BEHAVIOURS THAT CHALLENGE IN *SATB2*-ASSOCIATED SYNDROME

Volume II

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List of Appendices

Appendices

Appendix 1: Background Questionnaire

ID_____

BACKGROUND INFORMATION

Please tick or write your response to these questions concerning background details:
Please answer the following about the person you care for:

1. **Today's date:** _____
2. **Gender:** Male ☐ Female ☐
3. **Date of Birth**: ___/___/____ **Age:_____**
4. **Is the person you care for verbal?** (i.e. more than 30 signs/words in their vocabulary)

   Yes/No (delete as appropriate)

5. **Is the person you care for able to walk unaided?**

   Yes/No (delete as appropriate)

6. **Has the person you care for been diagnosed with a syndrome?** Yes/No (delete as appropriate)

If yes, please indicate which syndrome in 5a. and answer questions 6 to 8. If no, please move on to question 9

6.a

Cornelia de Lange syndrome	☐	Cri du Chat syndrome	☐
Prader-Willi syndrome	☐	Rubinstein Taybi syndrome	☐
Fragile X syndrome	☐	Down syndrome	☐
Lowe syndrome	☐	Soto Syndrome	☐
Rubinstein-Taybi syndrome	☐	9q34 deletion	
8p23deletion	☐	Tuberous Sclerosis	☐
Other _____			

7. **What is the genetic mechanism causing the syndrome in the person you care for?**

Uni-parental disomy	☐	Sequence repetition	☐
Deletion	☐	Translocation	☐
Unknown	☐	Other _____	

8. **When was the person you care for diagnosed?** _____
9. **Who diagnosed the person you care for?**

Paediatrician	☐	Clinical Geneticist	☐
GP	☐	Other _____	

10. **Has the person you care for had any medical/health difficulties in the last six months? If yes, please give details:**

_____

_____

In the information sheet and consent form we informed you that we may need to contact your child's/person you care for's GP in order to clarify any information regarding your child's health and diagnostic status (see consent form and information sheet for more information). If you have already indicated on the consent form that you are happy for us to do this, please complete the relevant details below:

11. Name of your child's/person you care for's GP_____

GP Address_____

GP Telephone number_____

The following questions ask for background information about you and your family. Please tick the appropriate boxes or write in the spaces provided.

Are you male or female? Male ☐ Female ☐

2. What was your age in years on your last birthday? _____years

3. Please tick the highest level of your educational qualifications.

No formal educational qualifications..... ☐
Fewer than 5 GCSE's or O Level's (grades A-C), NVQ 1, or BTEC First Diploma……. …. ☐
5 or more GCSE's or O Level's (grades A-C), NVQ 2, or equivalent……..…….. ☐
3 or more 'A' Levels, NVQ 3, BTEC National, or equivalent..... ☐
Polytechnic/University degree, NVQ 4, or equivalent..... ☐
Masters/Doctoral degree, NVQ 5, or equivalent……..... ☐

4. What is your relationship to your child with a genetic syndrome (e.g. mother, father, stepmother, grandmother, adoptive parent)? _____

5. In total how many people currently live in your home? _____ Adults _____ Children

6. Does your child with a genetic syndrome normally live with you?

Yes ☐ No ☐

If no, then where do they live?_____

7. What is your current marital status?

Married, and living with spouse..... ☐

Living with partner..... ☐

Divorced/Separated/Widowed/Single and NOT living with a partner..... ☐

If living with partner/spouse, please answer the following questions, if not, please go to question 12.

8. Is your partner male or female? Male ☐ Female ☐

9. What was their age in years on their last birthday? _____ years

10. Please tick the highest level of your partner/spouse's educational qualifications.

No formal educational qualifications..... ☐

Fewer than 5 GCSE or O Level (grades A-C), NVQ 1, or BTEC First Diploma…… ☐

5 or more GCSE or O Level (grades A-C), NVQ 2, or equivalent……..… ☐

3 or more 'A' Levels, NVQ 3, BTEC National, or equivalent..... ☐

Polytechnic/University degree, NVQ 4, or equivalent..... ☐

Masters/Doctoral degree, NVQ 5, or equivalent……..... ☐

11. What is your partner/spouse's relationship to your child with a genetic syndrome (e.g., mother, father, stepmother, adoptive parent)? _____

12. Recent data from research with families of children with special needs has shown that a family's financial resources are important in understanding family member's views and experiences. With this in mind, we would be very grateful if you could answer the additional question below. We are not interested in exactly what your family income is, but we would like to be able to look at whether those with high versus lower levels of financial resources have different experiences.

What is your current total annual family income? Please include a rough estimate of total salaries and other income (including benefits) before tax and national insurance/pensions.

Please tick one box only:

Less than £15,000……..……...… ☐

£15,001 to £25,000……...……. ☐

£25,001 to £35,000……...…..……. ☐

£35,001 to £45,000……...……. ☐

£45,001 to £55,000……...……..…. ☐

£55,001 to £65,000……..……..... ☐

£65,001 or more……..……..……. ☐

Please check your answers and go on to the next questionnaire.

Appendix 2: Wessex Questionnaire

WESSEX QUESTIONNAIRE

These items refer to the person you care for. For each question (A, B, C, D etc …), please enter the appropriate code in each box.

A) Wetting (nights)	**1** = frequently	**2** = occasionally	**3** = never	☐
B) Soiling (nights)	**1** = frequently	**2** = occasionally	**3** = never	☐
C) Wetting (days)	**1** = frequently	**2** = occasionally	**3** = never	☐
D) Soiling (days)	**1** = frequently	**2** = occasionally	**3** = never	☐
E) Walk with help*	**1** = not at all	**2** = not up stairs	**3** = up stairs and elsewhere	☐

*(note: if this person walks *by himself/herself* upstairs and elsewhere, please also code '3' for 'walk with help')

F) Walk by himself	**1** = not at all	**2** = not up stairs	**3** = up stairs and elsewhere	☐
G) Feed himself	**1** = not at all	**2** = with help	**3** = without help	☐
H) Wash himself	**1** = not at all	**2** = with help	**3** = without help	☐
I) Dress himself	**1** = not at all	**2** = with help	**3** = without help	☐

J) Vision	**1** = blind or almost	**2** = poor	**3** = normal	☐
K) Hearing	**1** = deaf or almost	**2** = poor	**3** = normal	☐
L) Speech	**1** = never a word	**2** = odd words only	**3** = sentences and normal	**4** = can talk but doesn't ☐

If this person talks in sentences, is his/her speech:

1 = Difficult to understand even by acquaintances, impossible for strangers?

2 = Easily understood for acquaintances, difficult for strangers?

3 = Clear enough to be understood by anyone? ☐

M) Reads	**1** = nothing	**2** = a little	**3** = newspapers and/or books	☐
N) Writes	**1** = nothing	**2** = a little	**3** = own correspondence	☐
O) Counts	**1** = nothing	**2** = a little	**3** = understands money values	☐

Appendix 3: Social Communication Questionnaire – Lifetime Version

Social Communication Questionnaire – Lifetime Version (SCQ-L) removed due to copyright restrictions

Appendix 4: Health Questionnaire

PART A

Instructions:

- Have these problems **EVER** affected your child or person you care for?
- Please rate as **0** – if the problem has never affected the person you care for, **1** – if it has been a mild problem, **2** - if the problem has been moderately serious, or **3** – if the problem has been severe.
- If the person you care for has had these problems please state whether any treatment has been implemented by circling **yes** or **no**.

	Never	Mild	Moderate	Severe
1a. Eye Problems (e.g. glaucoma / blocked tear duct/s).....	**0**	**1**	**2**	**3**
1b. Corrective surgery / medication / treatment: **yes / no**				
2a. Ear Problems (e.g. infections, glue ear) …….	**0**	**1**	**2**	**3**
2b. Corrective surgery / medication / treatment (e.g. grommets): **yes / no**				
3a. Dental Problems (e.g. toothache / gum problems / mouth ulcers / delayed eruption of teeth).....	**0**	**1**	**2**	**3**
3b.Dental surgery / treatment (e.g. teeth removal): **yes / no**				
4a. Cleft Palate.....	**0**	**1**	**2**	**3**
4b. Repaired: **yes / no**				
5a. Gastrointestinal Difficulties (e.g. reflux / stomach problems).....	**0**	**1**	**2**	**3**
5b. Corrective surgery / medication / treatment (e.g. nissen fundoplication): **yes / no**				
6a. Bowel Problems (e.g. obstruction).....	**0**	**1**	**2**	**3**
6b. Corrective surgery / treatment: **yes / no**				
7a. Heart Abnormalities or Circulatory Problems (e.g. congenital heart lesions or murmur).....	**0**	**1**	**2**	**3**
7b. Corrective surgery / medication / treatment: **yes / no**				
8a. Problems with Genitalia (e.g. prostate/ testicular problems i.e. undescended testes) ……..	**0**	**1**	**2**	**3**
8b. Corrective surgery / treatment: **yes / no**				
9a. Hernia (e.g. inguinal or hiatal).....	**0**	**1**	**2**	**3**
9b. Repair / treatment: **yes / no**				
10. Limb Abnormalities (e.g. malformed arm).....	**0**	**1**	**2**	**3**
11a. Epilepsy / Seizures / Neurological Referrals.....	**0**	**1**	**2**	**3**
11b. Medication: **yes / no**				
12a. Lung or Respiratory Problems (asthma/bronchitis).....	**0**	**1**	**2**	**3**
12b. Corrective surgery / medication / treatment: **yes / no**				
13a. Liver or Kidney Problems.....	**0**	**1**	**2**	**3**
13b. Corrective surgery / medication / treatment: **yes / no**				
14a. Diabetes or Thyroid Function Problems.....	**0**	**1**	**2**	**3**
14b. Corrective surgery / medication / treatment: **yes / no**				

15a. Skin Problems (e.g. tinea, eczema, psoriasis, dry skin)…….....	**0**	**1**	**2**	**3**
15b. Medication / treatment: **yes / no**				
16a. Other (please specify problem, severity from 0-3).....	**0**	**1**	**2**	**3**
16b. Corrective surgery / medication / treatment: **yes / no**				

PART B

Instructions:

- Have these medical problems affected the person you care for in the past **MONTH**
- Please rate as **0** – if your child has not been affected by this problem in the past month, **1** - if they have been mildly affected, **2** – if the problem has moderately affected your child and **3** - if your child has been severely affected by the problem.

	No	**Mild**	**Moderate**	**Severe**
17. Eye Problems (e.g. glaucoma / blocked tear duct/s).....	**0**	**1**	**2**	**3**
18. Ear Problems (e.g. infections, glue ear).....	**0**	**1**	**2**	**3**
19. Dental Problems (e.g. toothache / gum problems / mouth ulcers / delayed eruption of teeth).....	**0**	**1**	**2**	**3**
20. Cleft Palate.....	**0**	**1**	**2**	**3**
21. Gastrointestinal Difficulties (e.g. reflux / stomach problems).....	**0**	**1**	**2**	**3**
22. Bowel Problems (e.g. obstruction).....	**0**	**1**	**2**	**3**
23. Heart Abnormalities or Circulatory Problems (e.g. congenital heart lesions or murmur)…… ……...	**0**	**1**	**2**	**3**
24. Problems with Genitalia (e.g. prostate / testicular problems i.e. undescended testes)……..	**0**	**1**	**2**	**3**
25. Hernia (e.g. inguinal or hiatal).....	**0**	**1**	**2**	**3**
26. Limb Abnormalities (e.g. malformed arm).....	**0**	**1**	**2**	**3**
27. Epilepsy / Seizures / Neurological Referrals.....	**0**	**1**	**2**	**3**
28. Lung or Respiratory Problems (asthma / bronchitis).....	**0**	**1**	**2**	**3**
29. Liver or Kidney Problems.....	**0**	**1**	**2**	**3**
30. Diabetes or Thyroid Function Problems.....	**0**	**1**	**2**	**3**
31. Skin Problems (e.g. tinea, eczema, psoriasis, dry skin).....	**0**	**1**	**2**	**3**
32. Other (please specify problem and severity from 0-3) ……..	**0**	**1**	**2**	**3**

Appendix 5: The Gastro-oesophageal Distress Questionnaire

The GDQ

Instructions:

- This questionnaire asks about behaviours sometimes shown by people with learning disabilities.
- Please read the questions and examples carefully and indicate how often each behaviour has occurred in the **last two weeks** by circling the appropriate answer.

Does the person you care for:	**More than once an hour**	**Once an hour**	**Once a day**	**Once a week**	**Not occurred**
1. Arch his/her back, lie over arms of chairs or people on his/her back?	**4**	**3**	**2**	**1**	**0**
2. Lie over an object on his/her stomach? e.g. a side of an arm chair.	**4**	**3**	**2**	**1**	**0**
3. Salivate excessively?	**4**	**3**	**2**	**1**	**0**
4. Fidget, wriggle or move their body a great deal?	**4**	**3**	**2**	**1**	**0**
5. Place their hands or fingers in back of their mouth?	**4**	**3**	**2**	**1**	**0**
6. Chew on his/her clothes, fingers, hands or other parts of the body, objects or material?	**4**	**3**	**2**	**1**	**0**
7. Grind their teeth?	**4**	**3**	**2**	**1**	**0**
8. Scratch, hit, press or rub around the upper chest or throat?	**4**	**3**	**2**	**1**	**0**
9. Drink, request or seek out an excessive amount of fluids?	**4**	**3**	**2**	**1**	**0**
10. Cough, gag or regurgitate?	**4**	**3**	**2**	**1**	**0**
11. Appear in pain or discomfort (cry, groan or moan)?	**4**	**3**	**2**	**1**	**0**
12. Refuse food even though they are probably hungry?	**4**	**3**	**2**	**1**	**0**

13. Does the person you care for appear indecisive about food (edging towards table or food then moving away repeatedly, taking food and putting it back)? *(please tick)* **Yes** ☐ **No** ☐

14. Does the person you care for wake during the night?	**Never**	**Once a week**	**Most nights**	**Every night**
15. Does the person you care for sleep sitting or propped up?	**Never**	**Once a week**	**Most nights**	**Every night**
16. Does the person you care for seem to have bad breath?	**Never**	**Once a week**	**At the same time everyday**	**All day every day**

17. Has the person you care for prone to respiratory tract infections? *(please tick)* **Yes** ☐ **No** ☐

If 'yes' please indicate how often they occur:	**Monthly**	**Quarterly**	**Every six months**	**Annually**

Other (please specify)_____

Please check your answers and go on to the next questionnaire.

Appendix 6: The Repetitive Behaviour Questionnaire

THE RBQ

INSTRUCTIONS:

1. **The questionnaire asks about 19 different behaviours.**
2. **Each behaviour is accompanied by a brief definition and examples. The examples given for each behaviour are not necessarily a complete list but may help you to understand the definitions more fully.**
3. **Please read the definitions and examples carefully and circle the appropriate number on the scale to indicate how frequently the person you care for has engaged in each of the behaviours WITHIN THE LAST MONTH.**
4. **If a particular behaviour does not apply to the person you care for because they are not mobile or verbal please circle the number 0 on the scale.**

	Never	Once a month	Once a week	Once a day	More than once a day
1. Object stereotypy: Repetitive, seemingly purposeless movement of objects in an unusual way *E.g. twirling or twiddling objects, twisting or shaking objects, banging or slapping objects.*	**0**	**1**	**2**	**3**	**4**
2. Body stereotypy: Repetitive, seemingly purposeless movement of whole body or part of body (other than hands) in an unusual way. *E.g. body rocking, or swaying or spinning, bouncing, head shaking, body posturing.* Does not include self-injurious behaviour.	**0**	**1**	**2**	**3**	**4**
3. Hand stereotypy: Repetitive, seemingly purposeless movement of hands in an unusual way. *E.g. finger twiddling, hand flapping, wiggling or flicking fingers, hand posturing.* Does not include self-injurious behaviour.	**0**	**1**	**2**	**3**	**4**
4. Cleaning: Excessive cleaning, washing or polishing of objects or parts of the body *E.g. polishes windows and surfaces excessively, washes hands and face excessively.*	**0**	**1**	**2**	**3**	**4**
5. Tidying up: Tidying away any objects that have been left out. This may occur in situations when it is inappropriate to put the objects away. Objects may be put away into inappropriate places. *E.g. putting cutlery left out for dinner in the bin, removes all objects from surfaces.*	**0**	**1**	**2**	**3**	**4**
6. Hoarding: Collecting, storing or hiding objects to excess, including rubbish, bits of paper, and pieces of string or any other unusual items.	**0**	**1**	**2**	**3**	**4**
7. Organising objects: Organising objects into categories according to various characteristics such as colour, size, or function. *E.g. ordering magazines according to size, ordering toy cars according to colour, ordering books according to topic.*	**0**	**1**	**2**	**3**	**4**
8. Attachment to particular people: Continually asking to see, speak or contact a particular 'favourite' person. *E.g. continually asks to see or speak to particular friend, carer, babysitter or schoolteacher.*	**0**	**1**	**2**	**3**	**4**
9. Repetitive questions: Asking specific questions over and over. *E.g. always asking people what their favourite colour is, asking who is taking them to school the next day over and over.*	**0**	**1**	**2**	**3**	**4**

	Never	Once a month	Once a week	Once a day	More than once a day
10. Attachment to objects: Strong preference for a particular object to be present at all times. *E.g. carrying a particular piece of string everywhere, taking a particular red toy car everywhere, attachment to soft toy or particular blanket.*	**0**	**1**	**2**	**3**	**4**
11. Repetitive phrases/signing: Repeating particular sounds, phrases or signs that are unrelated to the situation over and over. *E.g. repeatedly signing the word 'telephone'.*	**0**	**1**	**2**	**3**	**4**
12. Rituals: carrying out a sequence of unusual or bizarre actions before, during or after a task. The sequence will always be carried out when performing this task and will always occur in the same way. *E.g. turning round three times before sitting down, turning lights on and off twice before leaving a room, tapping door frame twice when passing through it.*	**0**	**1**	**2**	**3**	**4**
13. Restricted conversation: Repeatedly talks about specific, unusual topics in great detail. *E.g. conversation restricted to: trains, buses, dinosaurs, particular film, country or sport.*	**0**	**1**	**2**	**3**	**4**
14. Echolalia: Repetition of speech that has either just been heard or has been heard more than a minute earlier. *E.g. Mum: ' Jack don't do that' Jack: 'Jack don't do that'.*	**0**	**1**	**2**	**3**	**4**
15. Preference for routine: Insists on having the same household, school or work schedule everyday*. E.g. likes to have the same activities on the same day at the same time each week, prefers to eat lunch at exactly the same time every day, wearing the same jumper everyday.*	**0**	**1**	**2**	**3**	**4**
16. Lining up or arranging objects*:* Arrangement of objects into lines or patterns. *E.g. placing toy cars in a symmetrical pattern, precisely lining up story books.*	**0**	**1**	**2**	**3**	**4**
17. Just right behaviour: Strong insistence that objects, furniture and toys always remain in the same place. *E.g. all chairs, pictures and toys have a very specific place that cannot be changed.*	**0**	**1**	**2**	**3**	**4**
18. Completing behaviour: Insists on having objects or activities 'complete' or 'whole'. *E.g. Must have doors open or closed not in between, story must be read from beginning to end, not left halfway through.*	**0**	**1**	**2**	**3**	**4**
19. Spotless behaviour: Removing small, almost unnoticeable pieces of lint, fluff, crumbs or dirt from surfaces, clothes and objects. *E.g. picking fluff off a jumper, removing crumbs from the kitchen table.*	**0**	**1**	**2**	**3**	**4**

Please check your answers and go on to the next questionnaire.

Appendix 7: The Activity Questionnaire

The Activity Questionnaire

Please read each item carefully and consider whether the behaviour has occurred in the last four weeks. Circle the appropriate number on the scale. Please ensure that you indicate a response for every item. If the particular behaviour does not apply, for example, if the person is not verbal, please circle 0 on the scale.

1) Does the person wriggle/squirm about when seated or lying down?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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2) Does the person fidget or play with their hands and/or feet when seated or lying down?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

---

3) Does the person find it difficult holding still?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

---

4) Does the person find it difficult to remain in their seat even when in situations where it would be expected?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

---

5) Does the person prefer to be moving around or becomes frustrated if left in one positions for too long?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

---

6) When the person is involved in a leisure activity (e.g. watching TV, playing a game etc.) do they make a lot of noise?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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7) When the person is involved in an activity, are they boisterous and/or rough?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

---

8) Does the person act as if they are “driven by a motor” (i.e. often very active)?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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9) Does the person seem like they need very little rest to recharge their battery?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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10) Does the person often talk excessively?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

---

11) Does the person’s behaviour seem difficult to manage/contain whilst out and about (e.g. in town, in supermarkets etc.)?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

---

12) Do you feel that you need to “keep an eye” on the person at all times?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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13) Does the person you care for seem to act/do things without stopping to think first?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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14) Does the person blurt out answers before questions have been completed?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

---

15) Does the person start to respond to instructions before they have been fully given or without seeming to understand them?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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16) Does the person want things immediately?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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17) Does the person find it difficult to wait?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

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18) Does the person disturb others because they have difficulty waiting for things or waiting their turn?

Never/almost never 0	Some of the time 1	Half of the time 2	A lot of the time 3	Always/almost all of the time 4

Appendix 8: Challenging Behaviour Questionnaire – Original Version

Challenging Behaviour Questionnaire – Original Version

1) Has the person shown self-injurious behaviour **in the last month**? (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing).

Yes ☐ No ☐

If the behaviour has not occurred, please go to question 2.
If the behaviour has occurred during the past month, please answer questions 1.1 to 1.4.:

1.1) Place a tick next to the items in the following list of behaviours, which the person displays in a repetitive manner (repeats the same movement/ behaviour twice or more in succession):

Hits self with body part (e.g. slaps head or face)……..… ☐
Hits self against surface or object (e.g. bangs head on floor or table)…….. ☐
Hits self with object…….. ☐
Bites self (e.g. bites hand on wrist or arm)……..…….. ☐
Pulls (e.g. pulls hair or skin)……..... ☐
Rubs or scratches self (e.g. rub marks on arm or leg)…….……. ☐
Inserts finger or objects (e.g. eye poking)……..... ☐
Other form of self-injury, please specify:……..…… ☐

1.2) In the last month, for how long did the **longest** episode or burst of this behaviour last? *(Please circle one number)*

1	2	3	4	5
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

1.3) In the last month, as a result of this behaviour, has physical contact, prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	1	2	3	4
Never	At least once a month	At least once a week	At least once a day	At least once an hour

1.4) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	2	3	4	5
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

2) Has the person shown aggression **in the last month**? (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing).

Yes ☐ No ☐

3) Has the person shown disruption and destruction of property or the environment **in the last month**? (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal).

Yes ☐ No ☐

4) Has the person shown stereotyped behaviours **in the last month**? (e.g. rocking twiddling objects, patting or tapping part of the body, constant hand movements, eye pressing).

Yes ☐ No ☐

Appendix 9: The Anxiety Depression and Mood Scale

THE ANXIETY DEPRESSION AND MOOD SCALE (ADAMS)

Instructions

The Anxiety Depression and Mood Scale (ADAMS) contains a list of behaviors that can be found among individuals with intellectual disability. Please describe the individual's behavior over the last 6 months.

0 behavior has not occurred, or is not a problem
1 behavior occurs occasionally, or is a mild problem
2 behavior occurs quite often, or is a moderate problem
3 behavior occurs a lot, or is a severe problem

	Not a problem	Mild problem	Moderate problem	Severe problem
1. Nervous	0	1	2	3
2. Problems initiating communication	0	1	2	3
3. Does not relax or settle down	0	1	2	3
4. Has period of over-activity	0	1	2	3
5. Sleeps more than normal	0	1	2	3
6. Withdraws from other people	0	1	2	3
7. Tense	0	1	2	3
8. Engages in ritualistic behaviours	0	1	2	3
9. Depressed mood	0	1	2	3
10. Sad	0	1	2	3
11. Worried	0	1	2	3
12. Has developing difficulty staying on task or completing work	0	1	2	3
13. Shy	0	1	2	3
14. Easily fatigued (not due to being overweight)	0	1	2	3
15. Anxious	0	1	2	3
16. Repeatedly checks items	0	1	2	3
17. Easily distracted	0	1	2	3
18. Lacks energy	0	1	2	3
19. Avoids others, spends much of time alone	0	1	2	3
20. Easily upset if ritualistic behaviours are interrupted	0	1	2	3
21. Lacks emotional facial expressions	0	1	2	3
22. Has shown difficulty in starting routine tasks	0	1	2	3
23. Listless	0	1	2	3
24. Experiences panic attacks	0	1	2	3
25. Avoids eye contact	0	1	2	3
26. Trembles when frightening situations are not present	0	1	2	3
27. Avoids peers	0	1	2	3
28. Tearful	0	1	2	3

Appendix 10: Mood, Interest and Pleasure Questionnaire

MOOD, INTEREST AND PLEASURE QUESTIONNAIRE

This questionnaire contains 12 questions – <u>you should complete all 12 questions</u>. Each question will ask for your opinion about particular behaviours, which you have observed in the <u>LAST 2 WEEKS</u>. For every question you should circle <u>the most appropriate</u> response e.g.

6) In the LAST TWO WEEKS, how interested did the person appear to be in his/her surroundings?

interested all of the time	***interested most of the time***	***interested about half of the time***	***interested some of the time***	***never interested***

1) In the last two weeks, did the person seem…

sad all of the time	sad most of the time	sad about half of the time	sad some of the time	never sad

Please comment if anything has happened in the last two weeks which you feel might explain sadness if it has been observed (e.g. a bereavement):

2) In the last two weeks, how often did you hear positive vocalizations* when the person was engaged in activities*?

all of the time	most of the the time	about half of the time	some of the time	never

*<u>positive vocalizations</u>: e.g. laughing, giggling, "excited sounds" etc.
*<u>engaged in activities</u>: i.e. when someone is actively involved in any activity such as a mealtime, a social interaction, a self-care task or social outing etc.

3) In the last two weeks, do you think the facial expression of the person looked "flat"*…

all of the time	most of the the time	about half of the time	some of the time	never

*<u>flat expression</u>: expression seems lifeless; lacks emotional expression; seems unresponsive.

4) In the last two weeks, would you say the person…

cried every day	cried nearly every day	cried 3-4 times each week	cried once or twice each week	cried less than once each week

5) In the last two weeks, how interested did the person appear to be in his/her surroundings?

interested all of the time	interested most of the time	interested about half of the time	interested some of the time	never interested

6) In the last two weeks, did the person seem to have been enjoying life…

all of the time	most of the the time	about half of the time	some of the time	never

Please comment if there are any reasons why this person might not have been enjoying him/herself e.g. illness, being in pain, experiencing a loss etc.:

7) In the last two weeks, would you say the person smiled…

at least once every day	at least once nearly every day	3-4 times each week	once or twice each week	less than once each week

8) In the last two weeks, how disinterested did the person seem to be in his/her surroundings?

disinterested all of the time	disinterested most of the time	disinterested about half of the time	disinterested some of the time	never disinterested

9) In the last two weeks, when the person was engaged in activities*, to what extent did his/her facial expressions* suggest that s/he was interested in the activity?

interested all of the time	interested most of the time	interested about half of the time	interested some of the time	never interested

*engaged in activities: i.e. when someone is actively involved in any activity such as a mealtime, social interaction, self-care task or social outing etc.
*facial expressions: interest might be indicated by the degree to which the person's gaze is being directed at the person/things involved in an activity.

10) In the last two weeks, would you say that the person…

laughed every day	laughed nearly every day	laughed 3-4 times each week	laughed once or twice each week	laughed less than once each week

11) In the last two weeks, how often did you see gestures which appeared to demonstrate enjoyment* when the person was engaged in activities*?

all of the time	most of the the time	about half of the time	some of the time	never

*gestures which appear to demonstrate enjoyment: e.g. clapping, waving hands in excitement etc.
*engaged in activities: i.e. when someone is actively involved in any activity such as a meal time, social interaction, self-care task or social outing etc.

12) In the last two weeks, did the person's vocalizations* sound distressed…

all of the time	most of the the time	about half of the time	some of the time	never

*vocalizations: any words, noises or utterances.

Please feel free to make any additional comments about the behaviour of the person over the last two weeks:

Appendix 11: Chapter Two comparisons of health, behavioural, autism and emotional characteristics

Appendices Table 1

Comparative group analyses across categories of behaviour that challenges for health characteristics, behavioural characteristics, autism spectrum disorder characteristics and emotional characteristics.

	Behavioural category											
	Self-injury		Comparative analyses		Aggression		Comparative analyses		Property Destruction		Comparative analyses	
	Yes (n=34)	No (n=47)	Statistic [1]	*p* value	Yes (n=61)	No (n=18)	Statistic [1]	*p* value	Yes (n=39)	No (n=40)	Statistic [1]	*p* value
Health characteristics												
Current health problem frequency	2.00 (1.00-4.00)	1.00 (0.00-2.00)	449.50	**.001**	2.00 (0.50-3.00)	1.00 (1.00-3.00)	526.50	.788	1.00 (0.00-3.00)	1.50 (1.00-3.00)	750.50	.768
Current health problem severity	3.50 (1.00-7.00)	1.00 (0.00-3.00)	437.50	**<.001**	2.00 (0.50-4.50)	1.00 (1.00-4.00)	527.00	.794	2.00 (0.00-5.00)	2.00 (1.00-4.00)	776.00	.968
GDQ clinical signs	7.00 (5.00-9.00)	4.00 (2.00-6.00)	339.00	**<.001**	5.62 (*3.21*)	4.17 (*2.18*)	-2.22	.032[c]	6.00 (4.00-9.00)	4.00 (2.25-6.00)	551.00	.024
Behavioural characteristics												
TAQ impulsivity	22.00 (18.00-23.00)	16.00 (11.00-21.00)	465.00	**.001**	21.00 (15.00-23.00)	13.50 (7.75-17.25)	259.50	**.001**	21.00 (15.00-23.00)	17.00 (10.63-21.00)	579.00	.048
TAQ overactivity	21.50 (15.00-25.00)	12.00 (5.00-18.00)	387.00	**<.001**	16.95 (*9.27*)	11.94 (*8.82*)	-2.09	.045	17.18 (*9.90*)	14.48 (*8.71*)	-1.29	.201
TAQ impulsive speech [a]	3.33 (*3.57*)	2.82 (*2.53*)	-.42	.710 [c]	3.00 (0.50-5.50)	2.00 (0.00-6.00)[3]	26.50	.658[h]	2.89 (*2.98*)	3.47 (*2.85*)	.47	.646[c]

Appendices Table 1 Continued

	Behavioural category											
	Self-injury		Comparative analyses		Aggression		Comparative analyses		Property Destruction		Comparative analyses	
	Yes (n=34)	No (n=47)	Statistic [1]	*p* value	Yes (n=61)	No (n=18)	Statistic [1]	*p* value	Yes (n=39)	No (n=40)	Statistic [1]	*p* value
ASD characteristics												
RBQ stereotyped behaviour	8.00 (3.00-11.25)	3.00 (0.00-7.00)	436.00	**<.001**	5.00 (0.00-9.00)	3.00 (0.00-7.25)	427.50	.150	8.00 (1.00-9.00)	3.00 (0.00-8.00)	574.000	.040
RBQ compulsive behaviour	6.00 (2.75-15.00)	5.00 (1.00-12.00)	709.50	.390	6.00 (3.50-15.00)	1.00 (0.00-6.25)	317.00	**.006**	8.00 (4.00-15.00)	3.50 (0.00-9.00)	511.000	**.008**
RBQ insistence on sameness	3.00 (0.75-6.00)	3.00 (0.00-5.00)	719.00	.437	4.00 (1.00-6.00)	1.00 (0.00-4.00)	344.00	.015	4.00 (1.00-6.00)	3.00 (0.00-5.00)	662.500	.243
RBQ restricted preferences [a]	6.22 (*4.02*)	5.88 (*3.98*)	-.21	.840 [c]	7.00 (4.00-9.00)	1.00 (0.00-10.00)	18.00	.236 [h]	7.33 (*3.39*)	5.73 (*4.10*)	-1.034	.314[c]
RBQ repetitive language [a]	8.00 (2.50-8.50)	5.00 (1.00-9.00)	65.00	.532	6.00 (1.00-8.50)	2.00 (2.00-11.00)	31.00	.965 [h]	6.00 (5.00-8.50)	4.00 (1.00-10.00)	57.5000	.548
SCQ communication [b]	7.00 (4.88-11.38)	4.94 (3.19-8.13)	452.00	.065	4.88 (3.25-8.13)	8.13 (5.00-12.19)	223.50	.030	7.02 (*3.91*)	5.83 (*3.85*)	-1.278	.206
SCQ restricted, repetitive, and stereotyped behaviours [b]	5.00 (3.50-7.00)	3.00 (1.00-5.00)	331.50	**.001**	4.00 (2.00-6.00)	3.00 (1.00-4.00)	279.00	.188	5.00 (3.00-6.00)	3.00 (1.50-5.00)	414.000	.029
SCQ reciprocal social interaction [b]	7.00 (4.00-10.50)	4.00 (3.00-7.00)	372.50	**.005** [d]	5.68 (*3.17*)	6.77 (*3.96*)	.41	.684[d, g]	7.00 (4.00-8.75)	4.00 (2.50-7.00)	408.000	.024

Appendices Table 1 Continued

	Behavioural category											
	Self-injury		Comparative analyses		Aggression		Comparative analyses		Property Destruction		Comparative analyses	
	Yes (n=34)	No (n=47)	Statistic [1]	*p* value	Yes (n=61)	No (n=18)	Statistic [1]	*p* value	Yes (n=39)	No (n=40)	Statistic [1]	*p* value
Emotional characteristics												
MIPQ-S mood	19.50 (17.75-21.00)	21.00 (18.00-22.00)	606.50	.063	20.00 (18.00-22.00)	21.00 (19.75-22.00)	421.00	.131	20.00 (18.00-21.00)	21.00 (18.00-22.00)	683.000	.337
MIPQ-S interest and pleasure	18.00 (15.00-20.00)	20.00 (17.00-21.00)	562.50	.023	18.00 (16.00-21.00)	19.50 (17.00-22.00)	431.50	.167	18.00 (14.00-20.00)	19.50 (18.00-21.00)	593.500	.066
ADAMS manic or hyperactive behaviour	11.00 (7.00-12.00)	6.00 (4.00-9.00)	438.50	**.001**	9.00 (6.00-12.00)	4.00 (1.00-7.25)	220.500	**<.001**	8.85 (*3.64*)	6.80 (*3.94*)	-2.40	.019
ADAMS depressed mood	4.00 (1.00-7.25)	2.00 (0.00-4.00)	600.50	.054	2.00 (1.00-5.00)	1.00 (0.00-3.00)	374.500	.038	3.00 (1.00-7.00)	2.00 (0.00-4.00)	623.00	.118
ADAMS social avoidance	6.00 (3.75-10.00)	5.00 (2.00-7.00)	586.50	.041	6.70 (*4.18*)	4.39 (*3.07*)	-2.280	.025[f, g]	6.00 (4.00-10.00)	4.00 (2.00-7.00)	554.50	.026
ADAMS general anxiety	8.00 (4.75-10.00)	3.00 (1.00-6.00)	418.50	**<.001**	6.00 (3.00-9.00)	1.50 (0.00-3.25)	237.500	**<.001**	8.00 (4.00-9.00)	2.50 (1.00-5.75)	384.00	**<.001**
ADAMS compulsive behaviour	5.00 (3.00-7.00)	2.00 (0.00-5.00)	451.50	**.001**	5.00 (1.50-6.00)	2.00 (0.00-3.25)	354.500	.022	5.00 (2.00-6.00)	2.00 (0.00-5.75)	547.50	.021

Notes. Significant group differences highlighted in **bold**.

[a] subscales only calculated for verbal participants (self-injury presence: n=9, self-injury absence: n=17; aggression presence: n=21, aggression absent: n=3; property destruction present: n=9, property destruction absent: n=15).

[b] SCQ only valid for individuals aged 4 years and over; 10 participants excluded from SCQ analyses (self-injury group: n=29, no self-injury group: n=42; physical aggression group: n=56, no physical aggression: n=13; property destruction group: n=36, no property destruction group: n=33).

[c] unequal variance (Welch's) *t*-test reported to account for unequal group sample sizes.

[d] *t*-test with logarithmic transformation.

[e] *t*-test with reflect and square root transformation.

[f] *t*-test with square root transformation.

[g] Descriptive values derived from untransformed raw data for greater representativeness.

[h] IQR values not wholly representative as *n*=3.

[1] Median (IQR) reported where Mann Whitney *U* tests were conducted, mean (*SD*) reported where *t*-tests were conducted.

Appendix 12: Chapter Three preliminary search strategy

Appendices Table 2

Preliminary search strategy: search terms and MeSH headings for databases searched.

		Web of Science	**PsycINFO**
Intellectual disability and genetic syndromes	Search terms	(TS=(intellectual* disab* OR learning disab* OR developmental* disab* OR mental* retard* OR mental* handicap*)) *OR* (TS=("smith-magenis" OR "prader-willi" OR "angelman" OR "fragile X" OR "cri-du-chat" OR "cornelia-de-lange" OR "down* syndrome" OR "lowe syndrome" OR "williams syndrome"))	(((intellectual* disab* OR learning disab* OR developmental* disab* OR mental* retard* OR mental* handicap*).ab,ti,tw) *OR* ("smith-magenis".mp.) *OR* ("prader-willi".mp.) *OR* ("angelman".mp.) *OR* ("fragile x".mp.) *OR* ("cri-du-chat".mp) *OR* ("cornelia de lange".mp.) *OR* ("down* syndrome".mp.) *OR* ("williams syndrome".mp.) *OR* ("lowe syndrome".mp.))
	MeSH headings	-	exp Intellectual Development Disorder/ *OR* exp Developmental Disabilities/ *OR* exp Prader-Willi Syndrome/ *OR* exp Fragile X Syndrome/ *OR* exp Crying Cat Syndrome/ *OR* exp Cornelia de Lange Syndrome/ *OR* exp Down's Syndrome/ *OR* exp Williams Syndrome/
Behaviours that challenge	Search terms	TS=(("challeng*" OR "problem*" OR "maladaptive" OR "aggress*" OR "destruct*" OR "disrupt*" OR "difficult*" OR "concern*") NEAR/2 ("behavio$r*"))	(("challeng*" OR "problem*" OR "maladaptive" OR "aberrant" OR "aggress*" OR "destruct*" OR "disrupt*" OR "difficult*" OR "concern*") adj2 "behavio$r*").ti,ab,tw.)
	MeSH headings	-	-
Measurement tools	Search terms	(TS=("tool*" OR "checklist" OR "questionnaire*" OR "interview*" OR "measure*" OR "inventor*" OR "survey*" OR "score")) *OR* (TS=(("parent*" OR "carer" OR "caregiver*" OR "informant" OR "teacher*" OR "self") NEAR/1 ("report*" OR "rated")))	(("tool*" OR "checklist" OR "questionnaire*" OR "interview*" OR "measure*" OR "interview*" OR "inventor*" OR "survey*" OR "score").ti,ab,tw.) *OR* ((("parent*" OR "carer" OR "caregiver*" OR "informant" OR "teacher*" OR "self") adj2 ("report*" OR "rated")).ti,ab,tw.)
	MeSH headings	-	-
Inclusion dates		Time span 2001-date of search (20.10.2021)	Time span 2001-date of search (20.10.2021)

Appendices Table 3

Preliminary search inclusion and exclusion criteria for screening of returned papers and identified measurement tools.

Screening of returned papers	
Inclusion criteria	**Exclusion criteria**
Studies published as full text original articles.	Non-human studies.
Studies published in English or that have an English language translation available.	Conference abstracts/papers, book chapters, letters, patents, editorial material, reviews, published protocols.
Studies including a standardised, quantitative measure in English Language (e.g., questionnaire, inventory, quantitative interview) described to assess BtC or problem behaviour.	Qualitative studies
Studies including participants reported to have an ID (or IQ < 70), diagnosis of a genetic syndrome associated with ID, or diagnosis of autism and comorbid ID (excluding high-functioning autism).	Studies published in non-peer reviewed journals.
Screening of measurement tools	
Inclusion criteria	**Exclusion criteria**
Measure could be obtained; the measure is freely available online or could be obtained by contacting the authors.	<50% of items assess BtC relevant to the definition of BtC in the current review; *and/or* measure does not quantitatively assess functions of BtC; *and/or* measure is a descriptive clinical interview without scoring criteria (purposed to guide clinical judgement).
Items in the measure include ratings of harm, frequency, severity, intensity, or difficulty managing behaviour to provide indication of the impact of BtC.	Previous version of a measure in which the latest version was published over 10 years ago at the time of search (e.g., if the latest version of a measure was published in 2008, previous versions of the measure are ineligible for inclusion). *However*, if the latest version of a measure was published *less than* 10 years ago at the time of search (e.g., in 2020), the previous version of the measure is eligible for inclusion.

Appendices Table 4

Measurement tools meeting criteria for inclusion in the review

Measurement tool	Brief description	BtC measurement tool	Function measurement tool
Aberrant Behavior Checklist – Irritability subscale (ABC-I; Aman & Singh, 1986; 1994; 2017)	The ABC is a 58-item measure containing five subscales. The 15-item *irritability subscale* met inclusion criteria. Items are rated from 0 ('not at all a problem') to 3 ('the problem is severe in degree).	+	-
Adult Behaviour Checklist (ABCL; Achenbach & Rescorla, 2003)	The ABCL is a 126-item measure comprising three high-order factor scales (internalising problems, externalising problems, and total problems) and eight syndrome subscales. The measure is suitable for adults aged 18-59 years. The 16-item *aggressive behaviour* syndrome subscale met criteria for inclusion. Items are rated from 0 ('Not True') to ('Very True or Often True').	+	-
Achenbach Youth Self-Report – 11-18 – Aggressive behaviour subscale (YSR 11-18-AB; Achenbach & Rescorla, 2001)	The YSR 11-18 is a 112-item measure comprising three high-order factor scales (internalising problems, externalising problems, and total problems) and eight syndrome subscales. The 17-item *aggressive behaviour* syndrome subscale met criteria for inclusion. Items are rated from 0 ('Not True') to ('Very True or Often True').	+	-
Adult Scale of Hostility and Aggression Reactive/Proactive (A-SHARP; Matlock & Aman, 2011)	The A-SHARP is a 48-item measure comprising five subscales. The 9-item *verbal aggression* subscale and 13-item *physical aggression* subscale met criteria for inclusion. Two dimensions of behaviour are evaluated: frequency/severity (problem scale) of behaviour in the past month, rated on a 4-point Likert scale from 0 'never happened' to 3 'severe or frequent', and provocation scale rated on a 5-point scale from -2 'only when provoked' to +2 'always the first to act'.	+	-
Aggressive Behaviour Scale (ABS; Perlman & Hirdes, 2008)	The ABS is a 4-item scale for aggressive behaviour based on the Minimum Data Set 2.0 (MDS 2.0). All 4-items met inclusion criteria as topographies of BtC. Each item is rated for its frequency on a 4-point Likert scale (0-3). The frequency scores for each item are summed to form a total score (0-12). The total score is divided into four levels from 'none' to 'very severe', indicating the overall frequency/severity of behaviour.	+	-
Aggression and Self-Injurious Behaviour Questionnaire (ASIQ; Bello-Mojeed et al., 2016)	The ASIQ was modified from the Challenging Behaviour Questionnaire (Hyman et al., 2002) and Behavior Problems Inventory (Rojahn et al., 2001). The ASIQ contains two subscales: a 10-item *self-injurious behaviour* subscale and 12-item *aggressive behaviour* against a person or property subscale. Each item represents a behaviour topography and is rating on four scales: frequency, severity, duration and need for physical restraint. Scores are summed on each subscale to form a total score, with higher scores indicating more difficult or challenging behaviour.	+	-

Appendices Table 4 Continued

Measurement tool	Brief description	BtC measurement tool	Function measurement tool
Autism Spectrum Disorders – Behaviour Problems for Adults (ASD-BPA; Matson & Rivet, 2007)	The ASD-BPA is a 20-item measure comprising four subscales. The -item A*ggression/Destruction* subscale and 3-item *Self-injurious Behavior* subscale met criteria for inclusion. Items are rated on a binary scale from 0 (not a problem, no impairment) to 1 (problem, impairment).	+	-
Autism Spectrum Disorders – Behavior Problems for Children (ASD-BPC; Matson et al., 2008)	The ASD-BPC (also reported as the Autism-Spectrum Disorders – Problem Behaviors for Children; ASD-PBC) is an 18-item measure comprising two subscales: externalizing and internalizing. The 8-item *externalizing* subscale met criteria for inclusion. Items are rated for severity on a 3-point Likert scale from 0 (not different; no impairment) to 2 (very different; severe impairment).	+	-
Behavior Problems Inventory-01 (BPI-01; Rojahn et al., 2001)	The BPI-01 is a 49-item measure containing three subscales. The *self-injurious behavior* and *aggressive/destructive behavior* subscales met inclusion criteria. Items are rated separately for occurrence, severity (1 'slight' to 3 'severe') and frequency (1 'monthly' to 4 'hourly').	+	-
Behavior Problems Inventory – Short form (BPI-S; Rojahn et al., 2012)	A shortened 30-item version of the BPI-01. The BPI-S contains three subscales. The *self-injurious behavior* and *aggressive/destructive behavior* subscales met inclusion criteria. Items are rated separately for occurrence, severity (1 'slight' to 3 'severe') and frequency (1 'monthly' to 4 'hourly').	+	-
Baby and Infant Screen for Children with aUtism Traits (BISCUIT – Part 3; Matson et al., 2007)	The BISCUIT-Part 3 is a 17-item measure designed to assess problem behaviors in infants and toddlers with ASD. The 10-item *Aggressive/Disruptive behavior* subscale and 2-item *Self-injurious behavior* subscale met criteria for inclusion. Items are rated on a 3-point Likert scale from 0 (not a problem or impairment, not at all) to 2 (severe problem or impairment).	+	-
Challenging Behaviour Attributions Scale (CHABA; Hastings, 1997)	The CHABA is a 39-item scale comprising seven subscales relating to causal models of BtC. The CHABA was developed to assess staff attributions of BtC circumstances. However, several studies have modified the scale so that parents or caregivers are asked to rate specific incidents of BtC in their child or person they care for (e.g., Grey et al., 2002; Rose, 2002). Subscales are *learned positive, learned negative, emotional, biomedical, physical, environment*, and *stimulation*. Items are rated on a 5-point Likert scale from 'very unlikely' to 'very likely'.	-	+
Challenging Behaviour Interview (CBI; Oliver et al., 2003)	The CBI defines five topographies of behaviours which are rated in line with the Emerson (2001) definition of BtC. Behaviour topographies are each rated on frequency, duration, physical harm, social disturbance and emotional upset, effects on environment, and management responses.	+	-
Challenging Behaviour Questionnaire (CBQ; Hyman et al., 2002)	The CBQ is a 7-item informant questionnaire designed to identify the phenomenology of BtC in individuals with ID and is based on the CBI. The measure assesses the presence (frequency) of behaviour topographies over the past month. Topographies include *self-injury*, *aggression,* and *property destruction*. Additional ratings are available for the severity of self-injurious behaviour and the presence or absence of eight different self-injurious behaviour topographies. The severity rating includes ratings for frequency of intervention and duration of behaviour.	+	-

Appendices Table 4 Continued

Measurement tool	Brief description	BtC measurement tool	Function measurement tool
Challenging Behaviour Questionnaire – Adapted (CBQ-A; Waite et al., 2017)	The CBQ-A is a modified version of the CBQ. In the original measure, Likert scales for the severity of behaviour are administered for the self-injury item only. These scales are replacing for physical aggression and destruction of property in the modified version. The modified version also includes ratings for the presence or absence of behaviour topographies for each category of BtC. Behaviour topographies are rated in relation to the previous 8 weeks. The frequency and severity of behaviour topographies are rated.	+	-
Challenging Behaviour Scale (CBS; Moniz-Cook et al., 2001)	The CBS is a 25-item measure originally designed to measure BtC among elders with dementia in nursing homes and has been used in several research studies with ID samples. The CBS met criteria for inclusion with >50% of items being identified as a BtC topography. Items are rated for occurrence over the previous 8 weeks, frequency (on a 4-point Likert scale from occasionally present' to 'daily') and severity (on a 4-point scale from 'easily managed' to 'very difficult to manage'),	+	-
Checklist of Challenging Behaviour (CCB; Harris et al., 1994)	A 32-item measure with two subscales; a 14-item subscale of *physical aggression against others*, and an 18-item subscale of *'other' challenging behaviour;* both subscales met inclusion criteria. Items are rated separately for frequency and management difficulty. The aggression subscale includes additional ratings for severity of injuries (1 'no injury' to 5 'very serious injury').	+	-
Child Behaviour Checklist – 1.5-5 – Aggressive behaviour subscale (CBCL 1.5-5-Aggressive behaviour subscale; Achenbach & Rescorla, 2001)	The CBCL 1.5-5 is a 99-item measure comprising three high-order factor scales (internalising problems, externalising problems, and total problems) and seven syndrome subscales. The 19-item *aggressive behaviour* syndrome subscale met inclusion. Items are rated from 0 ('Not True') to ('Very True or Often True').	+	-
Child Behaviour Checklist – 6-18 – Aggressive behaviour subscale (CBCL 6-18-AB; Achenbach & Rescorla, 2001)	The CBCL 6-18 is a 112-item measure comprising three high-order factor scales (internalising problems, externalising problems, and total problems) and eight syndrome subscales. The 18-item *aggressive behaviour* syndrome subscale met inclusion criteria. Items are rated from 0 ('Not True') to ('Very True or Often True').	+	-
Child Behaviour Checklist – Teacher Report Form – Aggressive behaviour subscale (CBCL-TRF-AB; Achenbach & Rescorla, 2000)	The CBCL-TRF is a 112-item measure comprising three high-order factor scales (internalising problems, externalising problems, and total problems) and eight syndrome subscales. The 20-item *aggressive behaviour* syndrome subscale met inclusion criteria. Items are rated from 0 ('Not True') to ('Very True or Often True').	+	-
Children Scale of Hostility and Aggression – Reactive/Proactive (C-SHARP; Farmer & Aman, 2009)	The C-SHARP is a 58-item measure comprising five subscales. The 12-item *verbal aggression* subscale, 12-item *bullying* subscale, and 8-item *physical aggression* subscale met inclusion criteria. Two dimensions of behaviour are evaluated: frequency/severity (problem scale) of behaviour in the past month, rated on a 4-point Likert scale from 0 'never happened' to 3 'severe or frequent', and provocation scale rated on a 5-point scale from -2 'only when provoked' to +2 'always the first to act'.	+	-

Appendices Table 4 Continued

Measurement tool	Brief description	BtC measurement tool	Function measurement tool
Contextual Assessment Inventory (CAI; MaAtee et al., 2004; Carr et al., 2008)	The CAI is a 24-item measure that assesses the contexts associated with the presence of problem behaviour in an individual. Respondents rate the presence of different BtC topographies, and the frequency of each topography on a 5-point scale from 'rarely' to 'often'. In addition, the CAI comprises four broad scale and nine subscales of contextual variables. Broad scales (and subscales) are social/cultural (negative interactions; disappointments), tasks/activity (factors related to tasks; factors related to daily routines), physical (uncomfortable environment; changes in the environment), and biological (medication; illness; physiological stress). Contextual items are rated on a 5-point Likert scale from 0 'not likely' to 5 'very likely'.	+	+
Comprehensive Assessment of Triggers for Behaviours of Concern Scale (CATS; Limbu et al., 2021)*	The CATS is a 333-item measure that assesses triggers and antecedents for behaviours of concern. Respondents rate the presence of triggers as present or absent. Triggers are categorised under five contextual categories *external environment, internal environment, expression of volition, characteristics associated with intellectual disability or autism,* and *specific activities/events*. Each of the five contextual categories are subdivided into subcategories, with the CATS comprising 12 subcategories in total, e.g., the *internal environment* categories consists of subcategories for *aversive physical states, medical conditions, mental health problems,* and *emotional states.*	-	+
Developmental Disabilities Profile – Maladaptive Behavior Scale (DDP-MBS; Brown et al., 1986)	The *DDP-MBS* is a 13-item scale. The scale met criteria for inclusion with >50% of items being identified as a BtC topography. Behaviour topographies are rated for their frequency on a 6-point scale from 1 'not this year' to 5 'once a day or more'.	+	-
Diagnostic Assessment for Severely Handicapped – II (DASH-II; Matson, 1995)	The DASH-II is an 84-item measure designed to assess psychopathology in individuals with severe and profound ID. The measure includes thirteen subscales. The five-item *self-injurious behaviour* subscale and 17-item *impulse control and miscellaneous behaviour* problems subscale met criteria for inclusion. Items are rated for their frequency on a 3-point Likert scale from 0 'not at all' to 2 'more than 10 times', duration on a 3-point Likert scale from 0 'less than a month' to 2 'over 12 months', and severity on a 3-point Likert scale from 0 'no disruption or damage' to 2 'property damage or injury'.	+	-
Early Childhood Behaviour Screen (ECBS; Holtz & Fox, 2012)	The ECBS is a 20-item measure with two subscales: *challenging behaviours* and prosocial behaviours. The 10-item *challenging behaviour* subscale met inclusion criteria. Behaviour topographies are rated for frequency on a 3-point Likert scale from 1 'almost never' to 3 'often'.	+	-
Extended Modified Overt Aggression Scale (Extended-MOAS; Crocker et al., 2006)	The Extended-MOAS is a modified version of the MOAS (Kay et al., 1988). The scale was modified with an additional behaviour topography: *sexual aggressive behaviour*.	+	-
Functional Analysis Checklist (FAC; Sturmey, 2001)	The FAC is a 41-item questionnaire used to assess whether a specified behaviour is associated with *biological factors, physical environment, communication, escape/demand factors, elicited or adjunctive behaviour, activity transitions*, and/or *positive reinforcement*. Respondents respond 'yes' or 'no' to each question. Further details are provided for each item answered with 'yes'.	-	+

Appendices Table 4 Continued

Measurement tool	Brief description	BtC measurement tool	Function measurement tool
Functional Assessment for Multiple Causality (FACT; Matson et al., 2003)	The FACT is a 35-item measure designed to identify the most prominent function associated with the occurrence of problem behaviors. The FACT comprises five subscales, each containing 7-items: Tangible, Physical, Attention. Escape, Non-social. The FACT has a novel forced-choice response format. Respondents must choose between three response options for each item. Function subscale scores are formed by tallying the frequency of function choices endorsed.	-	+
Functional Assessment Screening Tool (FAST; Iwata et al., 2013)	The FAST is a 16-item measure of the antecedent and consequent events that might be associated with the occurrence of behaviour. The FAST comprises four subscales: *social* (*attention/preferred items*), *social* (*escape from task/activities*), *automatic* (*sensory stimulation*) and *automatic* (*pain attenuation*).	+	+
Great Outcomes for Kids Impacted by Severe Developmental Disabilities – Brief Maladaptive Scale (GO4KIDDS-MBS; Esteves et al., 2021)	The GO4KIDDS was designed for use in children and adolescents with severe developmental disability. The measure comprises two scales for adaptive and maladaptive behaviour. The *brief maladaptive behaviour scale* met criteria for inclusion with >50% of items being identified as a topography of BtC, e.g., self-injurious behaviour and aggressive/destructive behaviour. Each topography is rated for its frequency over the previous 2 months.	+	-
Learning Disability Needs Assessment Tool (LDNAT; Painter et al., 2016)	The LDNAT is a holistic needs assessment tool developed from the Health of the Nation Outcome Scales (HoNOS; Wing et al., 1993) and the Mental Health Clustering Tool (MHCT; Self et al., 2008). The LDNAT consists of twenty-three components. The 8-item *challenging behaviour* component met criteria for inclusion. The challenging behaviour component contains 8 scales, each with anchor point descriptors adhering to a common underlying set of response options from 0 'no problem' to 4 'severe problem'.	+	-
Modified Overt Aggression Scale (MOAS; Kay et al., 1988)	The MOAS is a modified version of the OAS. The scale was modified by adding zero points to indicate non-occurrence of each topography (verbal aggression, physical aggression against objects, physical aggression against self, and physical aggression against other people). Ratings of intervention method severity were removed, and a system of weights was introduced to reflect the relative severities of the topographies.	+	-
Motivation Assessment Scale (MAS; Durrand & Crimmins, 1992)	The MAS is 16-item 7-point rating scale to assess four functions of BtC: *attention, escape, tangible*, and *sensory*. Parents or caregivers report on how likely their child's behaviour is to occur in various situations on a 7-point Likert scale from 0 ('never') to 6 ('always').	-	+
Overt Aggression Scale (OAS; Yudofsky et al., 1986)	The OAS includes four aggression topographies, each meeting criteria for inclusion as a topography of BtC. Topographies are verbal aggression, physical aggression against objects, physical aggression against self, and physical aggression against other people. Each behavioural topography is rated on severity and intervention method severity. The scale is completed for each episode of aggression.	+	-

Appendices Table 4 Continued

Measurement tool	Brief description	BtC measurement tool	Function measurement tool
Overt Aggression Scale – Modified for Neurorehabilitation (OAS-MNR; Alderman et al., 1997)	The OAS-MNR is a modified version of the OAS. The scale was modified by adding ratings for antecedents (set one: contributing factors and set two: contributing factors observed directly before behaviour), methods of intervention, and consequences of behaviour.	+	+
Individual Schedule of the Challenging Behaviour Survey (Alborz et al., 1994)	This measure consists of two parts. Part 1 collects information of the characteristics of the individual. Part 2 includes questions on topographies of BtC. Part 2 met criteria for inclusion with >50% of behavioural topographies being identified as BtC. Each behavioural topography is rated for occurrence and frequency. Additional questions include triggers for behaviour, circumstances for behaviour, explanations for behaviour (functions and setting events), and methods of intervention.	+	+
Initial Behavioural Assessment and Protective Equipment Decision Key (IBA-PEDK; Daraiseh et al., 2018)	The IBA is a risk assessment tool to identify recent history of BtC. The PEDK aggregates behaviours from the IBA to reveal the appropriate PPE to be worn by direct-care staff. The IBA-PEDK met inclusion criteria with >50% of behaviours being identified as a BtC topography. Behaviours are rated for their frequency (from 'low' to 'high') and intensity based on the degree of harm to themselves and others.	+	-
Institute for Basic Research – Modified Overt Aggression Scale (IBR-MOAS; Cohen et al., 2010)	The IBR-MOAS modified the OAS with the addition of a fifth behaviour topography; verbal aggression against self. Each behavioural topography is rated for frequency of occurrence for each item over the past year.	+	-
Nisonger Child Behaviour Rating Form (NCBRF; Aman et al., 1996)	The NCBRF is a 76-item parent report measure that comprises six 'problem behaviour' subscales and two 'social competence' subscales. The 7-item *self-injury/stereotypic* subscale met criteria for inclusion with >50% of items being identified as a topography of BtC. Behaviour topographies are rated for the past month for their frequency/extent problematic (from 0 'behaviour did not occur or was not a problem' to 3 'behaviour occurred a lot and was a severe problem').	+	-
Pervasive Developmental Disorder Behavior Inventory – Aggression subscale (PDDBI-A; Cohen et al., 2003)	The PDDBI is a parent and caregiver report measure comprising two scales: adaptive and maladaptive behavior. The maladaptive behaviour scale contains eight subscales. The 20-item *aggression* subscale met criteria for inclusion. Items are rated on a 4-point Likert scale from 0 ('Does not show behavior') to 3 ('Usually/typically shows behavior').	+	-
Problem Behaviour Checklist (PBCL; Tyrer et al., 2016)	The PBCL is a 7-item scale comprising seven behavioural topographies. The PBCL was developed to provide a short and comprehensive scale to assess BtC more quickly than commonly used scales. The scale met criteria for inclusion with >50% of items being identified as a BtC topography. Behaviour topographies are rated using anchor point descriptors adhering to a common underlying set of response options from 0 ('behaviour absent') to 4 ('extreme behaviour leasing to threat of loss of life or permanent injury and damage').	+	-

Appendices Table 4 Continued

Measurement tool	Brief description	BtC measurement tool	Function measurement tool
Psychopathology Checklists for Adults with Intellectual Disabilities (P-AID; Hove & Havik, 2008)	The P-AID checklists comprise ten psychiatric scales and eight problem behaviour scales. The problem behaviours scales met criteria for inclusion with >50% of behavioural topographies being identified as BtC. Each behaviour topography is rated for occurrence, with further binary yes/no questions used to indicate the severity, impact, risk of harm or injury, persistence, presence across situations and frequency of behaviour. Responses are summed to form a subscale score for each topography.	+	-
Questions About Behavioural Function (QABF; Matson & Vollmer, 1995)	The QABF is a 25-item measure of functions that might maintain problem behaviours in individuals with ID. The QABF comprises five subscales: attention, escape, tangible, non-social and physical. Items are rated on a 4-point Likert scale from 0 'never' to 3 'often'.	-	+
Sanfilippo Behaviour Rating Scale (SBRS; Shapiro et al., 2015)	The SBRS comprises three sections: Section I communication, Section II tantrums and Section III behaviour. Section II is composed of the frequency, duration and emotions expressed during tantrums and met criteria for inclusion. Section III contains fourteen behaviour clusters. The 7-item mood, anger and aggression cluster met criteria for inclusion. Items in the cluster are rated for frequency on a 7-point Likert scale from 0 ('never') to 6 ('always').	+	-
Scales of Independent Behaviour – Revised – Problem Behaviour Subscale (SIB-R-PB; Bruininks et al., 1996)	The SIB-R contains two subscales: adaptive behaviour and problem behaviour. The problem behaviour scale contains eight items, including *self-injury, aggression*, and *destructive behaviour*, which meet the inclusion criteria requiring 50% of items to be a topography of BtC. Each behaviour is rated for its frequency and severity.	+	-
Self-injury, Aggression and Destruction Screening Questionnaire (SAD-SQ; Davies & Oliver, 2006; Adams et al., 2018)	The SAD-SQ is a 30-item screening questionnaire to assess risk for and presence of self-injurious, aggressive, or destructive behaviour. The SAD-SQ is based on the CBQ (Hyman et al., 2002). In addition to ratings of putative risk markers for BtC, behavioural topographies are rated for their presence, severity, frequency, and level of concern. Items are summed to achieve a total severity score for each form of challenging behaviour.	+	-
Self-Injury Trauma Scale (SITS; Iwata et al., 1990)	The SITS is used to quantify visible injuries caused by self-injurious behaviour. Self-injury behaviour topographies are rated for their presence, the location of self-injury and severity of self-injury.	+	-
Staff Observation Aggression Scale – Revised (SOAS-R; Nijman et al., 1999)	The SOAS-R is used for the evaluation and monitoring of the nature, frequency, and severity of aggressive behaviour. It comprises ratings for five dimensions of aggressive behaviour: cause or provoking factors aggression, method of aggression, target of aggression, consequences, and intervention method to stop aggression.	+	-
Structured Interview for Skin Picking in Prader-Willi syndrome (Bull et al., 2021)	A semi-structured interview including fixed choice questions to assess the frequency and duration of skin picking, based on the CBI (Oliver et al., 2003).	+	+

Appendices Table 4 Continued

Measurement tool	Brief description	BtC measurement tool	Function measurement tool
Temper Outburst Interview (TOI; Tunnicliffe et al., 2014)	A semi-structured interview intended to elicit a phenomenological account of temper outbursts from a caregiver perspective. The TOI covers the latency and duration of outbursts, common antecedents, precursor behaviours, type, and sequence of behaviours during a typical outburst, and the success or otherwise of management strategies used by caregivers to alleviate harm or reduce outbursts. A proportion of questions were taken directly from the CBI (Oliver et al., 2003). Coding instructions are available for each question enabling quantitative analysis.	+	+

Note. *At the time of the main search, the CATS was identified as newly published measure of function that was published after the preliminary search took place. Terms for the CATS were not included in the main search strategy. However, forwards and backwards searches revealed no published evidence of IC, IRR and TRTR for this measure.

Appendices Table 5

Measurement tools excluded and reasons for their exclusion.

Measurement tool	Could not be obtained for assessment of eligibility	Not available	Not developed for broader use	Not available in English language	<50% of items in scale or subscale are BtC	Measure does not quantitatively assess functions of BtC	Descriptive clinical interview without scoring criteria	No ratings indicative of impact or harm
AAMR Adaptive and Maladaptive Behaviour Scale (AAMR-ABS; Nihira et al., 1993)		✓[a]						
Aberrant Behavior Checklist-Fragile X Syndrome (ABC-FXS; Sansone et al., 2011)					✓	✓		
About My Child – 26 (AMY-26; Rosenbaum et al., 2008)					✓	✓		
Assessment of Concerning Behaviour (ACB; Tarver et al., 2021)					✓	✓		
The Behaviour Assessment System for Children (BASC; Reynolds & Kamphaus, 2004)	✓							
Behaviour Assessment Guide Problem Behaviour Inventory (BAS-PBI; Willis et al., 1989)					✓	✓	✓	
Behaviour Problems Index (Peterson & Zill, 1986)					✓	✓		
Brief Infant Toddler Social Emotional Assessment (BITSEA; Briggs-Gowan & Carter 2002)					✓	✓		
C21st Health Check (Glasgow University Affiliated Programme, 2001)					✓	✓		✓
Challenging Behaviour Perception Questionnaire (CBPQ; Williams & Rose, 2007)	✓							
Challenging Behaviour Rating Scale (CBRS; Balakrishman & Matheen, 2022)*				✓				
Children's Adjustment and Parent Self-efficacy Scale – Developmental Disability (CAPES-DD; Emser et al., 2016)					✓	✓		
Child Challenging Behaviour Scale (CCBS; Bourke-Taylor et al., 2010)					✓	✓		
Children's Social Behaviour Questionnaire (CSBQ; Luteijn et al., 2000)					✓	✓		
Clinical Behaviour Checklist for Persons with Intellectual Disabilities (CBCPID; Marston et al., 1997)					✓	✓		
Clinical Global Impression Scale – Improvement/Severity (CGI-I/S; Guy, 1976)					✓[b]	✓		
Conners Parent Rating Scales (Conners 1997; 2008)					✓	✓		
Conners Teacher Rating Scales (Conners 1989; 2008)	✓							
Conditional Reasoning Problems (CRP; James et al., 2005)	✓							
Current Risk of Violence (CuRV; Lofthouse et al., 2014)					✓	✓		

Appendices Table 5 Continued

Measurement tool	Could not be obtained for assessment of eligibility	Not available	Not developed for broader use	Not available in English language	<50% of items in scale or subscale are BtC	Measure does not quantitatively assess functions of BtC	Descriptive clinical interview without scoring criteria	No ratings indicative of impact or harm
Developmental Behaviour Checklist – Primary caregiver (DBC-P; Einfeld et al., 2002)					✓	✓		
Developmental Behaviour Checklist – Teacher (DBC-T; Einfeld et al., 2002))					✓	✓		
Developmental Behaviour Checklist – Adult (DBC-A; Mohr et al., 2005)					✓	✓		
Developmental Behaviour Checklist – Under 4 (DBC-U4; Einfeld et al., 2002)		✓[c]			✓	✓		
Developmental Behaviour Checklist – Short Form (DBC-P24; Taffe et al., 2007)					✓	✓		
Developmental Behaviour Checklist – Monitoring (DBC-M; Einfeld et al., 2002)					✓[b]	✓		
Difficult Behaviour Assessment Form (DBAF; Hudson et al., 2008)					✓	✓		
Direct Aggression and Restriction Observation Checklist for Routine Observation (AROC; Nagy et al., 2019)				✓[d]				
Disability Assessment Schedule for Behaviour Problems (DAS-B; Holmes et al., 1982)					✓	✓		
Disruptive Behavioural Social Problems (DBSP; Young et al., 2003)					✓	✓		
Emotion Problems Scale – Behaviour Report Scale (EPS-BRS; Prout & Strohmer, 1991)	✓							
Eyberg Child Behaviour Inventory (ECBI; Eyberg & Pincus, 1999)					✓	✓		
Functional Assessment Interview (FAI; O'Neill et al., 1997; O'Neill et al., 1990)							✓	
Global Assessment of Individual Behaviour – Prader Willi Syndrome (GAIB-PWS; Tasse et al., 2002)					✓	✓		
Initial Behaviour Observation and Rating Scale (IBORS; Buskermolen et al., 2013)			✓					
InterRAI-Intellectual Disability (InterRAI-ID; Martin et al., 2007)					✓	✓		
Interview Protocol for Challenging Behaviour (Davis, 2004)							✓	
Inventory for Client and Agency Planning (ICAP; Bruininks et al., 1986)	✓[e]							
Health Crisis Assessment Scale (HCAS; Kalb et al., 2017)					✓	✓		
Learning Disability Casemix Scale (LDCS; Pendaries, 1997)	✓							
NeuroPsychiatric Inventory – Intellectual Disability (NPI-ID; Lundqvist et al., 2020)					✓	✓		

Appendices Table 5 Continued

Measurement tool	Could not be obtained for assessment of eligibility	Not available	Not developed for broader use	Not available in English language	<50% of items in scale or subscale are BtC	Measure does not quantitatively assess functions of BtC	Descriptive clinical interview without scoring criteria	No ratings indicative of impact or harm
Novaco Anger Scale (NAS; Novaco & Taylor, 2014)					✓	✓		
Oklahoma Quality Assurance Questionnaire (Oklahoma State University, 1992)					✓	✓		
Paediatric Behaviour Scale (Lindgren & Koeppl, 1987)		✓						
Paediatric Symptom Checklist (Jellineck et al., 1986)					✓	✓		
Prader-Willi Syndrome Behaviour Questionnaire (PWSBQ; Avrahamy et al., 2015)					✓	✓		
Problem and Target Scales (Marks et al., 1977)					✓	✓		
Psychiatric Assessment Schedule for Adults with Developmental Disability (PASS-ADD; Moss et al., 1993)					✓	✓		
Reiss Scale for Children's Dual Diagnosis (RSCDD; Reiss & Valenti-Hein, 1994)	✓							
Reiss Scale for Maladaptive Behaviour (RSMB; Reiss, 1998)	✓							
Rett Syndrome Behavioural Questionnaire (RSBQ; Mount et al., 2002)					✓	✓		
Rossago Behavioural Checklist (Politi et al., 2008)					✓	✓		
Self-injurious Behaviour Questionnaire (SIBQ; Gualtieri & Schroeder, 1989)					✓	✓		
Self-injury and Self-restraint checklist (Powell et al., 1996)								✓
Short Dynamic Risk Scale (SDRS; Quinsey, 2004)					✓	✓		
Social Skills Improvement - Rating System (SSIS-RS; Gresham & Elliott, 2008)	✓							
Spielberger State-Trait Anger Expression Inventory (STAXI; Spielberger, 1988)	✓							
Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997; 2001)					✓	✓		
Sutter-Eyberg Student Behaviour Inventory (SESBI; Eyberg & Pincus, 1999)					✓	✓		
Systematic Screening for Behaviour Disorders (SSBD; Walker & Severson, 1990)					✓	✓		
Tuberous Sclerosis Associated Neuropsychiatric Disorders (TAND; De Vries et al., 2015)					✓	✓		
Vineland Adaptive Behaviour Scales – Maladaptive Behaviour Domain (VABS-MBD; Sparrow et al., 1984)					✓	✓		

Appendices Table 5 Continued

Measurement tool	Could not be obtained for assessment of eligibility	Not available	Not developed for broader use	Not available in English language	<50% of items in scale or subscale are BtC	Measure does not quantitatively assess functions of BtC	Descriptive clinical interview without scoring criteria	No ratings indicative of impact or harm
Violence Risk Appraisal Guide (VRAG; Quinsey et al., 2006)					✓	✓		
Violence Risk Screening – 10 (V-RISK-10; Hartvig et al., 2007)					✓	✓		

Note. a= measure out of print and no longer available. b= measurement tool does not have a specific set of behavioural items; the tool can be used to rate behaviour defined by the user. c= measurement tool not widely available at the time of this review. d= measurement tool is only available in German. e= The SIB-R is the latest version of the ICAP; see Appendices Table 4. *The CBRS (Balakrishnan & Matheen, 2022) was identified as newly published measure of BtC published after the preliminary search took place. As of 2023, the measure has been administered in non-English language; therefore, the CBRS did not meet criteria for inclusion in this review.

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Appendix 13: Chapter Three full psychometric search strategy per database

Appendices Table 6

Intellectual disability, measurement tool and psychometric property search terms for each database

Web of Science (Core Collection)

Inclusion dates: All years (1900-2023); Date of search: 05.05.2023

Intellectual disability	TS=((intellectual* disab* OR learning disab* OR developmental* disab* OR mental* retard* OR mental* handicap*) OR ("Smith-Magenis syndrome" OR "Smith Magenis" OR "17p- syndrome" OR "17p11.2 monosomy" OR "Chromosome 17p deletion syndrome" OR "Deletion 17p syndrome" OR "partial monosomy 17p" OR "del(17)(p11.2)") OR (“Angelman*” OR “Angelman* syndrome” OR “Happy puppet syndrome” OR “Happy puppet”) OR (“Prader-Willi syndrome” OR “Prader-Labhart-Willi syndrome” OR “Willi-Prader syndrome”) OR (“Fragile X” OR “Fragile-X” OR “Fragile X syndrome” OR “FRAXA syndrome” OR “AFRAX” OR “Martin-Bell* syndrome” OR “Marker X syndrome” OR “fraX syndrome” OR “fra(X) syndrome” OR “X-linked mental retardation” OR “Macroorchidism” OR “Escalante* syndrome” OR “Escalante*”) OR (“Cri-du-chat syndrome” OR “Cat cry syndrome” OR “5p deletion syndrome” OR “5p- syndrome” OR “chromosome 5p- syndrome” OR “Monosomy 5p” OR “chromosome 5p deletion syndrome”) OR (“Cornelia de Lange* syndrome” OR “CDLS” OR “De Lange* syndrome” OR “Branchmann-De Lange* syndrome” OR “BDLS” OR “Brachmann* syndrome” OR “Amstelodamensis typus degenerativus” OR “Amsterdam dwarf syndrome” OR “Amsterdam dwarfism” OR “Typus degenerativus amstelodamensis”) OR (“Down* syndrome” OR “Trisomy 21” OR “Trisomy G” OR “47,XX,+21” OR “47,XY,+21”) OR (“Lowe syndrome” OR “Cerebrooculorenal syndrome” OR “Lowe oculocerebrorenal syndrome” OR “Oculocerebrorenal syndrome” OR “Oculocerebrorenal syndrome of Lowe” OR “Phosphatidylinositol-4,5-bisphosphate-5-phosphatase deficiency”) OR (“7q11.23*” OR “7q11.23 duplication syndrome” OR “7q11.23 microduplication syndrome” OR “chromosome 7q11.23 duplication” OR “chromosome 7q11.23 duplication syndrome” OR “dup(7)(q11.23)” OR “Somerville-Van der Aa syndrome” OR “trisomy 7q11.23” OR “WBS duplication syndrome” OR “Williams-Beuren region duplication syndrome” OR “william* syndrome”) OR (“Velocardiofacial syndrome” OR “Velo-cardio-facial syndrome” OR “DiGeorge* syndrome” OR “Conotruncal anomaly face syndrome” OR “CATCH22” OR “Autosomal dominant Opitz G/BBB syndrome” OR “Autosomal dominant Opitz G BBB syndrome” OR “Cayler cardiofacial syndrome” OR “Deletion 22q11/2 syndrome” OR “22q11/2 deletion syndrome” OR “22q11/2DS” OR “22q11 deletion syndrome” OR “Sedlackova* syndrome” OR “Shprintzen* syndrome”) OR (“Rett* syndrome” OR “Rett* disorder” OR “Cerebroatrophic hyperammonemia” OR “Autism-dementiaataxia-loss of purposeful hand use syndrome”) OR (“CHARGE syndrome” OR “CHARGE association” OR “Hall-Hittner* syndrome” OR “Hall* Hittner* syndrome” OR “Coloboma”) OR (“Tuberous sclerosis” OR “Tuberous sclerosis syndrome” OR “Bourneville* disease” OR “Bourneville* phakomatosis” OR “Cerebral sclerosis” OR “Cerebral sclerosis syndrome” OR “Epiloia” OR “Sclerosis tuberose” OR “Tuberose sclerosis” OR “Tuberose sclerosis syndrome” OR “Tuberous sclerosis complex”) OR (“Rubinstein-Taybi* syndrome” OR “RSTS” OR “Broad thumb-hallux syndrome”) OR (“Phelan-McDermid* syndrome” OR “Phelan-McDermid*” OR “22q13 deletion syndrome” OR “Deletion 22q13 syndrome” OR “Deletion 22q13.3 syndrome” OR “Monosomy 22q13” OR “22q13.3 deletion syndrome”) OR (“KBG syndrome”) OR (“Pitt-Hopkin*” OR “Pitt-Hopkin* syndrome” OR “PTHS”) OR (“48,XXYY syndrome” OR “XXYY syndrome”) OR (“ADNP syndrome” OR “ADNP-related intellectual disability and autism spectrum disorder” OR “ADNP-related multiple congenital anomalies-intellectual disability-autism spectrum disorder” OR “Helsmoortel-van der Aa syndrome” OR “HVDAS” OR “Mental

retardation, autosomal dominant 28" OR "MRD28" OR "ADNP-Related ID/ASD" OR "ADNP-Related disorder") OR ("SYNGAP1-related intellectual disability" OR "SYNGAP-ID" OR "SYNGAP1 syndrome" OR "MRD5" OR "SYNGAP1-related developmental and epileptic encephalopathy") OR ("Phenylketonuria" OR "Phenylalanine hydroxylase" OR "Folling* disease" OR "Folling* syndrome" OR "PAH deficiency" OR "PAH deficiency disease" OR "Phenylalanine hydroxylase deficiency disease" OR "Phenylalanine hydroxylase deficiency" OR "PKU" OR "Oligophrenia phenylpyruvica" OR "Deficiency Disease, Phenylalanine Hydroxylase") OR ("Sotos*" OR "Sotos* syndrome" OR "Cerebral gigantism" OR "Sotos* sequence") OR ("1p36 deletion syndrome" OR "1p36DS" OR "chromosome 1p36 deletion syndrome" OR "distal monosomy 1p36" OR "monosomy 1p36 syndrome") OR ("8p23 deletion syndrome" OR "8p23DS") OR ("Joubert*" OR "Joubert* syndrome" OR "Joubert-Bolthauser* syndrome" OR "JBTS" OR "Cerebello-oculo-renal syndrome" OR "Cerebello-oculo-renal syndrome 1" OR "Cerebellooculorenal syndrome 1" OR "Cerebellooculorenal syndrome" OR "Cerebellar vermis agenesis" OR "Cerebelloparenchymal disorder 4" OR "Cerebelloparenchymal disorder" OR "CPD4" OR "Familial aplasia of the vermis") OR ("16p11.2 deletion syndrome" OR "16p11.2DS" OR "AUTS14A") OR ("Lesch-Nyhan syndrome" OR "Choreoathetosis self-mutilation syndrome" OR "Complete HPRT deficiency" OR "Complete hypoxanthine-guanine phosphoribosyltransferase deficiency" OR "HPRT deficiency" OR "Deficiency of guanine phosphoribosyltransferase" OR "Deficiency of hypoxanthine phosphoribosyltransferase" OR "HGPRT deficiency" OR "Hypoxanthine guanine phosphoribosyltransferase deficiency" OR "Hypoxanthine phosphoribosyltransferase deficiency" OR "Juvenile gout, choreoathetosis, mental retardation syndrome" OR "Juvenile hyperuricemia syndrome" OR "Lesch-Nyhan disease" OR "Primary hyperuricemia syndrome" OR "Total HPRT deficiency" OR "Total hypoxanthine-guanine phosphoribosyl transferase deficiency" OR "X-linked hyperuricemia" OR "X-linked primary hyperuricemia" OR "X-linked uric aciduria enzyme defect") OR ("Cohen* syndrome" OR "Norio* syndrome" OR "Obesity-hypotonia syndrome" OR "Pepper* syndrome" OR "Prominent incisors-besityhypotonia syndrome" OR "Hypotonia obesity and prominent incisors") OR ("Dup15q syndrome" OR "Duplication/inversion 15q11" OR "Idic(15)" OR "Inv dup(15)" OR "Inverted duplication 15" OR "Isodicentric chromosome 15" OR "Isodicentric chromosome 15 syndrome" OR "Non-distal tetrasomy 15q" OR "15q duplication syndrome" OR "15q11-q13 duplication syndrome") OR ("Malan* syndrome" OR "MALNS") OR ("Marshall-smith* syndrome" OR "MRSHSS") OR ("Distal 18q deletion syndrome" OR "18q deletion syndrome" OR "18q- syndrome" OR "Chromosome 18 long arm deletion syndrome" OR "Chromosome 18q deletion syndrome" OR "Chromosome 18q monosomy" OR "Chromosome 18q- syndrome" OR "Del(18q) syndrome" OR "Monosomy 18q" OR "proximal 18q deletion syndrome" OR "tetrasomy 18p" OR "18p isochromosome" OR "18p tetrasomy") OR ("Kleefstra syndrome" OR "9q subtelomeric deletion syndrome" OR "9q- syndrome" OR "9q34.3 deletion syndrome" OR "9q34.3 microdeletion syndrome" OR "Chromosome 9q deletion syndrome" or "9q34 deletion" OR "9Qstds") OR ("Potocki-Lupski syndrome" OR "17p11.2 duplication syndrome" OR "17p11.2 microduplication syndrome" OR "chromosome 17p11.2 duplication syndrome" OR "Dup(17)(p11.2p11.2)" OR "Duplication 17p11.2 syndrome" OR "PTLS") OR ("Bardet-Biedl syndrome" OR "Bardet-Biedl*" OR "Laurence-Moon-Bardet-Biedl syndrome" OR "LMBBS") OR ("2q37 deletion syndrome" OR "2q37DS" OR "2q37 microdeletion syndrome" OR "Albright hereditary osteodystrophy-like syndrome" OR "Brachydactyly-mental retardation syndrome" OR "Chromosome 2q37 deletion syndrome" OR "Deletion 2q37" OR "Monosomy 2q37") OR ("Mucopolysaccharidosis type III" OR "MPS III" OR "Mucopolysaccharidosis III" OR "Sanfilippo* syndrome") OR ("Cardiofaciocutaneous syndrome" OR "Cardio-facio-cutaneous syndrome" OR "CFC syndrome") OR ("Smith-Lemli-Opitz syndrome" OR "7-dehydrocholesterol reductase deficiency" OR "RSH syndrome" OR "SLO syndrome") OR ("Mowat-Wilson* syndrome" OR "Hirschsprung disease-mental retardation syndrome" OR "Hirschsprung Disease – Intellectual Disability Syndrome"))

Measurement tools

(TS=((“aberrant behavio$r checklist” OR “aberrant behavio$r checklist-community” OR “aberrant behavio$r checklist – community” OR “ABC-C” OR “aberrant behavio$r checklist-residential” OR “aberrant behavio$r checklist – residential”) OR (“Aggression and Self-Injurious Behaviour Questionnaire” OR “ASIQ”) OR (“autism spectrum disorder* behavio$r problem* for adult*” Or “autism spectrum disorder* - behavio$r problem* for adult*” or “autism spectrum disorder* problem behavio$r for adult*” or “autism spectrum disorder* - problem behavio$r for adult*” or “ASD-BPA” OR “ASD-PBA” OR “autism spectrum disorder* behavio$r problem* for children” OR “autism spectrum disorder* - behavio$r problem* for children” OR “autism spectrum disorder* problem behavio$r for children” OR “autism spectrum disorder* - problem behavio$r for children” OR “ASD-BPC” OR “ASD-PBC”) OR (“baby and infant screen for children with autism traits” OR “BISCUIT”) OR (“child behavio$r checklist” OR “CBCL 1.5-5” OR “CBCL preschool” OR “CBCL 6-18” OR “TRF 6-18” OR “CBCL TRF” OR “teacher report form” OR “youth self-report” OR “youth self-report 11-18” OR “YSR” OR “adult behavio$r checklist” OR “ABCL” OR “achenbach checklist*”) OR (“children$s scale of hostility and aggression” OR “children$s scale of hostility and aggression – reactive/proactive ” OR “C-SHARP” OR “adult* scale of hostility and aggression” OR “adult* scale of hostility and aggression – reactive/proactive” OR “A-SHARP”) OR (“aggressive behavio$r scale”) OR (“behavio$r problem* inventory-01” OR “behavio$r problem* inventory” OR “BPI-Short form” OR “behavio$r problem* inventory – short form” OR “BPI-S”) OR (“challenging behavio$r perception questionnaire” OR “CHABA”) OR (“challenging behavio$r interview”) OR (“challenging behavio$r questionnaire” OR “CBQ”) OR (“challenging behavio$r scale”) OR (“checklist* of challenging behavio$r”) OR (“contextual assessment inventory for problem behavio$r” OR “contextual assessment inventory”) OR (“developmental disabilities profile – maladaptive behavio$r domain” OR “maladaptive behavio$r domain from the developmental disabilities profile” OR “developmental disabilities profile”) OR (“diagnostic assessment for severely handicapped” OR “DASH-II” OR “DASH-2”) OR (“the early childhood behavio$r screen” OR “ECBS”) OR (“functional assessment for multiple causality”) OR (“overt aggression scale” OR “Overt aggression scale modified for neurorehabilitation” OR “OAS-MNR” OR “modified overt aggression scale” OR “MOAS” OR “extended modified overt aggression scale” OR “extended MOAS” OR “institute for basic research modified overt aggression scale” OR “IBR-MOAS”) OR (“functional analysis checklist”) OR (“functional assessment screening tool” OR “FAST”) OR (“great outcomes for kids impacted by severe developmental disabilities” OR “GO4KIDDS”) OR (“learning disability needs assessment tool” OR “LDNAT”) OR (“motivation assessment scale”) OR (“initial behavio$r* assessment and protective equipment decision key” OR “IBA-PEDK”) OR (“Nisonger child behavio$r rating form” OR “NCBRF”) OR (“pervasive developmental disorder behavio$r inventory” OR “PDDBI”) OR (“problem behavio$r checklist”) OR (“psychopathology checklists for adults with intellectual disability*” OR “P-AID”) OR (“questions about behavio$r* function” OR “QABF”) OR (“Sanfilippo behavio$r rating scale*”) OR (“scales of independent behavio$r revised” OR “scales of independent behavio$r-revised” OR “SIB-R”) OR (“self-injury, aggression and destruction screening questionnaire” OR “SAD-SQ”) OR (“self-injury trauma scale”) OR (“staff observation aggression scale” OR “staff observation aggression scale – revised” OR “SOAS” OR “SOAS-R”)) OR TS=((interview) NEAR/2 ("temper outburst*")) OR TS=(("skin picking") NEAR/10 ("semi-structured interview")) OR ALL=("individual schedule of the challenging behavio$r survey"))

Psychometric properties	TS=((psychometr* OR clinimetr* OR clinometr* OR "outcome assessment" OR "outcome measure" OR "observer variation" OR reproducib* OR reliab* OR unreliab* OR valid* OR coefficient OR homogeneity OR homogenous OR "internal consistency" OR agreement OR precision OR imprecision OR "precise values" OR test-retest OR stability OR interrater OR inter-rater OR intrarater OR intra-rater OR intertester OR inter-tester OR intratester OR intra-tester OR interobserver OR inter-observer OR intraobserver OR intra-observer OR intertechnician OR inter-technician OR intratechnician OR intra-technician OR interexaminer OR inter-examiner OR intraexaminer OR intra-examiner OR interassay OR inter-assay OR intraassay OR intra-assay OR interindividual OR inter-individual OR intraindividual OR intra-individual OR interparticipant OR inter-participant OR intraparticipant OR intra-participant OR kappa OR kappa's OR kappas OR repeatab* OR generaliza* OR generalisa* OR concordance OR discriminative OR "known group" OR "factor analys*" OR dimension* OR subscale* OR "item discriminant" OR "interscale correlation*" OR error* OR "individual variability" OR "standard error of measurement" OR sensitiv* OR responsive* OR "meaningful change" OR "ceiling effect" OR "floor effect" OR "Item response model" OR IRT OR Rasch OR "Differential item functioning" OR DIF OR "computer adaptive testing" OR "item bank" OR "cross-cultural equivalence" OR "cronbach* alpha*" OR "replicab* measure*" OR "replicab* finding*" OR "replicab* result*" OR "replicab* test*" OR "repeated measure*" OR "repeated finding*" OR "repeated result*" OR "repeated test*" OR "item correlation*" OR "item selection*" OR "item reduction*" OR "Test-retest" OR "intraclass correlation*" OR "multitrait scaling anays*" OR "uncertainty measur*" OR "variability analys*" OR "variability value*" OR "minimal* important change" OR "minimal* important difference" OR "minimal* significant change" OR "minimal* significant difference" OR "minimal* detectable change" OR "minimal* detectable difference" OR "clinical* important change" OR "clinical* important difference" OR "clinical* significant change" OR "clinical* significant difference" OR "clinical* detectable change" OR "clinical* detectable difference" OR "small* real change" OR "small* real difference" OR "small* detectable change" OR "small* detectable difference"))

Ovid PsycINFO

Inclusion dates: All years (1967-May week 1 2023); Date of search: 05.05.2023

Intellectual disability	(intellectual* disab* or learning disab* or developmental* disab* or mental* retard* or mental* handicap* or ("Smith-Magenis syndrome" or "Smith Magenis" or "17p- syndrome" or "17p11.2 monosomy" or "Chromosome 17p deletion syndrome" or "Deletion 17p syndrome" or "partial monosomy 17p" or "del(17)(p11.2)") or ("Angelman*" or "Angelman* syndrome" or "Happy puppet syndrome" or "Happy puppet") or ("Prader-Willi syndrome" or "Prader-Labhart-Willi syndrome" or "Willi-Prader syndrome") or ("Fragile X" or "Fragile-X" or "Fragile X syndrome" or "FRAXA syndrome" or "AFRAX" or "Martin-Bell* syndrome" or "Marker X syndrome" or "fraX syndrome" or "fra(X) syndrome" or "X-linked mental retardation" or "Macroorchidism" or "Escalante* syndrome" or "Escalante*") or ("Cri-du-chat syndrome" or "Cat cry syndrome" or "5p deletion syndrome" or "5p- syndrome" or "chromosome 5p- syndrome" or "Monosomy 5p" or "chromosome 5p deletion syndrome") or ("Cornelia de Lange* syndrome" or "CDLS" or "De Lange* syndrome" or "Branchmann-De Lange* syndrome" or "BDLS" or "Brachmann* syndrome" or "Amstelodamensis typus degenerativus" or "Amsterdam dwarf syndrome" or "Amsterdam dwarfism" or "Typus degenerativus amstelodamensis") or ("Down* syndrome" or "Trisomy 21" or "Trisomy G" or "47,XX,+21" or "47,XY,+21") or ("Lowe syndrome" or "Cerebrooculorenal syndrome" or "Lowe oculocerebrorenal syndrome" or "Oculocerebrorenal syndrome" or "Oculocerebrorenal syndrome of Lowe" or "Phosphatidylinositol-4,5-bisphosphate-5-phosphatase deficiency") or ("7q11.23*" or "7q11.23 duplication syndrome" or "7q11.23 microduplication syndrome" or "chromosome 7q11.23 duplication" or "chromosome 7q11.23 duplication syndrome" or "dup(7)(q11.23)" or "Somerville-Van der Aa syndrome" or "trisomy 7q11.23" or "WBS duplication syndrome" or "Williams-Beuren region duplication syndrome" or "William* syndrome") or ("Velocardiofacial syndrome" or "Velo-cardio-facial syndrome" or "DiGeorge* syndrome" or "Conotruncal anomaly face syndrome" or "CATCH22" or "Autosomal dominant Opitz G/BBB syndrome" or "Autosomal dominant Opitz G BBB syndrome" or "Cayler cardiofacial syndrome" or "Deletion 22q11/2 syndrome" or "22q11/2 deletion syndrome" or "22q11/2DS" or "22q11 deletion syndrome" or "Sedlackova* syndrome" or "Shprintzen* syndrome") or ("Rett* syndrome" or "Rett* disorder" or "Cerebroatrophic hyperammonemia" or "Autism-dementiaataxia-

loss of purposeful hand use syndrome") or ("CHARGE syndrome" or "CHARGE association" or "Hall-Hittner* syndrome" or "Hall* Hittner* syndrome" or "Coloboma") or ("Tuberous sclerosis" or "Tuberous sclerosis syndrome" or "Bourneville* disease" or "Bourneville* phakomatosis" or "Cerebral sclerosis" or "Cerebral sclerosis syndrome" or "Epiloia" or "Sclerosis tuberose" or "Tuberose sclerosis" or "Tuberose sclerosis syndrome" or "Tuberous sclerosis complex") or ("Rubinstein-Taybi* syndrome" or "RSTS" or "Broad thumb-hallux syndrome") or ("Phelan-McDermid* syndrome" or "Phelan-McDermid*" or "22q13 deletion syndrome" or "Deletion 22q13 syndrome" or "Deletion 22q13.3 syndrome" or "Monosomy 22q13" or "22q13.3 deletion syndrome") or "KBG syndrome" or ("Pitt-Hopkin*" or "Pitt-Hopkin* syndrome" or "PTHS") or ("48,XXYY syndrome" or "XXYY syndrome") or ("ADNP syndrome" or "ADNP-related intellectual disability and autism spectrum disorder" or "ADNP-related multiple congenital anomalies-intellectual disability-autism spectrum disorder" or "Helsmoortel-van der Aa syndrome" or "HVDAS" or "Mental retardation, autosomal dominant 28" or "MRD28" or "ADNP-Related ID/ASD" or "ADNP-Related disorder") or ("SYNGAP1-related intellectual disability" or "SYNGAP-ID" or "SYNGAP1 syndrome" or "MRD5" or "SYNGAP1-related developmental and epileptic encephalopathy") or ("Phenylketonuria" or "Phenylalanine hydroxylase" or "Folling* disease" or "Folling* syndrome" or "PAH deficiency" or "PAH deficiency disease" or "Phenylalanine hydroxylase deficiency disease" or "Phenylalanine hydroxylase deficiency" or "PKU" or "Oligophrenia phenylpyruvica" or "Deficiency Disease, Phenylalanine Hydroxylase") or ("Sotos*" or "Sotos* syndrome" or "Cerebral gigantism" or "Sotos* sequence") or ("1p36 deletion syndrome" or "1p36DS" or "chromosome 1p36 deletion syndrome" or "distal monosomy 1p36" or "monosomy 1p36 syndrome") or ("8p23 deletion syndrome" or "8p23DS") or ("Joubert*" or "Joubert* syndrome" or "Joubert-Bolthauser* syndrome" or "JBTS" or "Cerebello-oculo-renal syndrome" or "Cerebello-oculo-renal syndrome 1" or "Cerebellooculorenal syndrome 1" or "Cerebellooculorenal syndrome" or "Cerebellar vermis agenesis" or "Cerebelloparenchymal disorder 4" or "Cerebelloparenchymal disorder" or "CPD4" or "Familial aplasia of the vermis") or ("16p11.2 deletion syndrome" or "16p11.2DS" or "AUTS14A") or ("Lesch-Nyhan syndrome" or "Choreoathetosis self-mutilation syndrome" or "Complete HPRT deficiency" or "Complete hypoxanthine-guanine phosphoribosyltransferase deficiency" or "HPRT deficiency" or "Deficiency of guanine phosphoribosyltransferase" or "Deficiency of hypoxanthine phosphoribosyltransferase" or "HGPRT deficiency" or "Hypoxanthine guanine phosphoribosyltransferase deficiency" or "Hypoxanthine phosphoribosyltransferase deficiency" or "Juvenile gout, choreoathetosis, mental retardation syndrome" or "Juvenile hyperuricemia syndrome" or "Lesch-Nyhan disease" or "Primary hyperuricemia syndrome" or "Total HPRT deficiency" or "Total hypoxanthine-guanine phosphoribosyl transferase deficiency" or "X-linked hyperuricemia" or "X-linked primary hyperuricemia" or "X-linked uric aciduria enzyme defect") or ("Cohen* syndrome" or "Norio* syndrome" or "Obesity-hypotonia syndrome" or "Pepper* syndrome" or "Prominent incisors-besityhypotonia syndrome" or "Hypotonia obesity and prominent incisors") or ("Dup15q syndrome" or "Duplication/inversion 15q11" or "Idic(15)" or "Inv dup(15)" or "Inverted duplication 15" or "Isodicentric chromosome 15" or "Isodicentric chromosome 15 syndrome" or "Non-distal tetrasomy 15q" or "15q duplication syndrome" or "15q11-q13 duplication syndrome") or ("Malan* syndrome" or "MALNS") or ("Marshall-smith* syndrome" or "MRSHSS") or ("Distal 18q deletion syndrome" or "18q deletion syndrome" or "18q- syndrome" or "Chromosome 18 long arm deletion syndrome" or "Chromosome 18q deletion syndrome" or "Chromosome 18q monosomy" or "Chromosome 18q-syndrome" or "Del(18q) syndrome" or "Monosomy 18q" or "proximal 18q deletion syndrome" or "tetrasomy 18p" or "18p isochromosome" or "18p tetrasomy") or ("Kleefstra syndrome" or "9q subtelomeric deletion syndrome" or "9q- syndrome" or "9q34.3 deletion syndrome" or "9q34.3 microdeletion syndrome" or "Chromosome 9q deletion syndrome" or "q34 deletion" or "9Qstds") or ("Potocki-Lupski syndrome" or "17p11.2 duplication syndrome" or "17p11.2 microduplication syndrome" or "chromosome 17p11.2 duplication syndrome" or "Dup(17)(p11.2p11.2)" or "Duplication 17p11.2 syndrome" or "PTLS") or ("Bardet-Biedl syndrome" or "Bardet-Biedl*" or "Laurence-Moon-Bardet-Biedl syndrome" or "LMBBS") or ("2q37 deletion syndrome" or "2q37DS" or "2q37 microdeletion syndrome" or "Albright hereditary osteodystrophy-like syndrome" or "Brachydactyly-mental retardation syndrome" or "Chromosome 2q37 deletion syndrome" or "Deletion 2q37" or "Monosomy 2q37") or ("Mucopolysaccharidosis type III" or "MPS III" or "Mucopolysaccharidosis III" or "Sanfilippo* syndrome") or ("Cardiofaciocutaneous syndrome" or "Cardio-facio-cutaneous syndrome" or "CFC syndrome") or ("Smith-Lemli-Opitz syndrome" or "7-dehydrocholesterol reductase deficiency" or "RSH syndrome" or "SLO syndrome") or ("Mowat-Wilson* syndrome" or "Hirschsprung disease-mental retardation syndrome" or "Hirschsprung Disease – Intellectual Disability Syndrome")).ti,ab,tw. OR (exp Intellectual Development Disorder/) OR (exp Developmental Disabilities/) OR (exp Prader-Willi Syndrome/) OR (exp Fragile X

	Syndrome/) OR (exp Crying Cat Syndrome/) OR (exp Cornelia de Lange Syndrome/) OR (exp Downs Syndrome/) OR (exp Williams Syndrome/) OR (exp Rett Syndrome/) OR (exp Phenylketonuria/)
Measurement tools	("aberrant behavio?r checklist" or "aberrant behavio?r checklist-community" or "aberrant behavio?r checklist – community" or "ABC-C" or "aberrant behavio?r checklist-residential" or "aberrant behavio?r checklist – residential" or ("Aggression and Self-Injurious Behaviour Questionnaire" or "ASIQ") or ("autism spectrum disorder* behavio?r problem* for adult*" or "autism spectrum disorder* - behavio?r problem* for adult*" or "autism spectrum disorder* problem behavio?r for adult*" or "autism spectrum disorder* - problem behavio?r for adult*" or "ASD-BPA" or "ASD-PBA" or "autism spectrum disorder* behavio?r problem* for children" or "autism spectrum disorder* - behavio?r problem* for children" or "autism spectrum disorder* problem behavio?r for children" or "autism spectrum disorder* - problem behavio?r for children" or "ASD-BPC" or "ASD-PBC") or ("baby and infant screen for children with autism traits" or "BISCUIT") or ("child behavio?r checklist" or "CBCL 1.5-5" or "CBCL preschool" or "CBCL 6-18" or "TRF 6-18" or "CBCL TRF" or "teacher report form" or "youth self-report" or "youth self-report 11-18" or "YSR" or "adult behavio?r checklist" or "ABCL" or "achenbach checklist*") or ("children?s scale of hostility and aggression" or "children?s scale of hostility and aggression – reactive/proactive" or "C-SHARP" or "adult* scale of hostility and aggression" or "adult* scale of hostility and aggression – reactive/proactive" or "A-SHARP") or "aggressive behavio?r scale" or ("behavio?r problem* inventory-01" or "behavio?r problem* inventory" or "BPI-Short form" or "behavio?r problem* inventory – short form" or "BPI-S") or ("challenging behavio?r perception questionnaire" or "CHABA") or "challenging behavio?r interview" or ("challenging behavio?r questionnaire" or "CBQ") or "challenging behavio?r scale" or "checklist* of challenging behavio?r" or ("contextual assessment inventory for problem behavio?r" or "contextual assessment inventory") or ("developmental disabilities profile – maladaptive behavio?r domain" or "maladaptive behavio?r domain from the developmental disabilities profile" or "developmental disabilities profile") or ("diagnostic assessment for severely handicapped" or "DASH-II" or "DASH-2") or ("the early childhood behavio?r screen" or "ECBS") or "functional assessment for multiple causality" or ("overt aggression scale" or "Overt aggression scale modified for neurorehabilitation" or "OAS-MNR" or "modified overt aggression scale" or "MOAS" or "extended modified overt aggression scale" or "extended MOAS" or "institute for basic research modified overt aggression scale" or "IBR-MOAS") or "functional analysis checklist" or ("functional assessment screening tool" or "FAST") or ("great outcomes for kids impacted by severe developmental disabilities" or "GO4KIDDS") or ("learning disability needs assessment tool" or "LDNAT") or "motivation assessment scale" or ("initial behavio?r* assessment and protective equipment decision key" or "IBA-PEDK") or ("Nisonger child behavio?r rating form" or "NCBRF") or ("pervasive developmental disorder behavio?r inventory" or "PDDBI") or "problem behavio?r checklist" or ("psychopathology checklists for adults with intellectual disability*" or "P-AID") or ("questions about behavio?r* function" or "QABF") or "Sanfilippo behavio?r rating scale*" or ("scales of independent behavio?r revised" or "scales of independent behavio?r-revised" or "SIB-R") or ("self-injury, aggression and destruction screening questionnaire" or "SAD-SQ") or "self-injury trauma scale" or ("staff observation aggression scale" or "staff observation aggression scale – revised" or "SOAS" or "SOAS-R") or "individual schedule of the challenging behavio?r survey" or (interview adj2 "temper outburst*") or ("skin picking" adj10 "semi-structured interview")).ti,ab,tw.
Psychometric properties	(psychometr* or clinimetr* or clinometr* or "outcome assessment" or "outcome measure" or "observer variation" or reproducib* or reliab* or unreliab* or valid* or coefficient or homogeneity or homogenous or "internal consistency" or agreement or precision or imprecision or "precise values" or test-retest or stability or interrater or inter-rater or intrarater or intra-rater or intertester or inter-tester or intratester or intra-tester or interobserver or inter-observer or intraobserver or intra-observer or intertechnician or inter-technician or intratechnician or intra-technician or interexaminer or inter-examiner or intraexaminer or intra-examiner or interassay or inter-assay or intraassay or intra-assay or interindividual or inter-individual or intraindividual or intra-individual or interparticipant or inter-participant or intraparticipant or intra-participant or kappa or kappa's or kappas or repeatab* or generaliza* or generalisa* or concordance or discriminative or "known group" or "factor analys*" or dimension* or subscale* or "item discriminant" or "interscale correlation*" or error* or "individual variability" or "standard error of measurement" or sensitiv* or responsive* or "meaningful change" or "ceiling effect" or "floor effect" or "Item response model" or IRT or Rasch or "Differential item functioning" or DIF or "computer adaptive testing" or "item bank" or "cross-cultural equivalence" or ("cronbach* alpha*" or "replicab* measure*" or "replicab* finding*" or "replicab* result*" or "replicab* test*" or "repeated

measure*" or "repeated finding*" or "repeated result*" or "repeated test*" or "item correlation*" or "item selection*" or "item reduction*" or "Test-retest" or "intraclass correlation*" or "multitrait scaling anays*" or "uncertainty measur*" or "variability analys*" or "variability value*" or "minimal* important change" or "minimal* important difference" or "minimal* significant change" or "minimal* significant difference" or "minimal* detectable change" or "minimal* detectable difference" or "clinical* important change" or "clinical* important difference" or "clinical* significant change" or "clinical* significant difference" or "clinical* detectable change" or "clinical* detectable difference" or "small* real change" or "small* real difference" or "small* detectable change" or "small* detectable difference")).mp,ti,ab,tw. or "Psychometrics & Statistics & Methodology".mp. or "research methods & experimental design".mp. OR (exp Measurement/) OR (exp Error Analysis/) OR (exp Test Construction/) OR (exp Interrater Reliability/) OR (exp Content Analysis/) OR (exp "Error of Measurement"/) OR (exp Factor Structure/) OR (exp Testing Methods/) OR (exp Statistical Reliability/) OR (exp "Consistency (Measurement)"/) OR (exp Computerized Assessment/) OR (exp Factor Analysis/) OR (exp Prediction/) OR (exp Statistical Validity/) OR (exp Prediction Errors/) OR (exp Measurement Invariance/) OR (exp Variability Measurement/) OR (exp Psychometrics/)

Ovid Embase

Inclusion dates: All years (1900-2023 May 5); Date of search: 05.05.2023

Intellectual disability

(intellectual* disab* or learning disab* or developmental* disab* or mental* retard* or mental* handicap* or ("Smith-Magenis syndrome" or "Smith Magenis" or "17p- syndrome" or "17p11.2 monosomy" or "Chromosome 17p deletion syndrome" or "Deletion 17p syndrome" or "partial monosomy 17p" or "del(17)(p11.2)") or ("Angelman*" or "Angelman* syndrome" or "Happy puppet syndrome" or "Happy puppet") or ("Prader-Willi syndrome" or "Prader-Labhart-Willi syndrome" or "Willi-Prader syndrome") or ("Fragile X" or "Fragile-X" or "Fragile X syndrome" or "FRAXA syndrome" or "AFRAX" or "Martin-Bell* syndrome" or "Marker X syndrome" or "fraX syndrome" or "fra(X) syndrome" or "X-linked mental retardation" or "Macroorchidism" or "Escalante* syndrome" or "Escalante*") or ("Cri-du-chat syndrome" or "Cat cry syndrome" or "5p deletion syndrome" or "5p- syndrome" or "chromosome 5p- syndrome" or "Monosomy 5p" or "chromosome 5p deletion syndrome") or ("Cornelia de Lange* syndrome" or "CDLS" or "De Lange* syndrome" or "Branchmann-De Lange* syndrome" or "BDLS" or "Brachmann* syndrome" or "Amstelodamensis typus degenerativus" or "Amsterdam dwarf syndrome" or "Amsterdam dwarfism" or "Typus degenerativus amstelodamensis") or ("Down* syndrome" or "Trisomy 21" or "Trisomy G" or "47,XX,+21" or "47,XY,+21") or ("Lowe syndrome" or "Cerebrooculorenal syndrome" or "Lowe oculocerebrorenal syndrome" or "Oculocerebrorenal syndrome" or "Oculocerebrorenal syndrome of Lowe" or "Phosphatidylinositol-4,5-bisphosphate-5-phosphatase deficiency") or ("7q11.23*" or "7q11.23 duplication syndrome" or "7q11.23 microduplication syndrome" or "chromosome 7q11.23 duplication" or "chromosome 7q11.23 duplication syndrome" or "dup(7)(q11.23)" or "Somerville-Van der Aa syndrome" or "trisomy 7q11.23" or "WBS duplication syndrome" or "Williams-Beuren region duplication syndrome" or "William* syndrome") or ("Velocardiofacial syndrome" or "Velo-cardio-facial syndrome" or "DiGeorge* syndrome" or "Conotruncal anomaly face syndrome" or "CATCH22" or "Autosomal dominant Opitz G/BBB syndrome" or "Autosomal dominant Opitz G BBB syndrome" or "Cayler cardiofacial syndrome" or "Deletion 22q11/2 syndrome" or "22q11/2 deletion syndrome" or "22q11/2DS" or "22q11 deletion syndrome" or "Sedlackova* syndrome" or "Shprintzen* syndrome") or ("Rett* syndrome" or "Rett* disorder" or "Cerebroatrophic hyperammonemia" or "Autism-dementiaataxia-loss of purposeful hand use syndrome") or ("CHARGE syndrome" or "CHARGE association" or "Hall-Hittner* syndrome" or "Hall* Hittner* syndrome" or "Coloboma") or ("Tuberous sclerosis" or "Tuberous sclerosis syndrome" or "Bourneville* disease" or "Bourneville* phakomatosis" or "Cerebral sclerosis" or "Cerebral sclerosis syndrome" or "Epiloia" or "Sclerosis tuberose" or "Tuberose sclerosis" or "Tuberose sclerosis syndrome" or "Tuberous sclerosis complex") or ("Rubinstein-Taybi* syndrome" or "RSTS" or "Broad thumb-hallux syndrome") or ("Phelan-McDermid* syndrome" or "Phelan-McDermid*" or "22q13 deletion syndrome" or "Deletion 22q13 syndrome" or "Deletion 22q13.3 syndrome" or "Monosomy 22q13" or "22q13.3 deletion syndrome") or "KBG syndrome" or ("Pitt-Hopkin*" or "Pitt-Hopkin* syndrome" or "PTHS") or ("48,XXYY syndrome" or "XXYY syndrome") or ("ADNP syndrome" or "ADNP-related intellectual disability and autism spectrum disorder" or "ADNP-related multiple congenital anomalies-intellectual disability-autism spectrum disorder" or "Helsmoortel-van der Aa syndrome" or "HVDAS" or "Mental retardation, autosomal dominant 28" or "MRD28" or "ADNP-Related ID/ASD" or "ADNP-Related disorder") or ("SYNGAP1-related intellectual disability" or "SYNGAP-ID" or "SYNGAP1 syndrome" or

"MRD5" or "SYNGAP1-related developmental and epileptic encephalopathy") or ("Phenylketonuria" or "Phenylalanine hydroxylase" or "Folling* disease" or "Folling* syndrome" or "PAH deficiency" or "PAH deficiency disease" or "Phenylalanine hydroxylase deficiency disease" or "Phenylalanine hydroxylase deficiency" or "PKU" or "Oligophrenia phenylpyruvica" or "Deficiency Disease, Phenylalanine Hydroxylase") or ("Sotos*" or "Sotos* syndrome" or "Cerebral gigantism" or "Sotos* sequence") or ("1p36 deletion syndrome" or "1p36DS" or "chromosome 1p36 deletion syndrome" or "distal monosomy 1p36" or "monosomy 1p36 syndrome") or ("8p23 deletion syndrome" or "8p23DS") or ("Joubert*" or "Joubert* syndrome" or "Joubert-Bolthauser* syndrome" or "JBTS" or "Cerebello-oculo-renal syndrome" or "Cerebello-oculo-renal syndrome 1" or "Cerebellooculorenal syndrome 1" or "Cerebellooculorenal syndrome" or "Cerebellar vermis agenesis" or "Cerebelloparenchymal disorder 4" or "Cerebelloparenchymal disorder" or "CPD4" or "Familial aplasia of the vermis") or ("16p11.2 deletion syndrome" or "16p11.2DS" or "AUTS14A") or ("Lesch-Nyhan syndrome" or "Choreoathetosis self-mutilation syndrome" or "Complete HPRT deficiency" or "Complete hypoxanthine-guanine phosphoribosyltransferase deficiency" or "HPRT deficiency" or "Deficiency of guanine phosphoribosyltransferase" or "Deficiency of hypoxanthine phosphoribosyltransferase" or "HGPRT deficiency" or "Hypoxanthine guanine phosphoribosyltransferase deficiency" or "Hypoxanthine phosphoribosyltransferase deficiency" or "Juvenile gout, choreoathetosis, mental retardation syndrome" or "Juvenile hyperuricemia syndrome" or "Lesch-Nyhan disease" or "Primary hyperuricemia syndrome" or "Total HPRT deficiency" or "Total hypoxanthine-guanine phosphoribosyl transferase deficiency" or "X-linked hyperuricemia" or "X-linked primary hyperuricemia" or "X-linked uric aciduria enzyme defect") or ("Cohen* syndrome" or "Norio* syndrome" or "Obesity-hypotonia syndrome" or "Pepper* syndrome" or "Prominent incisors-besityhypotonia syndrome" or "Hypotonia obesity and prominent incisors") or ("Dup15q syndrome" or "Duplication/inversion 15q11" or "Idic(15)" or "Inv dup(15)" or "Inverted duplication 15" or "Isodicentric chromosome 15" or "Isodicentric chromosome 15 syndrome" or "Non-distal tetrasomy 15q" or "15q duplication syndrome" or "15q11-q13 duplication syndrome") or ("Malan* syndrome" or "MALNS") or ("Marshall-smith* syndrome" or "MRSHSS") or ("Distal 18q deletion syndrome" or "18q deletion syndrome" or "18q- syndrome" or "Chromosome 18 long arm deletion syndrome" or "Chromosome 18q deletion syndrome" or "Chromosome 18q monosomy" or "Chromosome 18q-syndrome" or "Del(18q) syndrome" or "Monosomy 18q" or "proximal 18q deletion syndrome" or "tetrasomy 18p" or "18p isochromosome" or "18p tetrasomy") or ("Kleefstra syndrome" or "9q subtelomeric deletion syndrome" or "9q- syndrome" or "9q34.3 deletion syndrome" or "9q34.3 microdeletion syndrome" or "Chromosome 9q deletion syndrome" or "q34 deletion" or "9Qstds") or ("Potocki-Lupski syndrome" or "17p11.2 duplication syndrome" or "17p11.2 microduplication syndrome" or "chromosome 17p11.2 duplication syndrome" or "Dup(17)(p11.2p11.2)" or "Duplication 17p11.2 syndrome" or "PTLS") or ("Bardet-Biedl syndrome" or "Bardet-Biedl*" or "Laurence-Moon-Bardet-Biedl syndrome" or "LMBBS") or ("2q37 deletion syndrome" or "2q37DS" or "2q37 microdeletion syndrome" or "Albright hereditary osteodystrophy-like syndrome" or "Brachydactyly-mental retardation syndrome" or "Chromosome 2q37 deletion syndrome" or "Deletion 2q37" or "Monosomy 2q37") or ("Mucopolysaccharidosis type III" or "MPS III" or "Mucopolysaccharidosis III" or "Sanfilippo* syndrome") or ("Cardiofaciocutaneous syndrome" or "Cardio-facio-cutaneous syndrome" or "CFC syndrome") or ("Smith-Lemli-Opitz syndrome" or "7-dehydrocholesterol reductase deficiency" or "RSH syndrome" or "SLO syndrome") or ("Mowat-Wilson* syndrome" or "Hirschsprung disease-mental retardation syndrome" or "Hirschsprung Disease – Intellectual Disability Syndrome")).ti,ab,tw. OR exp intellectual impairment/ OR exp mental deficiency/ OR exp developmental disorder/ OR exp Smith Magenis syndrome/ OR exp Prader Willi syndrome/ OR exp happy puppet syndrome/ OR exp fragile X syndrome/ OR exp cat cry syndrome/ OR exp Down syndrome/ OR exp Williams Beuren syndrome/ OR exp Lowe syndrome/ OR exp chromosome 5p/ OR exp chromosome deletion 5/ OR exp DiGeorge syndrome/ OR exp Rett syndrome/ OR exp syndrome CHARGE/ OR exp tuberous sclerosis/ OR exp Rubinstein syndrome/ OR exp Phelan-McDermid syndrome/ OR exp phenylketonuria/ OR exp Sotos syndrome/ OR exp Joubert syndrome/ OR exp Lesch Nyhan syndrome/ OR exp Bardet Biedl syndrome/ OR exp Sanfilippo syndrome/ OR exp Smith Lemli Opitz syndrome/

Measurement tools	("aberrant behavio?r checklist" or "aberrant behavio?r checklist-community" or "aberrant behavio?r checklist – community" or "ABC-C" or "aberrant behavio?r checklist-residential" or "aberrant behavio?r checklist – residential" or ("Aggression and Self-Injurious Behaviour Questionnaire" or "ASIQ") or ("autism spectrum disorder* behavio?r problem* for adult*" or "autism spectrum disorder* - behavio?r problem* for adult*" or "autism spectrum disorder* problem behavio?r for adult*" or "autism spectrum disorder* - problem behavio?r for adult*" or "ASD-BPA" or "ASD-PBA" or "autism spectrum disorder* behavio?r problem* for children" or "autism spectrum disorder* - behavio?r problem* for children" or "autism spectrum disorder* problem behavio?r for children" or "autism spectrum disorder* - problem behavio?r for children" or "ASD-BPC" or "ASD-PBC") or ("baby and infant screen for children with autism traits" or "BISCUIT") or ("child behavio?r checklist" or "CBCL 1.5-5" or "CBCL preschool" or "CBCL 6-18" or "TRF 6-18" or "CBCL TRF" or "teacher report form" or "youth self-report" or "youth self-report 11-18" or "YSR" or "adult behavio?r checklist" or "ABCL" or "achenbach checklist*") or ("children?s scale of hostility and aggression" or "children?s scale of hostility and aggression – reactive/proactive" or "C-SHARP" or "adult* scale of hostility and aggression" or "adult* scale of hostility and aggression – reactive/proactive" or "A-SHARP") or "aggressive behavio?r scale" or ("behavio?r problem* inventory-01" or "behavio?r problem* inventory" or "BPI-Short form" or "behavio?r problem* inventory – short form" or "BPI-S") or ("challenging behavio?r perception questionnaire" or "CHABA") or "challenging behavio?r interview" or ("challenging behavio?r questionnaire" or "CBQ") or "challenging behavio?r scale" or "checklist* of challenging behavio?r" or ("contextual assessment inventory for problem behavio?r" or "contextual assessment inventory") or ("developmental disabilities profile – maladaptive behavio?r domain" or "maladaptive behavio?r domain from the developmental disabilities profile" or "developmental disabilities profile") or ("diagnostic assessment for severely handicapped" or "DASH-II" or "DASH-2") or ("the early childhood behavio?r screen" or "ECBS") or "functional assessment for multiple causality" or ("overt aggression scale" or "Overt aggression scale modified for neurorehabilitation" or "OAS-MNR" or "modified overt aggression scale" or "MOAS" or "extended modified overt aggression scale" or "extended MOAS" or "institute for basic research modified overt aggression scale" or "IBR-MOAS") or "functional analysis checklist" or ("functional assessment screening tool" or "FAST") or ("great outcomes for kids impacted by severe developmental disabilities" or "GO4KIDDS") or ("learning disability needs assessment tool" or "LDNAT") or "motivation assessment scale" or ("initial behavio?r* assessment and protective equipment decision key" or "IBA-PEDK") or ("Nisonger child behavio?r rating form" or "NCBRF") or ("pervasive developmental disorder behavio?r inventory" or "PDDBI") or "problem behavio?r checklist" or ("psychopathology checklists for adults with intellectual disability*" or "P-AID") or ("questions about behavio?r* function" or "QABF") or "Sanfilippo behavio?r rating scale*" or ("scales of independent behavio?r revised" or "scales of independent behavio?r-revised" or "SIB-R") or ("self-injury, aggression and destruction screening questionnaire" or "SAD-SQ") or "self-injury trauma scale" or ("staff observation aggression scale" or "staff observation aggression scale – revised" or "SOAS" or "SOAS-R") or "individual schedule of the challenging behavio?r survey" or (interview adj2 "temper outburst*") or ("skin picking" adj10 "semi-structured interview")).ti,ab,tw.
Psychometric properties	("reliab*" or "valid*" or "coefficient" or "internal consistency" or "cronbach* alpha*" or "item correlation*" or "item selection*" or "item reduction*" or "agreement" or "precision" or "imprecision" or "precise values").mp,ti,ab,tw. OR ("test-retest" or ("test" and "retest") or ("reliab*" and ("test" or "retest")) or "stability" or "interrater" or "inter-rater" or "intrarater" or "intra-rater" or "intertester" or "inter-tester" or "intratester" or "intra-tester" or "interobserver" or "inter-observer" or "intraobserver" or "intra-observer" or "intertechnician" or "inter-technician" or "intratechnician" or "intra-technician" or "interexaminer" or "inter-examiner" or "intraexaminer" or "intra-examiner" or "interassay" or "inter-assay" or "intraassay" or "intra-assay" or "interindividual" or "inter-individual" or "intraindividual" or "intra-individual" or "interparticipant" or "inter-participant" or "intraparticipant" or "intra-participant").mp,ti,ab,tw. OR ("kappa" or "kappas" or "coefficient of variation" or "repeatab*" or (("replicab*" or "repeated") and ("measure" or "measures" or "findings" or "result" or "results" or "test" or "tests")) or "generaliza*" or "generalisab*" or "concordance" or ("intraclass" and "correlation*") or "discriminative" or "known group" or "factor analysis" or "factor analyses" or "factor structure" or "factor structures" or "dimensionality" or "subscale*" or "multitrait scaling analysis" or "multitrait scaling analyses" or "item discriminant" or "interscale correlation" or "interscale correlations" or (("error" or "errors") and ("measure*" or "correlat*" or "evaluat*" or "accuracy" or "accurate" or "precision" or "mean")) or "individual variability" or "interval variability" or "rate variability" or "variability analysis" or ("uncertainty" and ("measurement" or "measuring")) or "standard error of measurement" or "sensitiv*" or "responsiv*" or ("limit" and "detection") or "minimal detectable

concentration" or "interpretab*" or ("small*" and ("real" or "detectable") and ("change" or "difference")) or "meaningful change" or "minimal important change" or "minimal important difference" or "minimally important change" or "minimally important difference" or "minimal detectable change" or "minimal detectable difference" or "minimally detectable change" or "minimally detectable difference" or "minimal real change" or "minimal real difference" or "minimally real change" or "minimally real difference" or "ceiling effect" or "floor effect" or "item response model" or "irt" or "rasch" or "differential item functioning" or "dif" or "computer adaptive testing" or "item bank" or "cross-cultural equivalence").mp,ti,ab,tw. OR ("reproducib*" or "audit" or "psychometr*" or "clinimetr*" or "clinometr*").mp,ti,ab,tw. OR "observer variation".mp,ti,ab,tw. OR
exp intermethod comparison/ OR exp data collection method/ OR exp validation study/ OR exp feasibility study/ OR exp pilot study/ OR exp psychometry/ OR exp reproducibility/ OR exp observer variation/ OR exp discriminant analysis/ OR exp validity/ OR exp Outcome Assessment, Health Care/ OR exp internal consistency/ OR exp measurement precision/ OR exp measurement error/ OR exp measurement accuracy/ OR exp measurement/ OR exp intrarater reliability/ OR exp interrater reliability/ OR exp test retest reliability/ OR exp reliability/ OR exp factor analysis/ OR exp internal consistency/

Ovid Medline

Inclusion dates: All years (1946-May 5, 2023); Date of search: 05.05.2023

Intellectual disability

(intellectual* disab* or learning disab* or developmental* disab* or mental* retard* or mental* handicap* or ("Smith-Magenis syndrome" or "Smith Magenis" or "17p- syndrome" or "17p11.2 monosomy" or "Chromosome 17p deletion syndrome" or "Deletion 17p syndrome" or "partial monosomy 17p" or "del(17)(p11.2)") or ("Angelman*" or "Angelman* syndrome" or "Happy puppet syndrome" or "Happy puppet") or ("Prader-Willi syndrome" or "Prader-Labhart-Willi syndrome" or "Willi-Prader syndrome") or ("Fragile X" or "Fragile-X" or "Fragile X syndrome" or "FRAXA syndrome" or "AFRAX" or "Martin-Bell* syndrome" or "Marker X syndrome" or "fraX syndrome" or "fra(X) syndrome" or "X-linked mental retardation" or "Macroorchidism" or "Escalante* syndrome" or "Escalante*") or ("Cri-du-chat syndrome" or "Cat cry syndrome" or "5p deletion syndrome" or "5p- syndrome" or "chromosome 5p- syndrome" or "Monosomy 5p" or "chromosome 5p deletion syndrome") or ("Cornelia de Lange* syndrome" or "CDLS" or "De Lange* syndrome" or "Branchmann-De Lange* syndrome" or "BDLS" or "Brachmann* syndrome" or "Amstelodamensis typus degenerativus" or "Amsterdam dwarf syndrome" or "Amsterdam dwarfism" or "Typus degenerativus amstelodamensis") or ("Down* syndrome" or "Trisomy 21" or "Trisomy G" or "47,XX,+21" or "47,XY,+21") or ("Lowe syndrome" or "Cerebrooculorenal syndrome" or "Lowe oculocerebrorenal syndrome" or "Oculocerebrorenal syndrome" or "Oculocerebrorenal syndrome of Lowe" or "Phosphatidylinositol-4,5-bisphosphate-5-phosphatase deficiency") or ("7q11.23*" or "7q11.23 duplication syndrome" or "7q11.23 microduplication syndrome" or "chromosome 7q11.23 duplication" or "chromosome 7q11.23 duplication syndrome" or "dup(7)(q11.23)" or "Somerville-Van der Aa syndrome" or "trisomy 7q11.23" or "WBS duplication syndrome" or "Williams-Beuren region duplication syndrome" or "William* syndrome") or ("Velocardiofacial syndrome" or "Velo-cardio-facial syndrome" or "DiGeorge* syndrome" or "Conotruncal anomaly face syndrome" or "CATCH22" or "Autosomal dominant Opitz G/BBB syndrome" or "Autosomal dominant Opitz G BBB syndrome" or "Cayler cardiofacial syndrome" or "Deletion 22q11/2 syndrome" or "22q11/2 deletion syndrome" or "22q11/2DS" or "22q11 deletion syndrome" or "Sedlackova* syndrome" or "Shprintzen* syndrome") or ("Rett* syndrome" or "Rett* disorder" or "Cerebroatrophic hyperammonemia" or "Autism-dementiaataxia-loss of purposeful hand use syndrome") or ("CHARGE syndrome" or "CHARGE association" or "Hall-Hittner* syndrome" or "Hall* Hittner* syndrome" or "Coloboma") or ("Tuberous sclerosis" or "Tuberous sclerosis syndrome" or "Bourneville* disease" or "Bourneville* phakomatosis" or "Cerebral sclerosis" or "Cerebral sclerosis syndrome" or "Epiloia" or "Sclerosis tuberose" or "Tuberose sclerosis" or "Tuberose sclerosis syndrome" or "Tuberous sclerosis complex") or ("Rubinstein-Taybi* syndrome" or "RSTS" or "Broad thumb-hallux syndrome") or ("Phelan-McDermid* syndrome" or "Phelan-McDermid*" or "22q13 deletion syndrome" or "Deletion 22q13 syndrome" or "Deletion 22q13.3 syndrome" or "Monosomy 22q13" or "22q13.3 deletion syndrome") or "KBG syndrome" or ("Pitt-Hopkin*" or "Pitt-Hopkin* syndrome" or "PTHS") or ("48,XXYY syndrome" or "XXYY syndrome") or ("ADNP syndrome" or "ADNP-related intellectual disability and autism spectrum disorder" or "ADNP-related multiple congenital anomalies-intellectual disability-autism spectrum disorder"

or "Helsmoortel-van der Aa syndrome" or "HVDAS" or "Mental retardation, autosomal dominant 28" or "MRD28" or "ADNP-Related ID/ASD" or "ADNP-Related disorder") or ("SYNGAP1-related intellectual disability" or "SYNGAP-ID" or "SYNGAP1 syndrome" or "MRD5" or "SYNGAP1-related developmental and epileptic encephalopathy") or ("Phenylketonuria" or "Phenylalanine hydroxylase" or "Folling* disease" or "Folling* syndrome" or "PAH deficiency" or "PAH deficiency disease" or "Phenylalanine hydroxylase deficiency disease" or "Phenylalanine hydroxylase deficiency" or "PKU" or "Oligophrenia phenylpyruvica" or "Deficiency Disease, Phenylalanine Hydroxylase") or ("Sotos*" or "Sotos* syndrome" or "Cerebral gigantism" or "Sotos* sequence") or ("1p36 deletion syndrome" or "1p36DS" or "chromosome 1p36 deletion syndrome" or "distal monosomy 1p36" or "monosomy 1p36 syndrome") or ("8p23 deletion syndrome" or "8p23DS") or ("Joubert*" or "Joubert* syndrome" or "Joubert-Bolthauser* syndrome" or "JBTS" or "Cerebello-oculo-renal syndrome" or "Cerebello-oculo-renal syndrome 1" or "Cerebellooculorenal syndrome 1" or "Cerebellooculorenal syndrome" or "Cerebellar vermis agenesis" or "Cerebelloparenchymal disorder 4" or "Cerebelloparenchymal disorder" or "CPD4" or "Familial aplasia of the vermis") or ("16p11.2 deletion syndrome" or "16p11.2DS" or "AUTS14A") or ("Lesch-Nyhan syndrome" or "Choreoathetosis self-mutilation syndrome" or "Complete HPRT deficiency" or "Complete hypoxanthine-guanine phosphoribosyltransferase deficiency" or "HPRT deficiency" or "Deficiency of guanine phosphoribosyltransferase" or "Deficiency of hypoxanthine phosphoribosyltransferase" or "HGPRT deficiency" or "Hypoxanthine guanine phosphoribosyltransferase deficiency" or "Hypoxanthine phosphoribosyltransferase deficiency" or "Juvenile gout, choreoathetosis, mental retardation syndrome" or "Juvenile hyperuricemia syndrome" or "Lesch-Nyhan disease" or "Primary hyperuricemia syndrome" or "Total HPRT deficiency" or "Total hypoxanthine-guanine phosphoribosyl transferase deficiency" or "X-linked hyperuricemia" or "X-linked primary hyperuricemia" or "X-linked uric aciduria enzyme defect") or ("Cohen* syndrome" or "Norio* syndrome" or "Obesity-hypotonia syndrome" or "Pepper* syndrome" or "Prominent incisors-besityhypotonia syndrome" or "Hypotonia obesity and prominent incisors") or ("Dup15q syndrome" or "Duplication/inversion 15q11" or "Idic(15)" or "Inv dup(15)" or "Inverted duplication 15" or "Isodicentric chromosome 15" or "Isodicentric chromosome 15 syndrome" or "Non-distal tetrasomy 15q" or "15q duplication syndrome" or "15q11-q13 duplication syndrome") or ("Malan* syndrome" or "MALNS") or ("Marshall-smith* syndrome" or "MRSHSS") or ("Distal 18q deletion syndrome" or "18q deletion syndrome" or "18q- syndrome" or "Chromosome 18 long arm deletion syndrome" or "Chromosome 18q deletion syndrome" or "Chromosome 18q monosomy" or "Chromosome 18q-syndrome" or "Del(18q) syndrome" or "Monosomy 18q" or "proximal 18q deletion syndrome" or "tetrasomy 18p" or "18p isochromosome" or "18p tetrasomy") or ("Kleefstra syndrome" or "9q subtelomeric deletion syndrome" or "9q- syndrome" or "9q34.3 deletion syndrome" or "9q34.3 microdeletion syndrome" or "Chromosome 9q deletion syndrome" or "q34 deletion" or "9Qstds") or ("Potocki-Lupski syndrome" or "17p11.2 duplication syndrome" or "17p11.2 microduplication syndrome" or "chromosome 17p11.2 duplication syndrome" or "Dup(17)(p11.2p11.2)" or "Duplication 17p11.2 syndrome" or "PTLS") or ("Bardet-Biedl syndrome" or "Bardet-Biedl*" or "Laurence-Moon-Bardet-Biedl syndrome" or "LMBBS") or ("2q37 deletion syndrome" or "2q37DS" or "2q37 microdeletion syndrome" or "Albright hereditary osteodystrophy-like syndrome" or "Brachydactyly-mental retardation syndrome" or "Chromosome 2q37 deletion syndrome" or "Deletion 2q37" or "Monosomy 2q37") or ("Mucopolysaccharidosis type III" or "MPS III" or "Mucopolysaccharidosis III" or "Sanfilippo* syndrome") or ("Cardiofaciocutaneous syndrome" or "Cardio-facio-cutaneous syndrome" or "CFC syndrome") or ("Smith-Lemli-Opitz syndrome" or "7-dehydrocholesterol reductase deficiency" or "RSH syndrome" or "SLO syndrome") or ("Mowat-Wilson* syndrome" or "Hirschsprung disease-mental retardation syndrome" or "Hirschsprung Disease – Intellectual Disability Syndrome")).ti,ab,tw. or exp Intellectual Disability/ or exp Persons with Mental Disabilities/ or exp Developmental Disabilities/ or exp Learning Disabilities/ or exp Smith-Magenis Syndrome/ or exp Angelman Syndrome/ or exp Prader-Willi Syndrome/ or exp Fragile X Syndrome/ or exp Cri-du-Chat Syndrome/ or exp De Lange Syndrome/ or exp Down Syndrome/ or exp Oculocerebrorenal Syndrome/ or exp Williams Syndrome/ or exp DiGeorge Syndrome/ or exp Rett Syndrome/ or exp CHARGE Syndrome/ or exp Tuberous Sclerosis/ or exp Rubinstein-Taybi Syndrome/ or exp Klinefelter Syndrome/ or exp Phenylketonurias/ or exp Sotos Syndrome/ or exp Lesch-Nyhan Syndrome/ or exp Bardet-Biedl Syndrome/ or exp Mucopolysaccharidosis III/ or exp Smith-Lemli-Opitz Syndrome/

Measurement tools	("aberrant behavio?r checklist" or "aberrant behavio?r checklist-community" or "aberrant behavio?r checklist – community" or "ABC-C" or "aberrant behavio?r checklist-residential" or "aberrant behavio?r checklist – residential" or ("Aggression and Self-Injurious Behaviour Questionnaire" or "ASIQ") or ("autism spectrum disorder* behavio?r problem* for adult*" or "autism spectrum disorder* - behavio?r problem* for adult*" or "autism spectrum disorder* problem behavio?r for adult*" or "autism spectrum disorder* - problem behavio?r for adult*" or "ASD-BPA" or "ASD-PBA" or "autism spectrum disorder* behavio?r problem* for children" or "autism spectrum disorder* - behavio?r problem* for children" or "autism spectrum disorder* problem behavio?r for children" or "autism spectrum disorder* - problem behavio?r for children" or "ASD-BPC" or "ASD-PBC") or ("baby and infant screen for children with autism traits" or "BISCUIT") or ("child behavio?r checklist" or "CBCL 1.5-5" or "CBCL preschool" or "CBCL 6-18" or "TRF 6-18" or "CBCL TRF" or "teacher report form" or "youth self-report" or "youth self-report 11-18" or "YSR" or "adult behavio?r checklist" or "ABCL" or "achenbach checklist*") or ("children?s scale of hostility and aggression" or "children?s scale of hostility and aggression – reactive/proactive" or "C-SHARP" or "adult* scale of hostility and aggression" or "adult* scale of hostility and aggression – reactive/proactive" or "A-SHARP") or "aggressive behavio?r scale" or ("behavio?r problem* inventory-01" or "behavio?r problem* inventory" or "BPI-Short form" or "behavio?r problem* inventory – short form" or "BPI-S") or ("challenging behavio?r perception questionnaire" or "CHABA") or "challenging behavio?r interview" or ("challenging behavio?r questionnaire" or "CBQ") or "challenging behavio?r scale" or "checklist* of challenging behavio?r" or ("contextual assessment inventory for problem behavio?r" or "contextual assessment inventory") or ("developmental disabilities profile – maladaptive behavio?r domain" or "maladaptive behavio?r domain from the developmental disabilities profile" or "developmental disabilities profile") or ("diagnostic assessment for severely handicapped" or "DASH-II" or "DASH-2") or ("the early childhood behavio?r screen" or "ECBS") or "functional assessment for multiple causality" or ("overt aggression scale" or "Overt aggression scale modified for neurorehabilitation" or "OAS-MNR" or "modified overt aggression scale" or "MOAS" or "extended modified overt aggression scale" or "extended MOAS" or "institute for basic research modified overt aggression scale" or "IBR-MOAS") or "functional analysis checklist" or ("functional assessment screening tool" or "FAST") or ("great outcomes for kids impacted by severe developmental disabilities" or "GO4KIDDS") or ("learning disability needs assessment tool" or "LDNAT") or "motivation assessment scale" or ("initial behavio?r* assessment and protective equipment decision key" or "IBA-PEDK") or ("Nisonger child behavio?r rating form" or "NCBRF") or ("pervasive developmental disorder behavio?r inventory" or "PDDBI") or "problem behavio?r checklist" or ("psychopathology checklists for adults with intellectual disability*" or "P-AID") or ("questions about behavio?r* function" or "QABF") or "Sanfilippo behavio?r rating scale*" or ("scales of independent behavio?r revised" or "scales of independent behavio?r-revised" or "SIB-R") or ("self-injury, aggression and destruction screening questionnaire" or "SAD-SQ") or "self-injury trauma scale" or ("staff observation aggression scale" or "staff observation aggression scale – revised" or "SOAS" or "SOAS-R") or "individual schedule of the challenging behavio?r survey" or (interview adj2 "temper outburst*") or ("skin picking" adj10 "semi-structured interview")).ti,ab,tw.
Psychometric properties	("reliab*" or "valid*" or "coefficient" or "internal consistency" or "cronbach* alpha*" or "item correlation*" or "item selection*" or "item reduction*" or "agreement" or "precision" or "imprecision" or "precise values" or ("test-retest" or ("test" and "retest") or ("reliab*" and ("test" or "retest")) or "stability" or "interrater" or "inter-rater" or "intrarater" or "intra-rater" or "intertester" or "inter-tester" or "intratester" or "intra-tester" or "interobserver" or "inter-observer" or "intraobserver" or "intra-observer" or "intertechnician" or "inter-technician" or "intratechnician" or "intra-technician" or "interexaminer" or "inter-examiner" or "intraexaminer" or "intra-examiner" or "interassay" or "inter-assay" or "intraassay" or "intra-assay" or "interindividual" or "inter-individual" or "intraindividual" or "intra-individual" or "interparticipant" or "inter-participant" or "intraparticipant" or "intra-participant") or ((("kappa" or "kappas" or "coefficient of variation" or "repeatab*" or (("replicab*" or "repeated") and ("measure" or "measures" or "findings" or "result" or "results" or "test" or "tests")) or "generaliza*" or "generalisab*" or "concordance" or ("intraclass" and "correlation*") or "discriminative" or "known group" or "factor analysis" or "factor analyses" or "factor structure" or "factor structures" or "dimensionality" or "subscale*" or ("multitrait" and "scaling" and ("analysis" or "analyses")) or "item discriminant" or "interscale correlation" or "interscale correlations" or "error" or "errors") and ("measure*" or "correlat*" or "evaluat*" or "accuracy" or "accurate" or "precision" or "mean")) or "individual variability" or "interval variability" or "rate variability" or "variability analysis" or ("uncertainty" and ("measurement" or "measuring")) or "standard error of measurement" or "sensitiv*" or "responsiv*" or ("limit" and "detection") or "minimal detectable concentration" or "interpretab*" or ("small*" and ("real" or

	"detectable") and ("change" or "difference")) or "meaningful change" or "minimal important change" or "minimal important difference" or "minimally important change" or "minimally important difference" or "minimal detectable change" or "minimal detectable difference" or "minimally detectable change" or "minimally detectable difference" or "minimal real change" or "minimal real difference" or "minimally real change" or "minimally real difference" or "ceiling effect" or "floor effect" or "item response model" or "irt" or "rasch" or "differential item functioning" or "dif" or "computer adaptive testing" or "item bank" or "cross-cultural equivalence") or ("reproducib*" or "audit" or "psychometr*" or "clinimetr*" or "clinometr*") or "observer variation").ti,ab,tw. OR exp Psychometrics/ or exp Outcome Assessment, Health Care/ or exp Observer Variation/ or exp "Reproducibility of Results"/ or exp Discriminant Analysis/ or exp Data Collection/ or exp Validation Study/ or exp Feasibility Studies/ or exp Pilot Projects/ or exp Factor Analysis, Statistical/

Note. At the time of the main search, the Comprehensive Assessment of Triggers for Behaviours of Concern Scale (CATS; Limbo et al., 2021) was identified as newly published measure of function that was published after the preliminary search took place and terms for the CATS were not included in the main search strategy. However, forwards and backwards searches revealed no published evidence of IC, IRR and TRTR for this measure.

Limbu, B., Unwin, G., & Deb, S. (2021, Oct). Comprehensive Assessment of Triggers for Behaviours of Concern Scale (CATS): Initial Development. International Journal of Environmental Research and Public Health, 18(20), Article 10674. https://doi.org/10.3390/ijerph182010674.

Appendix 14: Chapter Three systematic process applied for inclusion of studies in the meta-analysis

Internal consistency

Several IC studies reported IC at multiple time points for the same participants (Rojahn et al., 2013b; Siegel et al., 2014), for these studies, IC values were extracted from the first timepoint. One study assessing the IC of the BPI-01 and NCBRF reported the sample size as a range, for this study, the median sample size was extracted (Rojahn et al., 2010).

Inter-rater reliability and test-retest reliability

Two IRR studies reported mean individual item percentage agreements between informants (Iwata et al., 1990; Iwata et al., 2013), one study of IRR and TRTR reported individual item kappa values (Matson & Rivet, 2008), and one IRR study reported Pearson correlation coefficients for individual items (Zarcone et al., 1991). For these studies, overall subscale values were calculated by taking the average of Fisher Z transformed Kappa values (Borenstein et al., 2009), or Pearson's coefficients, respectively.

Finally, one IRR study (Harris et al., 1994) reported the percentage agreement between informants, for this study, percentage agreement was converted into a Kappa coefficient using the following formula (Glen, 2014):

$$\kappa = \frac{Po - Pe}{1 - Pe} = 1 - \frac{1 - Po}{1 - Pe}$$

where:

P_o= the relative observed agreement among informants

P_e = the hypothetical probability of chance agreement

Appendix 14 References

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Appendix 15: Chapter Three summary of study characteristics for studies included in the meta-analytic syntheses

Appendices Table 7

Overall risk of bias ratings and study characteristics for each study assessing internal consistency, inter-rater reliability and test-retest reliability of measures of BtC

Measure	Authors (Paper number)	Risk of bias [a]			Measurement property						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
ABC	Aman et al. (1985) (5)	VG	VG-VG	VG	IC	Cronbach's alpha	927	100%	NR, NR, NR [c]	58.9%	Professional	NR	Healthcare setting
		D	A-D	A/D	IRR	Spearman correlation	25	100%	NR, NR, NR	NR	Professional-professional	NR	Healthcare setting
		D	VG-D	D	TRTR	Spearman correlation	184	100%	NR, NR, NR	NR	Professional	NR	Healthcare setting
	Aman et al. (1987a) (3)	VG	VG-VG	VG	IC	Cronbach's alpha	531	100%	402, 150, NR	61.4%	Professional	Paper	Community-based organisation
	Aman et al. (1987b) (4)	D	VG-D	VG	IRR	Pearson correlation	28	100%	NR, NR, NR [b]	NR	Professional-professional	NR	Healthcare setting
		D	VG-D	D	TRTR	Pearson correlation	28	100%	NR, NR, NR [b]	NR	Professional	NR	Healthcare setting
	Bihm & Poindexter (1991) (7)	VG	VG-VG	VG	IC	Cronbach's alpha	470	100%	325, 105, NR	53.0%	Professional	Paper	Community-based organisation
	Espie et al. (2003) (19)	VG	VG-VG	VG	IC	Cronbach's alpha	186	100%	NR, NR, NR	58.0%	Parent or caregiver	NR	Healthcare Setting
	Kaat et al. (2021) (32)	VG	VG-VG	VG	IC	Cronbach's alpha	120	100%	NR, NR, NR [b]	NR	Parent or caregiver	Online survey	Charity-organisation
	Newton & Sturmey (1988) (50)	VG	VG-VG	VG	IC	Cronbach's alpha	209	100%	NR, NR, NR	57.0%	Professional	Paper	Community-based organisation
	Rojahn & Heisel (1991) (59)	VG	VG-VG	VG	IC	Cronbach's alpha	199	76%	128, NR, 36-276	77.4%	Professional	Paper	Healthcare setting
		I	A-I	A/D	IRR	Pearson correlation	130	93%	NR, NR, NR [b]	NR [b]	Professional	Paper	Healthcare setting
	Rojahn et al. (2011) (63)	VG	VG-VG	VG	IC	Cronbach's alpha	254	100%	NR, NR, NR [b]	NR	Parent or caregiver, professional or educator	NR	Healthcare setting and community-based organisation
	Rojahn et al. (2013b) (66)	VG	VG-VG	VG	IC	Cronbach's alpha	97	NR	NR, NR, NR [b]	NR	Parent or caregiver	Interview	Charity-organisation
	Siegel et al. (2014) (71)	VG	VG-VG	VG	IC	Cronbach's alpha	38	55%	151, 35, 60-252	84.0%	Parent or caregiver	Paper or interview	Healthcare setting
	Sturmey & Bertman (1994) (77)	VG	VG-VG	VG	IC	Cronbach's alpha	27	100%	NR, NR, NR	NR	Professional	Paper	School

Appendices Table 7 Continued

Measure	Authors (Paper number)	Risk of bias [a]			Measurement property						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
ABC Continued	Sigafoos et al. (1997) (73)	D	VG-D	D	IRR	Spearman correlation	32	100%	51, NR, 20-72	75.0%	Parent or caregiver-Educator	NR	Community-based organisation
	Schroeder et al. (1997) (69)	I	VG-I	VG	IRR	Pearson correlation	30	100%	NR, NR, NR	NR	Professional-Professional	NR	Community-based organisation
		D	VG-D	VG/D	TRTR	Pearson correlation	30	100%	NR, NR, NR	NR	Professional-Professional	NR	Community-based organisation
ABC-C	Aman et al. (1995) (2)	VG	VG-VG	VG	IC	Cronbach's alpha	1024	100%	510, 170, 216-1068	58.9%	Professional	Paper	Community-based organisation
	Brown et al. (2002) (10)	VG	VG-VG	VG	IC	Cronbach's alpha	601	100%	158, NR, 72-264	56.0%	Parent or caregiver	Paper	School
	Freund & Reiss (1991) (24)	D	D-VG	VG	IC	Cronbach's alpha	110	NR	126, 60, 36-300	69.0%	Parent or caregiver	Paper	Healthcare Setting
		D	D-VG	VG	IC	Cronbach's alpha	94	NR	132, 60, 36-312	69.0%	Educator	Paper	Healthcare Setting
		D	VG-D	A/D	IRR	Pearson correlation	94	NR	132, 60, 36-312	69.0%	Parent or caregiver-Educator	Paper	Healthcare Setting
		D	A-D	D	TRTR	Pearson correlation	30	NR [b]	NR, NR, NR [b]	NR [b]	Parent or caregiver	Paper	Healthcare setting
		D	A-D	D	TRTR	Spearman correlation	25	NR [b]	NR, NR, NR [b]	NR [b]	Educator	Paper	Healthcare setting
	Marshburn & Aman (1992) (36)	D	D-VG	VG	IC	Cronbach's alpha	153	NR	NR, NR, NR [b]	NR	Educator	Paper	School
	Miller et al. (2003) (48)	VG	VG-VG	VG	IC	Cronbach's alpha	47	100%	NR, NR, NR [b]	NR	Educator	Paper	School
		D	VG-D	A	TRTR	Pearson correlation	48	100%	108, 21, 60-144	70.0%	Parent or caregiver	Paper	School
		D	VG-D	A	TRTR	Pearson correlation	22	100%	NR, NR, NR [b]	NR	Educator	Paper	School
	Hellings et al. (2005) (28)	I	VG-I	VG	IRR	Pearson correlation	8	100%	137, NR, 84-228	75.0%	Parent or caregiver-Educator	NR	Healthcare setting
A-SHARP	Matlock & Aman (2011) (38)	VG	VG-VG	VG	IC	Cronbach's alpha	512	100%	49, 148, 228-1008	61.7%	Professional	Paper	Community-based organisation
	Rojahn et al. (2017) (62)	VG	VG-VG	VG	IC	Cronbach's alpha	155	100%	NR, NR, 192-852	69.7%	Professional	NR	Community-based organisation
	Matlock & Aman (2014) (39)	D	VG-D	VG	IRR	ICC	39	100%	NR, NR, NR [b]	NR [b]	Parent or caregiver-Parent or caregiver	NR	Community-based organisation

Appendices Table 7 Continued

Measure	Authors (Paper number)	Risk of bias [a]			Measurement property						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
ASD-BPA	Matson & Rivet (2008) (40)	VG	VG-VG	VG	IC	Kuder-Richardson -20	171	100%	579, 138, 192-936	56.0%	Professional	Interview	Community-based organisation
		A	VG-A	VG	IRR	Kappa	171	100%	579, 138, 192-936	56.0%	Professional-Professional	Interview	Community-based organisation
		A	VG-A	VG	TRTR	Kappa	23	100%	NR, NR, NR [b]	NR [b]	Professional	Interview	Community-based organisation
BPI-01	Gonzalez et al. (2009) (25)	VG	VG-VG	VG	IC	Cronbach's alpha	425	100%	600, 163, 180-1044	55.3%	Professional	Interview	Community-based organisation
		D	VG-D	VG	IRR	Pearson correlation	100	100%	588, 164, 180-1032	63.0%	Professional-Professional	Interview	Community-based organisation
		I	VG-I	D	TRTR	Pearson correlation	80	100%	NR, NR, NR [b]	NR [b]	Professional	Interview	Community-based organisation
	Rojahn et al. (2013a) (60)	VG	VG-VG	VG	IC	Cronbach's alpha	179	100% [h]	202, 128, 18-737	42.2%	Parent or caregiver	Online survey	Charity-organisation
	Rojahn et al. (2001) (61)	D	D-VG	VG	IC	Cronbach's alpha	432	100%	NR, NR, 60-264	54.0%	Professional	Interview	Community-based organisation
		A	VG-A	VG	TRTR	ICC	247	100%	NR, NR, NR [b]	NR [b]	Professional	Interview	Community-based organisation
	Rojahn et al. (2010) (64)	VG	VG-VG	VG	IC	Cronbach's alpha	223-231 [i]	100%	NR, NR, NR [b]	NR	Educator	NR	School
		D	VG-D	A/VG	IRR	ICC	27	100%	NR, NR, NR [b]	NR [b]	Educator-Educator	NR	School
		D	VG-D	A/VG	IRR	ICC	63	100%	NR, NR, NR [b]	NR [b]	Educator-Professional	NR	School
		D	A-D	A	TRTR	ICC	24	100%	NR, NR, NR [b]	NR [b]	Educator	NR	School
	Rojahn et al. (2012a) (65)	VG	VG-VG	VG	IC	Cronbach's alpha	1122	100%	413, 244, 24-1080	57.8%	NR	NR	NR
	Rojahn et al. (2013b) (66)	VG	VG-VG	VG	IC	Cronbach's alpha	180	100% [j]	27, 10, 4-48	61.1%	Parent or caregiver	Interview	Charity-organisation
		I	VG-I	VG/I	TRTR	ICC	180	100% [j]	27, 10, 4-48	61.1%	Parent or caregiver	Interview	Charity-organisation
	van Ingen et al. (2010) (81)	VG	VG-VG	VG	IC	Cronbach's alpha	130	100%	476, 129, NR	70.8%	Professional	NR	Community-based organisation
		D	VG-D	VG/A	IRR	ICC	130	100%	476, 129, NR	70.8%	Professional-Professional	NR	Community-based organisation
		I	A-I	D	TRTR	ICC	130	100%	476, 129, NR	70.8%	Professional	NR	Community-based organisation
	Chan & Chien (2017) (12)	I	VG-I	VG/A	TRTR	ICC	42	100%	521, 131, NR	40.5%	Professional	NR	Healthcare setting

Appendices Table 7 Continued

Measure	Authors (Paper number)	Risk of bias [a]			Measurement property						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
BPI-Short form	Bowring et al. (2017) (9)	VG	VG-VG	VG	IC	Cronbach's alpha	265	100%	497, 195, NR	50.6%	Parent or caregiver	NR	Community-based organisation
	Mascitelli et al. (2015) (37)	D	VG-D	VG	IC	Cronbach's alpha	676	100%	438, 143, 192-852	67.7%	Professional	NR	Community-based organisation
		D	VG-D	VG/A	IRR	ICC	147	100%	NR, NR, NR [b]	69.6%	Professional-Professional	NR	Community-based organisation
		D	A-D	D	TRTR	Pearson correlation	147	100%	NR, NR, NR [b]	69.6%	Professional	NR	Community-based organisation
	Rojahn et al. (2012a) (65)	D	VG-D	VG	IC	Cronbach's alpha	1122	100%	413, 244, 24-1080	57.8%	NR	NR	NR
BISCUIT-Part 3	Matson et al. (2009a) (42)	VG	VG-VG	VG	IC	Cronbach's alpha	270	NR	27, 5, 17-37	72.2%	Parent or caregiver	Interview	Community-based organisation
	Matson et al. (2010) (43)	VG	VG-VG	VG	IC	Cronbach's alpha	644	NR	24, 5, 17-37	69.4%	Parent or caregiver	Interview	Community-based organisation
CBCL 1.5-5	Neo et al. (2021) (49)	VG	VG-VG	VG	IC	Cronbach's alpha	152	100% [g]	13, 10, 18-59	54.6%	Parent or caregiver	NR	Community-based organisation
	Pandolfi et al. (2009) (58)	D	D-VG	VG	IC	Cronbach's alpha	128	NR	42, 10, NR	89.0%	Parent or caregiver	Paper	Community-based organisation
CBCL 6-18	Esbensen et al. (2018) (18)	VG	VG-VG	VG	IC	Cronbach's alpha	88	100% [e]	136, 36, 72-216	61.4%	Parent or caregiver	Paper	Healthcare setting and charity-organisation
					IRR	ICC	88	100% [e]	136, 36, 72-216	61.4%	Parent or caregiver-Educator	Paper	Healthcare setting and charity-organisation
	Miller et al. (2003) (48)	VG	VG-VG	VG	IC	Cronbach's alpha	44	100%	NR, NR, NR [b]	NR	Parent or caregiver	Paper	School
		D	VG-D	A	TRTR	Pearson correlation	36	100%	NR, NR, NR [b]	NR	Parent or caregiver	Paper	School
CBCL-TRF	Miller et al. (2003) (48)	VG	VG-VG	VG	IC	Cronbach's alpha	47	100%	NR, NR, NR [b]	NR	Educator	Paper	School
		D	VG-D	A	TRTR	Pearson correlation	48	100%	108, 21, 60-144	70.0%	Parent or caregiver	Paper	School
		D	VG-D	A	TRTR	Pearson correlation	22	100%	NR, NR, NR [b]	NR [b]	Educator	Paper	School
CBI	Oliver et al. (2003) (54)	D	VG-D	VG	IRR	Pearson correlation	6-14 [l]	100%	NR, NR, NR [b]	NR [b]	Professional-Professional	Interview	Healthcare setting
		D	VG-D	A/D	TRTR	Pearson correlation	6-14 [l]	100%	NR, NR, NR [b]	NR [b]	Professional	Interview	Healthcare setting

Appendices Table 7 Continued

Measure	Authors (Paper number)	Risk of bias [a]			Measurement property						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
CCB	Harris et al. (1994) (27)	I	VG-I	VG	IRR	Kappa	4	100%	NR, NR, NR	NR	Professional-Professional	Interview	Community-based organisation
		I	D-I	D	TRTR	Spearman correlation	6	100%	NR, NR, NR	NR	Professional-Professional	Interview	Community-based organisation
C-SHARP	Farmer & Aman (2009) (20)	VG	VG-VG	VG	IC	Cronbach's alpha	372	100%	150, 44, 36-252	60.5%	Parent or caregiver	Paper	School
	Farmer & Aman (2010) (21)	I	VG-I	A	IRR	ICC	6-22 [k]	100%	NR, NR, NR [b]	NR [b]	Parent or caregiver-Parent or caregiver	Paper	School
	Farmer & Aman (2015) (22)	VG	VG-VG	VG	IC	Cronbach's alpha	8-12 [f]	53%	92, 54, NR	84.0%	Parent or caregiver	NR	Healthcare and research participant databases.
IBR-MOAS	Cohen et al. (2010) (14)	VG	VG-VG	VG	IC	Cronbach's alpha	3547	98%	592, 167, NR	60.0%	Professional	Paper	Healthcare setting
		VG	VG-VG	VG	IC	Cronbach's alpha	25	NR	458, 221, 72-780	56.0%	Professional, educator, or parent or caregiver	NR	Healthcare setting
		A	VG-A	VG	IRR	ICC	25	NR [b]	458, 221, 72-780	56.0%	Parent or caregiver-Professional	NR	Healthcare setting
		A	VG-A	VG	TRTR	ICC	16	NR [b]	NR, NR, NR [b]	NR [b]	Educator, Professional, or Parent or caregiver	NR	Healthcare setting
LDNAT	Painter et al. (2016) (57)	VG	VG-VG	VG	IC	Cronbach's alpha	1692	100%	500, NR, 216-1080	54.5%	Professional	NR	Healthcare setting
		I	A-I	D	TRTR	ICC	27	100%	NR, NR, NR [b]	NR [b]	Professional	NR	Healthcare setting
MOAS	Oliver et al. (2007) (55)	D	VG-D	VG/A	IRR	ICC	60	100%	420, NR, 276-696	64.3%	Parent or caregiver-Parent or caregiver	NR	Healthcare setting
NCBRF	Aman et al. (1996) (6)	VG	VG-VG	VG	IC	Cronbach's alpha	326	NR	81, 40, 36-192	65.0%	Parent or caregiver	Paper	Healthcare setting
		VG	VG-VG	VG	IC	Cronbach's alpha	260	NR	81, 41, 36-192	69.2%	Educator	Paper	Healthcare setting
		D	VG-D	D	IRR	Pearson correlation	189	NR	NR, NR, NR [b]	NR	Parent or caregiver-Educator	Paper	Healthcare setting

Appendices Table 7 Continued

Measure	Authors (Paper number)	Risk of bias [a]			Measurement property						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
NCBRF Continued	Norris & Lecavalier (2011) (53)	VG	VG-VG	VG	IC	Cronbach's alpha	399	100%	139, 46, 60-216	64.0%	Parent or caregiver	Paper	Healthcare setting and school
	Rojahn et al. (2010) (64)	VG	VG-VG	VG	IC	Cronbach's alpha	223-231 [i]	100%	NR, NR, NR [b]	NR	Educator	NR	School
		D	VG-D	A/VG	IRR	ICC	27	100%	NR, NR, NR [b]	NR [b]	Educator-Educator	NR	School
		D	VG-D	A/VG	IRR	ICC	63	100%	NR, NR, NR [b]	NR [b]	Educator-Professional	NR	School
		D	A-D	A	TRTR	ICC	24	100%	NR, NR, NR [b]	NR [b]	Educator	NR	School
OAS	Hellings et al. (2005) (28)	I	VG-I	D	IRR	Pearson correlation	8	100%	137, NR, 84-228	75.0%	Parent or caregiver-Educator	NR	Healthcare setting
PBCL	Tyrer et al. (2016) (79)	D	VG-D	VG/A/D	IRR	Kappa	38	NR	NR, NR, NR	NR	Professional-Professional	NR	Community-based organisation
PDDBI-Parent	Cohen et al. (2003) (13)	VG	VG-VG	VG	IC	Cronbach's alpha	311	NR [b]	NR, NR, NR [b]	NR [b]	Parent or caregiver	NR	School
		D	VG-D	D	IRR	ICC	270	NR [b]	NR, NR, NR [b]	NR [b]	Parent or caregiver-Educator	NR	School
PDDBI-Teacher	Cohen et al. (2003) (13)	VG	VG-VG	VG	IC	Cronbach's alpha	298	NR [b]	NR, NR, NR [b]	NR [b]	Educator	NR	School
		D	VG-D	A/D	IRR	ICC	49	NR [b]	NR, NR, NR [b]	NR [b]	Educator-Educator	NR	School
SIT	Iwata et al. (1990) (31)	I	VG-I	VG/I	IRR	Kappa	50	NR	NR, NR, 36-228	NR	Professional-Professional	Paper	NR
SOAS-ID-R	van den Bogaard et al. (2018) (80)	D	VG-D	VG	IRR	Pearson correlation	23	NR [b]	NR, NR, NR [b]	NR [b]	Professional-Professional	Paper	Healthcare setting

ABC=Aberrant behaviour checklist, ABC-C=Aberrant behaviour checklist-Community version, A-SHARP=Adult Scale of Hostility and Aggression, ASD-BPA=Autism Spectrum Disorder-Behavior Problems for Adults, BPI-01=Behavior Problems Inventory-01, BPI-Short Form=Behavior Problems Inventory-Short Form, BISCUIT-Part 3=Baby and Infant Screen for Children with aUtism Traits-Part 3, CBCL 1.5-5=Child Behavior Checklist 1.5-5, CBCL 6-18=Child Behavior Checklist 6-18, CBCL-TRF=Child Behavior Checklist-Teacher Report Form, CBI=Challenging Behaviour Interview, CCB=Checklist of Challenging Behaviour, C-SHARP=Children's Scale of Hostility and Aggression, IBR-MOAS=Institute for Basic Research-Modified Overt Aggression Scale, LDNAT=Learning Disability Needs Assessment Tool, MOAS=Modified Overt Aggression Scale, NCBRF=Nisonger Child Behavior Rating Form, OAS=Overt Aggression Scale, PBCL=Problem Behavior Checklist, PDDBI-Parent=Pervasive Developmental Disorder Behavior Inventory-Parent Version, PDDBI-Teacher=Pervasive Developmental Disorder Behavior Inventory-Teacher Version, SIT=Self-injury Trauma Scale, SOAS-ID-R=Staff Observation Aggression Scale – Revised, IC=Internal consistency, IRR=Inter-rater reliability, TRTR=Test-retest reliability, ID=Intellectual Disability, GS=Genetic Syndrome, NR=Not reported, ICC=Intra-class correlation, VG=Very good, A=Adequate, D=Doubtful, I=Inadequate.

Note. Paper numbers align with numbers of included papers listed in Appendix 21. [a] Internal consistency risk of bias ratings using the COSMIN risk of bias reliability criteria, inter-rater reliability and test-retest reliability ratings using the COSMIN risk of bias internal consistency criteria (Mokkink et al., 2018). VG=Very good, A=Adequate, D=Doubtful, I=Inadequate. A full

breakdown of risk of bias ratings for each study is available on request from the corresponding author. [b] Characteristics reported for overall sample but not for subset sample for IC/IRR/TRTR analysis. [c] Sample included mixture of ages but mean age and range were not reported. [d] Report ID as *primary* diagnosis in 37% of participants. [e] Participants with diagnosis of Downs syndrome. [f] Number of participants varied between C-SHARP subscales, n=12 for all problem scales, n=8 for all provocation scales. [g] Participants with genetic syndromes associated with intellectual disability. [h] Participants with diagnosis of Cornelia de Lange syndrome. [I] Number of participants reported as range between 223 and 231. [j] Participants at risk for intellectual and developmental disabilities (authors did not define at intellectual disability due to the low age range of participants). [k] Number of participants varied between C-SHARP subscales, n=22 for all problem scales, n=7 for the *verbal aggression provocation* scale, n=10 for the *bullying provocation* scale and n=6 for the *physical aggression provocation* scale. [l] Number of participants varied between CBI scales, n=14 for *physical aggression severity*, n=10 for *self-injury severity,* n=9 for *verbal aggression severity*, n=8 for *inappropriate vocalisation severity*, n=6 for *disruption of the environment severity*.

Appendices Table 8

Overall risk of bias ratings and study characteristics for each study assessing internal consistency, inter-rater reliability and test-retest reliability of measures of function

Measure	Authors (Paper number)	Risk of bias [a]			Reliability						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
CAI	McAtee et al. (2004) (46)	VG	VG-VG	VG	IC	Cronbach's alpha	20	100%	450, NR, 312-612	65.0%	Professional	Interview	Community-based organisation
		D	VG-D	A	IRR	ICC	20	100%	450, NR, 312-612	65.0%	Professional-Professional	Interview	Community-based organisation
		D	A-D	D	TRTR	Pearson correlation	20	100%	450, NR, 312-612	65.0%	Professional	Interview	Community-based organisation
FACT	Matson et al. (2003) Study 1 (44)	VG	VG-VG	VG	IC	KR-20	297	100%	567, 163, 108-1020	54.9%	Professional	Interview	Community-based organisation
	Matson et al. (2003) Study 2 (44)	VG	VG-VG	VG	IC	KR-20	197	100%	569, 160, 192-1020	56.9%	Professional	Interview	Community-based organisation
	Zaja et al. (2011) (82)	VG	VG-VG	VG	IC	Cronbach's alpha	130	100%	476, 128, 240-876	70.8%	Professional	NR	Community-based organisation
		D	VG-D	VG	IRR	ICC	130	100%	476, 128, 240-876	70.8%	Professional-Professional	NR	Community-based organisation
		I	VG-I	A	TRTR	ICC	130	100%	476, 128, 240-876	70.8%	Professional	NR	Community-based organisation
FAST	Zaja et al. (2011) (82)	VG	VG-VG	VG	IC	Cronbach's alpha	130	100%	476, 128, 240-876	70.8%	Professional	NR	Community-based organisation
		D	VG-D	VG	IRR	ICC	130	100%	476, 128, 240-876	70.8%	Professional-Professional	NR	Community-based organisation
		I	VG-I	A	TRTR	ICC	130	100%	476, 128, 240-876	70.8%	Professional	NR	Community-based organisation
	Iwata et al. (2013) (30)	I	VG-I	A	IRR	Kappa	196	NR	214, NR, 60-636	63.2%	Professional, Parent or caregiver, or Educator	NR	Healthcare setting and community-based organisation
MAS	Akande (1998) (1)	VG	VG-VG	VG	IC	Cronbach's alpha	102	100%	143, 20, 120-168	NR	Educator	NR	School
		D	VG-D	A/D	IRR	Pearson correlation	102	100%	143, 20, 120-168	NR	NR	NR	School
	Bihm et al. (1991) (8)	VG	VG-VG	VG	IC	Cronbach's alpha	118	100%	347, 109, NR	55.9%	Professional	Paper	Community-based organisation

Appendices Table 8 Continued

Measure	Authors (Paper number)	Risk of bias [a]			Reliability						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
MAS Continued	Duker & Sigafoos (1998) (16)	VG	VG-VG	VG	IC	Cronbach's alpha	86	100%	NR, NR, NR	NR	Professional, Parent or caregiver, or Educator	NR	Community-based organisation or School
		D	VG-D	D	IRR	Pearson correlation	86	100%	NR, NR, NR	NR	Parent or caregiver-Educator	NR	Community-based organisation
	Freeman et al. (2007) (23)	VG	VG-VG	VG	IC	Cronbach's alpha	83	100%	NR, NR, 23-216	NR [b]	Parent or caregiver	Paper	Healthcare setting
	Kearney et al. (2006) (34)	VG	VG-VG	VG	IC	Cronbach's alpha	335	100%	NR, NR, NR	NR	Professional	NR	Community-based organisation
	Koritsas & Iacono (2013) (35)	VG	VG-VG	VG	IC	Cronbach's alpha	70	100%	430, 156, 228-876	68.6%	Professional	Paper	Community-based organisation
		D	VG-D	VG	IRR	ICC	12	100%	NR, NR, NR [b]	NR [b]	Professional-Professional	Interview	Community-based organisation
	Newton & Sturmey (1991) (51)	VG	VG-VG	VG	IC	Cronbach's alpha	27	100%	NR, NR, NR	NR	Professional	NR	Community-based organisation
		D	VG-D	D	IRR	Pearson correlation	12	100%	NR, NR, NR	NR	Professional-Professional	NR	Community-based organisation
	Shogren & Rojahn (2003) (70)	VG	VG-VG	VG	IC	Cronbach's alpha	20	100%	NR, NR, 240-588	75.0%	Professional	Paper	Community-based organisation
		D	VG-D	VG/D	IRR	ICC	20	100%	NR, NR, 240-588	75.0%	Professional-Professional	Paper	Community-based organisation
		A	VG-A	VG	TRTR	ICC	20	100%	NR, NR, 240-588	75.0%	Professional	Paper	Community-based organisation
	Spreat & Connelly (1996) (75)	VG	VG-VG	VG	IC	Cronbach's alpha	47	100%	NR, NR, NR	51.1%	Educator or Professional	Paper	Community-based organisation
		D	VG-D	A/D	IRR	Pearson correlation	47	100%	NR, NR, NR	51.1%	Educator-Professional	NR	Community-based organisation
	Kearney et al. (1994) (33)	D	VG-D	D	IRR	Pearson correlation	42	100%	NR, NR, NR	64.3%	Professional-Professional	Paper	Community-based organisation
	Sigafoos et al. (1994) (72)	D	VG-D	VG/D	IRR	Pearson correlation	38	100%	312, NR, 168-480	83.3%	Professional-Professional	Paper	Community-based organisation
	Thompson & Emerson (1995) (78)	D	VG-D	A	IRR	ICC	42	100%	144, NR, 96-192	40.0%	NR	Paper	School

Appendices Table 8 Continued

Measure	Authors (Paper number)	Risk of bias [a]			Reliability						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
	Zarcone et al. (1991) (83)	D	VG-D	VG/D	IRR	Pearson correlation	39	100%	NR, NR, NR	NR	Professional-Professional	Interview	School
		D	VG-D	VG/D	IRR	Pearson correlation	16	100%	NR, NR, NR	NR	Educator-Educator	Paper	School
QABF	Freeman et al. (2007) (23)	VG	VG-VG	VG	IC	Cronbach's alpha	82	100%	NR, NR, 23-216	NR [b]	Parent or caregiver	Paper	Healthcare setting
	Koritsas & Iacono (2013) (35)	VG	VG-VG	VG	IC	Cronbach's alpha	70	100%	430, 156, 228-876	68.6%	Professional	Paper	Community-based organisation
		D	VG-D	VG	IRR	ICC	12	100%	NR, NR, NR [b]	NR [b]	Professional-Professional	Interview	Community-based organisation
	Nicholson et al. (2005) (52)	VG	VG-VG	VG	IC	Cronbach's alpha	118	100%	211, NR, 120-312	70.0%	Professional or Educator	Interview	School
		I	VG-I	D/I	IRR	Pearson correlation	118	100%	211, NR, 120-312	70.0%	Professional or Educator-Professional or Educator	Interview	School
	Rojahn et al. (2012b) (67)	VG	VG-VG	VG	IC	Cronbach's alpha	115	100%	477, 129, 240-876	70.4%	Professional	Paper	Community-based organisation
	Shogren & Rojahn (2003) (70)	VG	VG-VG	VG	IC	Cronbach's alpha	20	100%	NR, NR, 240-588	75.0%	Professional	Interview	Community-based organisation
		D	VG-D	VG/D	IRR	ICC	20	100%	NR, NR, 240-588	75.0%	Professional-Professional	Paper	Community-based organisation
		A	VG-A	VG	TRTR	ICC	20	100%	NR, NR, 240-588	75.0%	Professional	Paper	Community-based organisation
	Zaja et al. (2011) (82)	VG	VG-VG	VG	IC	Cronbach's alpha	130	100%	476, 128, 240-876	70.8%	Professional	NR	Community-based organisation
		D	VG-D	VG	IRR	ICC	130	100%	476, 128, 240-876	70.8%	Professional-Professional	NR	Community-based organisation
		I	VG-I	A	TRTR	ICC	130	100%	476, 128, 240-876	70.8%	Professional	NR	Community-based organisation
	Matson & Wilkins (2009) (41)	D	VG-D	D	IRR	Spearman correlation	64-80	100%	NR, NR, NR [b]	NR [b]	Professional-Professional	Interview	Community-based organisation
	Medeiros et al. (2013) (47)	D	VG-D	D	IRR	Pearson correlation	115	100%	362, 119, 204-720	70.0%	Professional-Professional	NR	Community-based organisation
		I	VG-I	I	TRTR	Pearson correlation	115	100%	362, 119, 204-720	70.0%	Professional	NR	Community-based organisation

Appendices Table 8 Continued

Measure	Authors (Paper number)	Risk of bias [a]			Reliability						Informant completing measure	Method of administration	Recruitment strategy
		Overall rating	Rating range	Mode rating	Type	Statistic	Participants in analysis (n)	% with ID or ID-associated GS	Mean age, SD, range (months)	% Male			
QABF-Short form	Singh et al. (2009) (74)	VG	VG-VG	VG	IC	Cronbach's alpha	75	100%	556, 150, 240-1020	49.3%	Professional	Interview	Community-based organisation
		D	VG-D	A/D	IRR	Pearson correlation	38	100%	NR, NR, NR [b]	NR [b]	Professional-Professional	NR	Community-based organisation
		D	VG-D	VG/A	TRTR	Pearson correlation	29	100%	NR, NR, NR [b]	NR [b]	Professional	NR	Community-based organisation

CAI=Contextual Assessment Inventory, FACT=Functional Assessment for Multiple CausaliTy, FAST=Functional Assessment Screening Tool, MAS=Motivation Assessment Scale, QABF=Questions About Behavioural Function Scale, QABF-Short Form=Questions About Behavioural Function-Short Form. IC=Internal consistency, IRR=Inter-rater reliability, TRTR=Test-retest reliability, ID=Intellectual Disability, GS=Genetic Syndrome, NR=Not reported, ICC=Intra-class correlation, KR-20=Kuder Richardson-20.

Note. Paper numbers align with numbers of included papers listed in Appendix 21. [a] Internal consistency risk of bias ratings using the COSMIN risk of bias reliability criteria, inter-rater reliability and test-retest reliability ratings using the COSMIN risk of bias internal consistency criteria (Mokkink et al., 2018). VG=Very good, A=Adequate, D=Doubtful, I=Inadequate. A full breakdown of risk of bias ratings for each study is available on request from the corresponding author. [b] Characteristics reported for overall sample but not for subset sample for IC/IRR/TRTR analysis.

Appendix 16: Chapter Three summary of study characteristics and reported reliability values for studies omitted from the meta-analytic syntheses

Appendices Table 9

Study characteristics and reported reliability values for studies omitted from quantitative analyses due to omission of information for individual subscales

Authors (paper number)	Measure	Reliability			Participants (n)	% with ID or ID-associated GS	Mean age (range) [d]	Informant completing measure	Method of administration	Recruitment strategy
		Type	Statistic	Reported value/s						
Chadwick et al. (2000) (11)	ABC-C	IRR	Pearson correlation	Range = 0.33-0.79 across subscales (stereotypies subscale = 0.33, all other subscale >0.70) [a]	139	82%	7.11 years (4.10-11.11 years)	Parent and schoolteacher or teaching assistant	Interview	UK special schools
Conroy et al. (1996) (15)	MAS	IRR	Pearson correlation	Range = -0.98-0.99 across subscales	20	100%	20.35 years (10-40 years)	Direct-care staff from different shifts	Interview	Group residential homes
Durand & Crimmins (1988) (17)	MAS	IRR	Pearson correlation	Range = 0.66-0.92 across subscales	50	100%	14.50 years (3.08-18.80 years)	Schoolteacher and assistant teacher	Paper	Schools
		TRTR	Pearson correlation	Range = 0.89-0.98 across subscales	50	100%	14.50 years (3.08-18.80 years)	Schoolteacher	Paper	Schools
Hustyi et al. (2013) (29)	FAST	IRR	Mean item-by-item % agreement	Mean agreement = 78.1%, range = 56.3-100%)	55	100% [b]	NR (6-25 years)	Mother and separate caregiver (e.g., Father)	Interview (telephone call)	Prader-Willi Syndrome groups and online parent support groups

Appendices Table 9 Continued

Authors (paper number)	Measure	Reliability			Participants (n)	% with ID or ID-associated GS	Mean age (range) [d]	Informant completing measure	Method of administration	Recruitment strategy
		Type	Statistic	Reported value/s						
Paclawskyj et al. (2000) (56)	QABF	IC	Cronbach's alpha	Range = 0.90-0.93 across subscales Overall alpha = 0.60	269	100%	NR (NR)	Direct-care staff	Interview	State developmental centre
		IRR	Pearson correlation	Range = 0.79-0.99 across subscales and the total score	57	100%	NR (NR)	Direct-care staff	Interview	State developmental centre
		TRTR	Pearson correlation	Range = 0.80-0.99 across subscales and the total score	34	100%	NR (NR)	Direct-care staff	Interview	State developmental centre
Sturmey (2001) (76)	FAC	IRR	Overall % agreement and Cohen's Kappa	Overall % agreement = 80 (range = 43-100%) Overall Kappa = 0.26 (range = 0.00-1.00)	30	100%	38 years (NR)	Direct-care staff	Paper	Residential facility
		TRTR	Overall % agreement and Cohen's Kappa	Overall % agreement = 87 (range = 63-100%) Overall Kappa = 0.53 (range = 0.00-1.00)	30	100%	38 years (NR)	Direct-care staff	Paper	Residential facility
Miller et al. (2003) (48)	ABC-C	IRR	Pearson correlation	Range = 0.72-0.80 across subscales [c]	48	100%	9 years (5-12 years)	Schoolteacher and teaching assistants	Paper	School special education classrooms
	CBCL-TRF	IRR	Pearson correlation	Range = 0.50-0.83 across subscales [c]	48	100%	9 years (5-12 years)	Schoolteacher and teaching assistants	Paper	School special education classrooms
Grey et al. (2010) (26)	BPI-01	IC	Cronbach's alpha	Overall alpha = 0.93 Subscale alphas's = 0.41 (self-injurious behaviour) and 0.86 aggression/destruction) [e]	159	100%	31.81 years (19-56 years)	Direct-care staff	Paper	Community-based services

Appendices Table 9 Continued

Authors (paper number)	Measure	Reliability			Participants (n)	% with ID or ID-associated GS	Mean age (range) [d]	Informant completing measure	Method of administration	Recruitment strategy
		Type	Statistic	Reported value/s						
Matson et al. (2009b) (45)	BISCUIT-Part 3	IC	Cronbach's alpha	Overall alpha = 0.91	276	100% [f]	26.83 months (17-37 months)	Primary caregiver	Interview	State funded program for children with developmental delay
Rose & Nelson (2016) (68)	BPI-01	IC	Cronbach's alpha	Self-injurious and aggressive/destructive behaviour subscale range = 0.64-0.85 [g]	46	100%	44.33 years (28-59 years)	Mothers	Interview or paper	Learning Disability Services and Schools

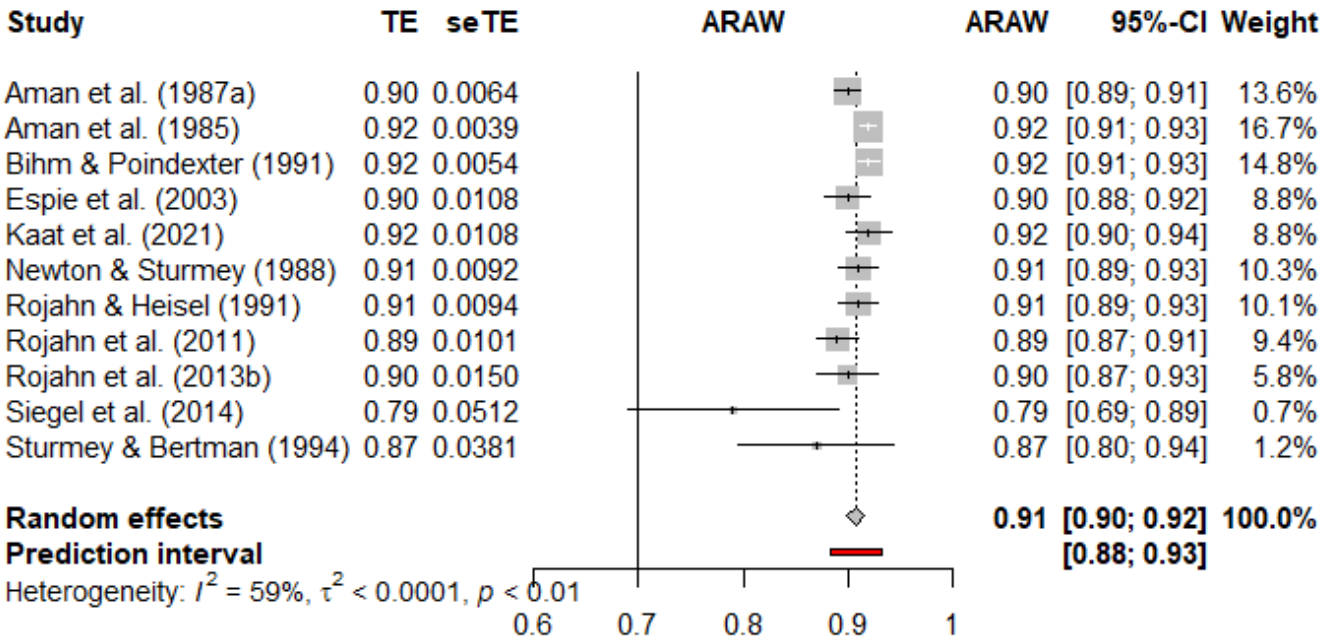
ABC-C=Aberrant Behaviour Checklist-Community Version, MAS=Motivation Assessment Scale, QABF=Questions About Behavioural Function Scale, FAC=Functional Analysis Checklist, CBCL-TRF=Child Behaviour Checklist-Teacher Report Form, BPI-01=Behaviour Problems Inventory-01, BISCUIT-Part 3= Baby and Infant Screen for Children with aUtism Traits-Part 3, IC=Internal consistency, IRR=Inter-rater reliability, TRTR=Test-retest reliability, ID=Intellectual Disability, GS=Genetic Syndrome, NR=Not reported.

Note. Paper numbers align with numbers of included papers listed in Appendix 21.

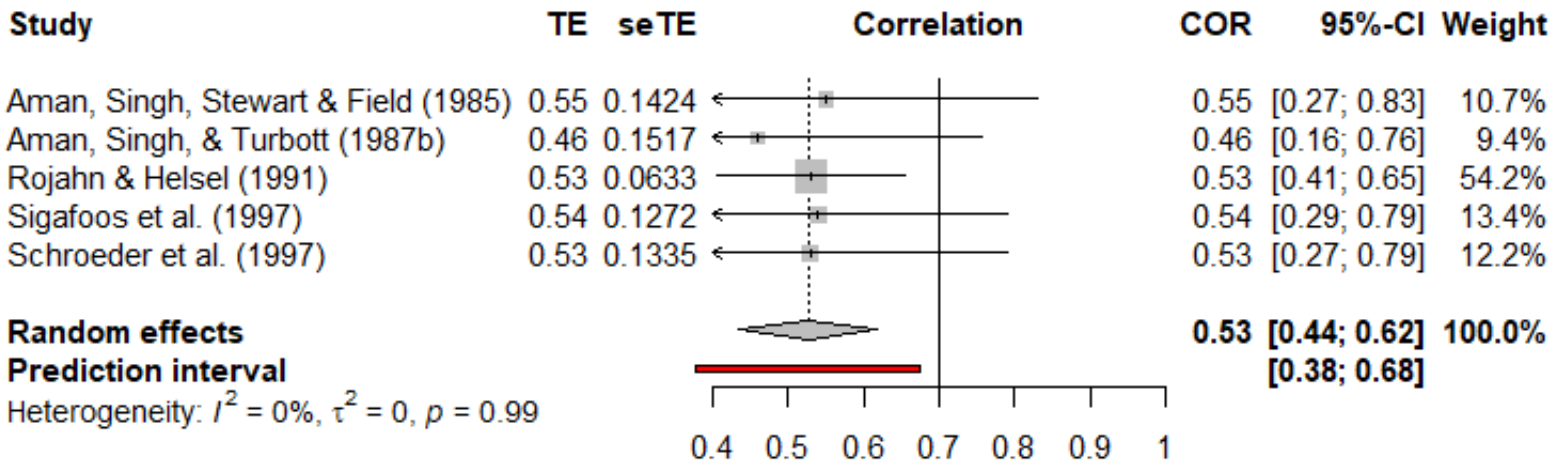
[a] IRR obtained from 65 (46.7%) of participants.
[b] All participants held a diagnosis of Prader-Willi Syndrome.
[c] IRR obtained from 22 (45.83%) of participants.
[d] Mean age and ranges are reported from overall study samples.
[e] Unclear whether authors used the frequency or severity self-injurious behaviour and aggressive/destructive behaviour subscales.
[f] Participants with developmental delay.
[g] Authors summed the severity and frequency subscale scores for each behaviour, producing a total self-injurious behaviour score and total aggressive/destructive behaviour score.

Appendix 17: Chapter Three forest plots of IC, IRR and TRTR per measure of behaviours that challenge

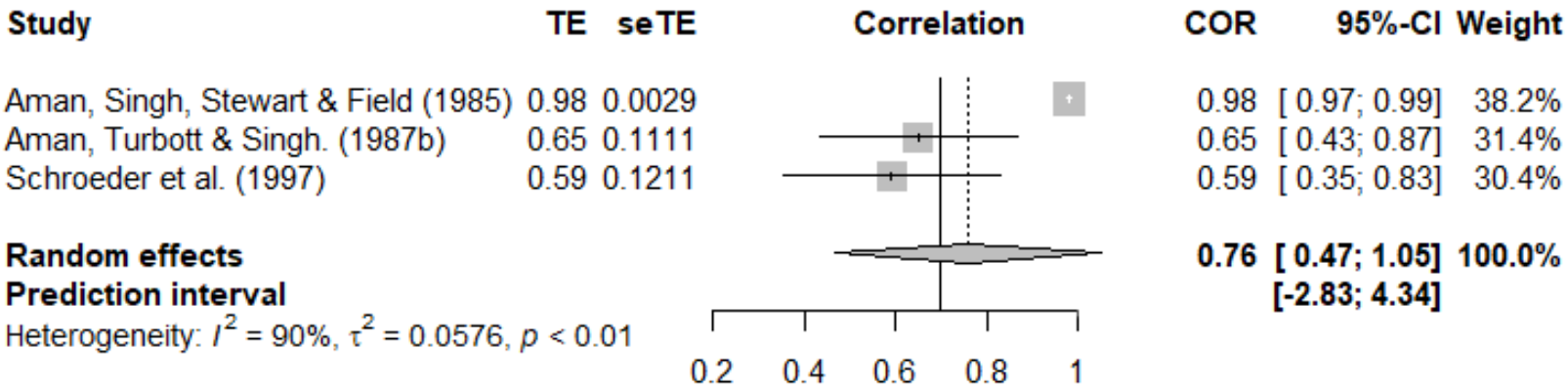
Aberrant Behaviour Checklist (ABC) *irritability* subscale



Appendices Figure 1. Forest plot for the internal consistency of the ABC *irritability* subscale using a random-effects model.

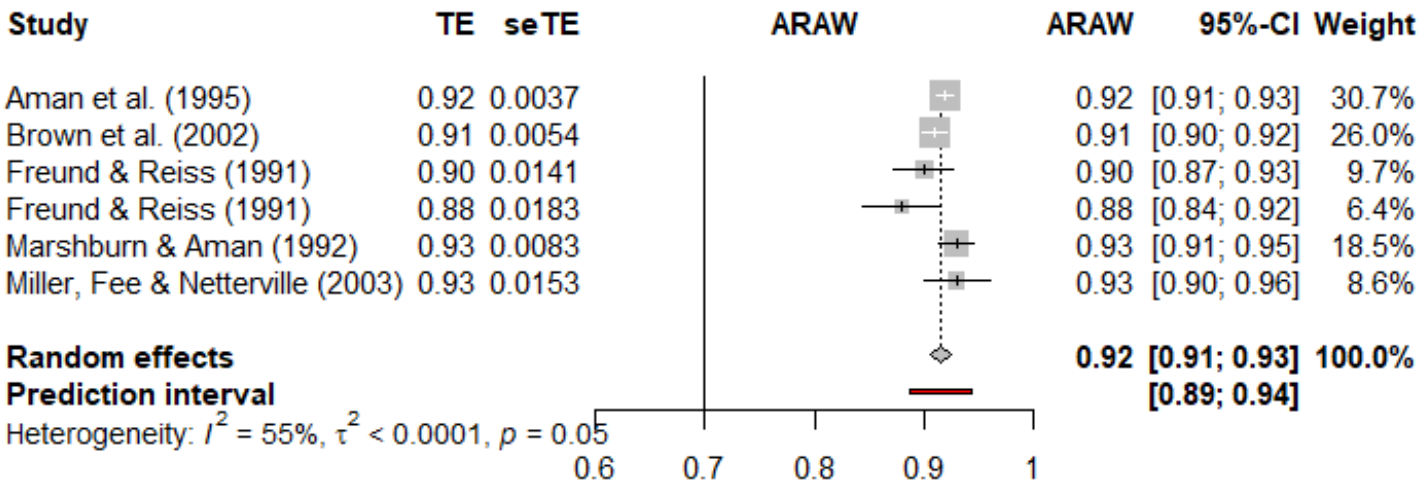


Appendices Figure 2. Forest plot for the inter-rater reliability of the ABC *irritability* subscale using a random-effects model.

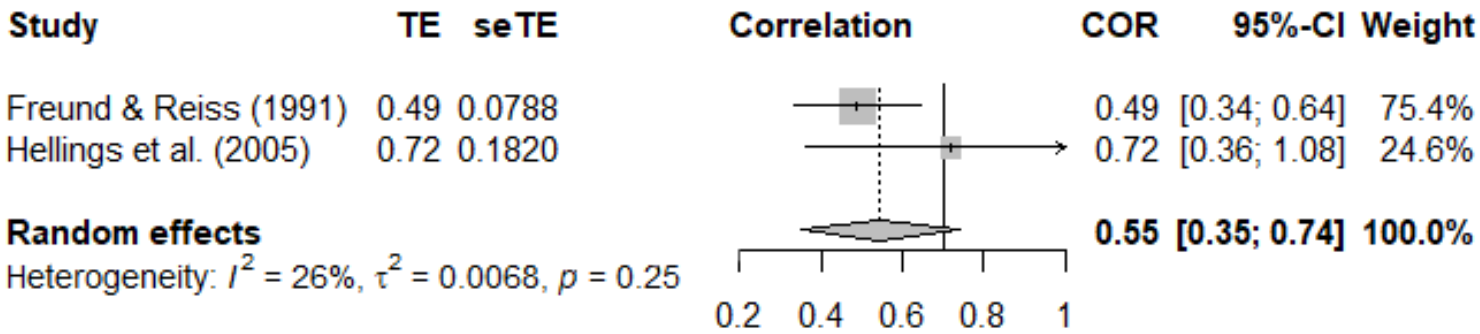


Appendices Figure 3. Forest plot for the test-retest reliability of the ABC *irritability* subscale using a random-effects model.

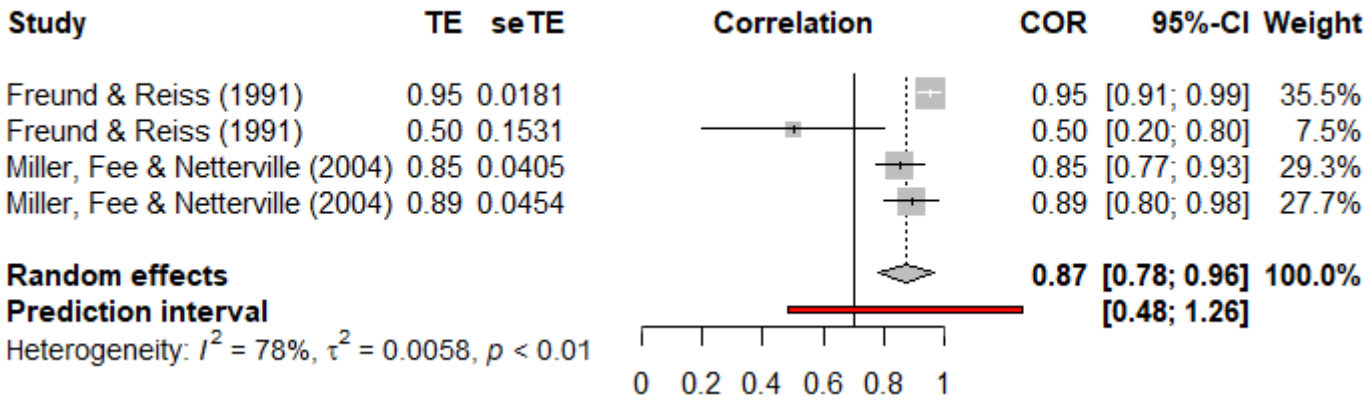
Aberrant Behaviour Checklist – Community (ABC-C) *irritability* subscale



Appendices Figure 4. Forest plot for the internal consistency of the ABC-C *irritability* subscale using a random-effects model.

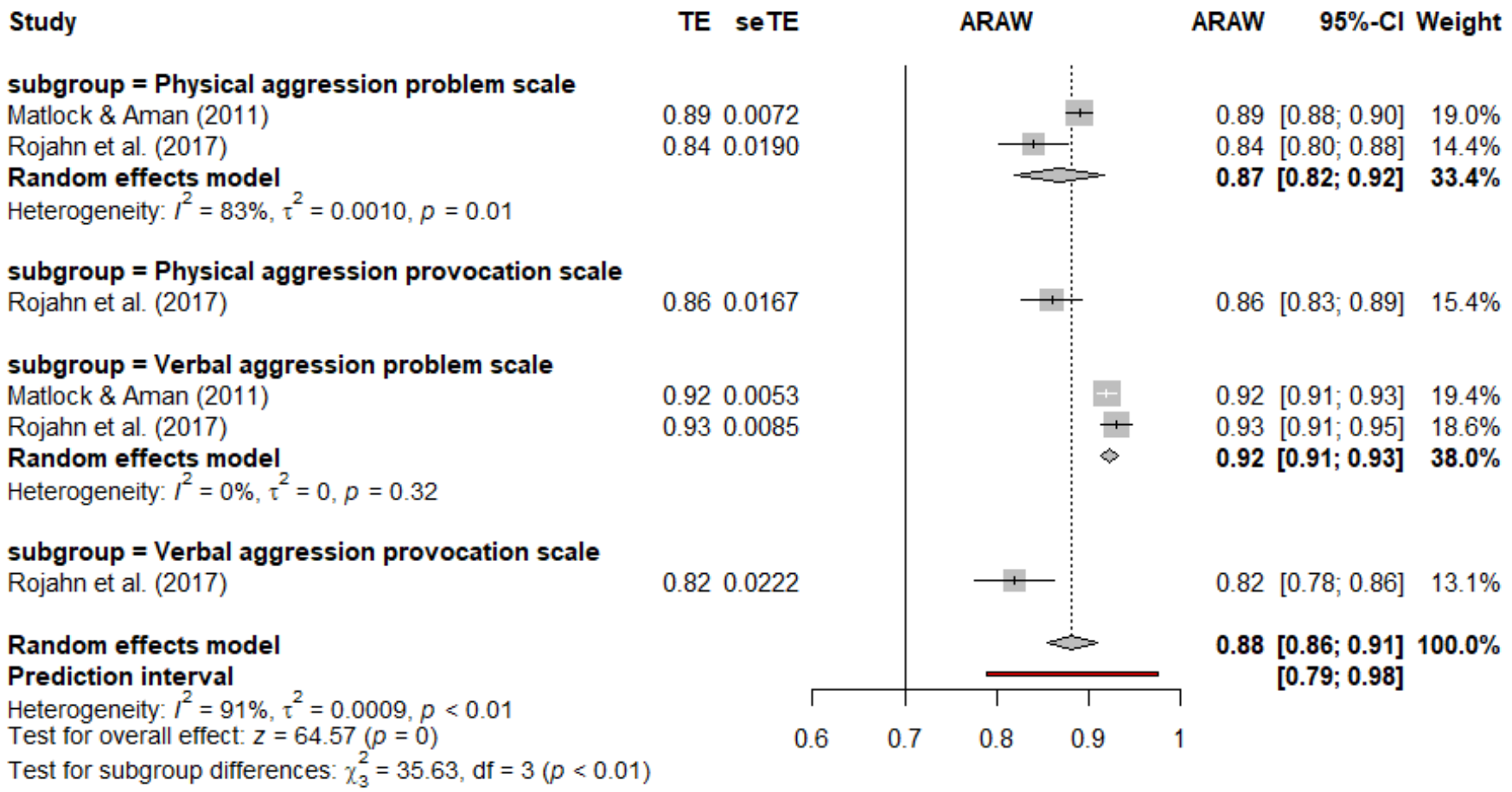


Appendices Figure 5. Forest plot for the inter-rater reliability of the ABC-C *irritability* subscale using a random-effects model.

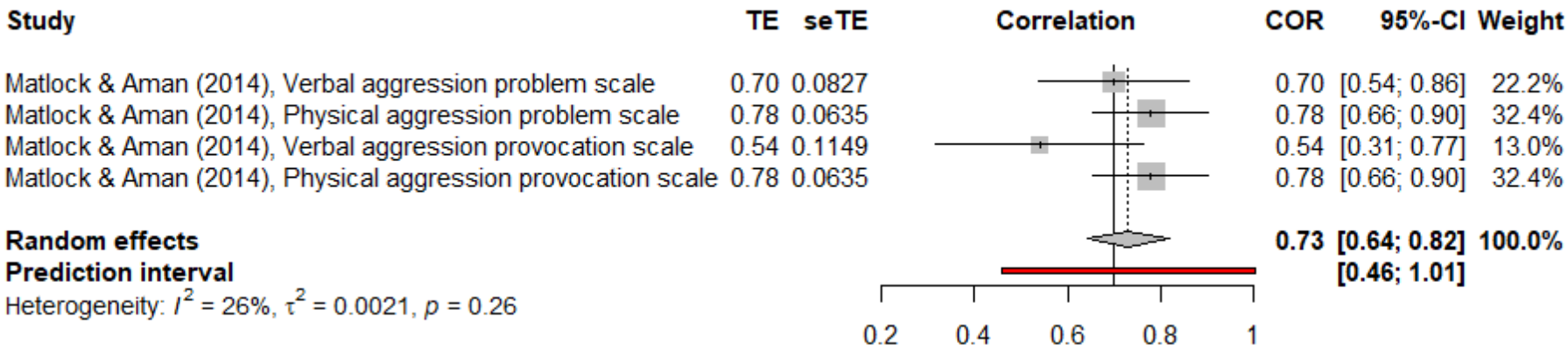


Appendices Figure 6. Forest plot for the test-retest reliability of the ABC-C *irritability* subscale using a random-effects model.

Adult Scale of Hostility and Aggression (A-SHARP)

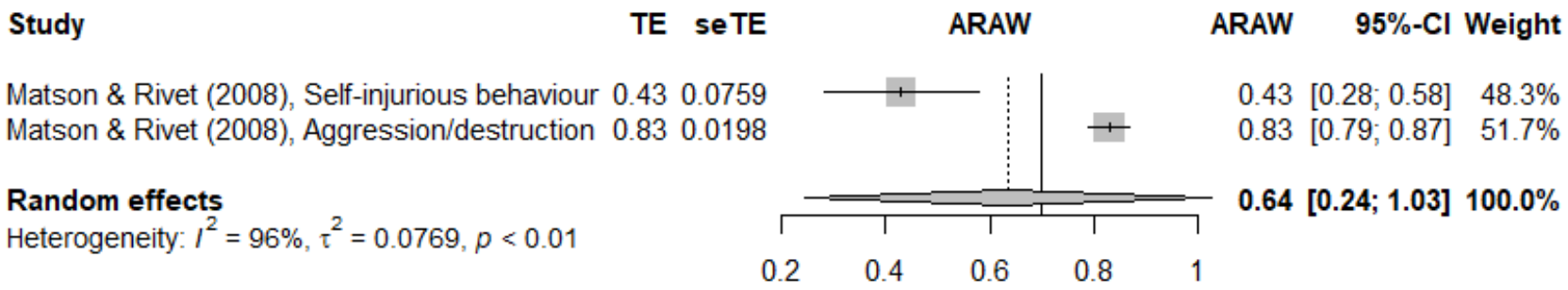


Appendices Figure 7. Forest plot for the internal consistency of the A-SHARP using a random-effects model.

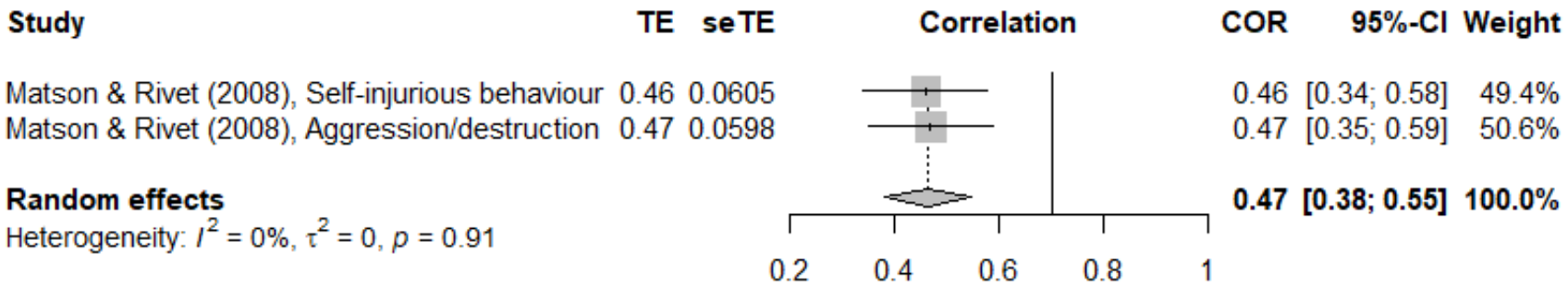


Appendices Figure 8. Forest plot for the inter-rater reliability of the A-SHARP using a random-effects model.

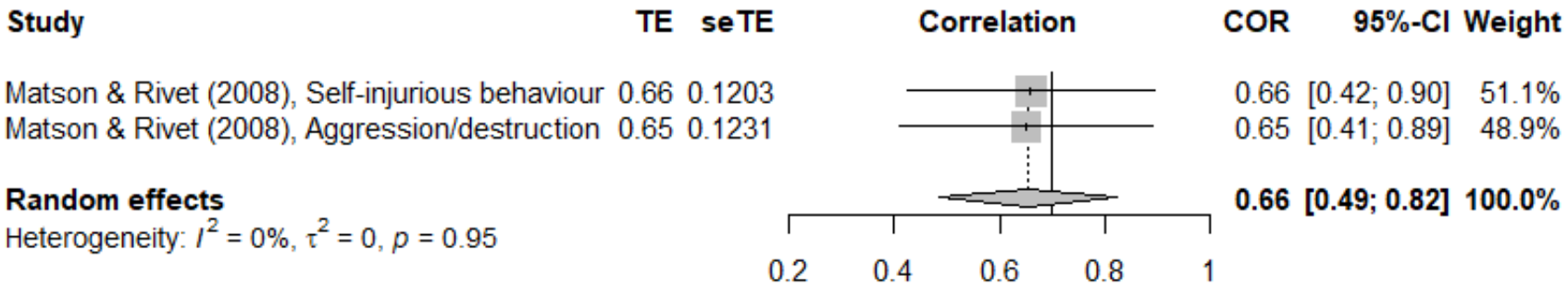
Autism Spectrum Disorder – Behavior Problems for Adults (ASD-BPA)



Appendices Figure 9. Forest plot for the internal consistency of the ASD-BPA using a random-effects model.

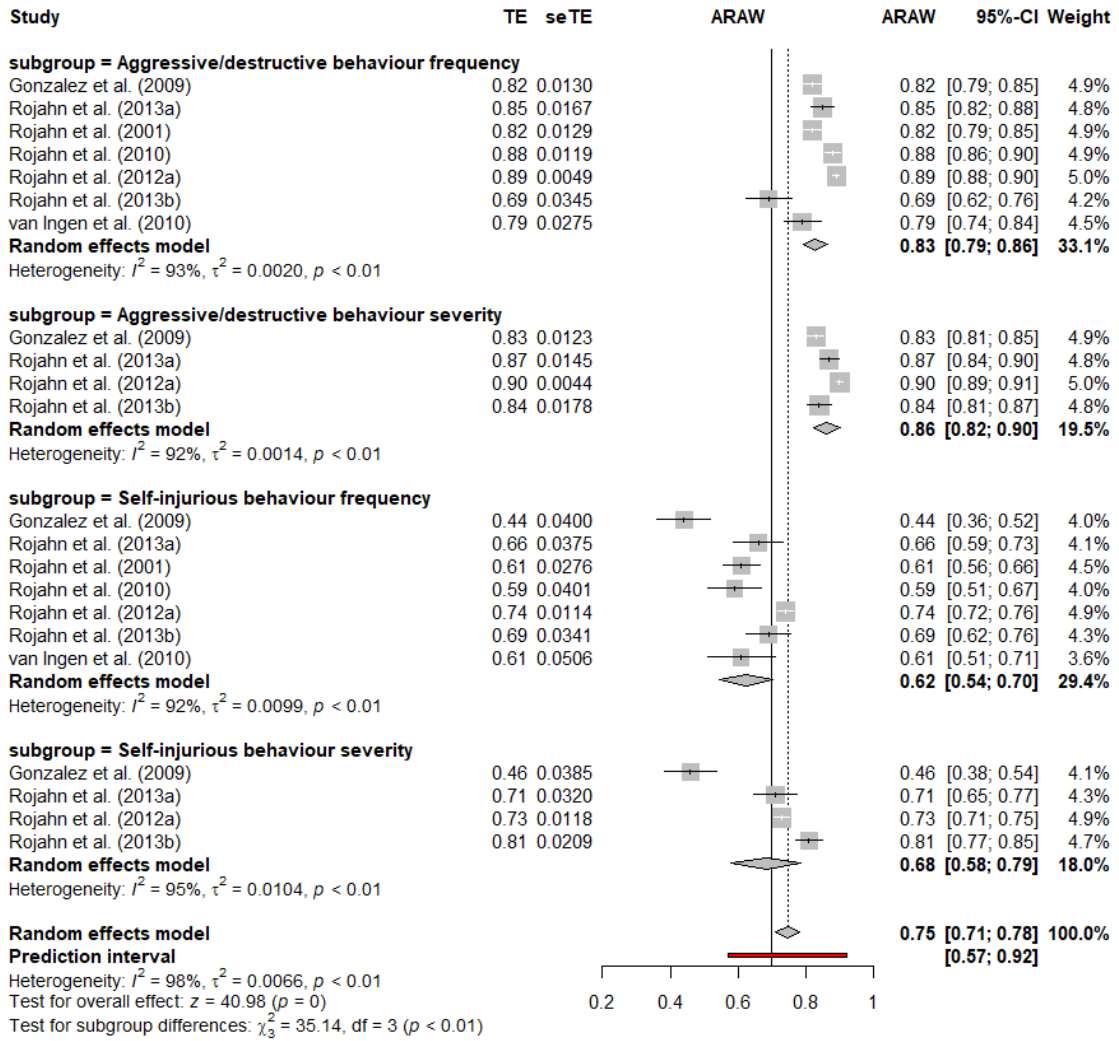


Appendices Figure 10. Forest plot for the inter-rater reliability of the ASD-BPA using a random-effects model.

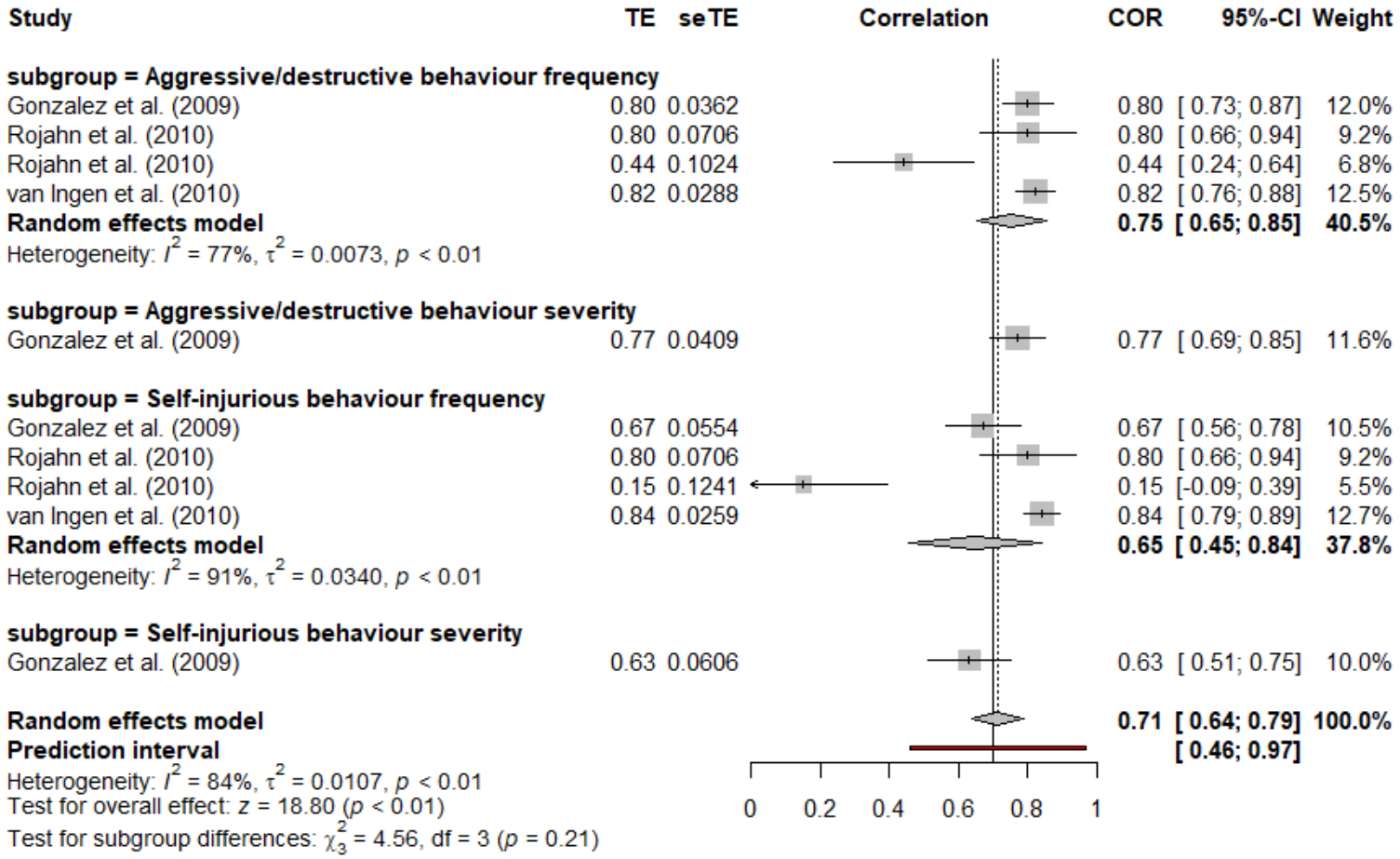


Appendices Figure 11. Forest plot for the test-retest reliability of the ASD-BPA using a random-effects model.

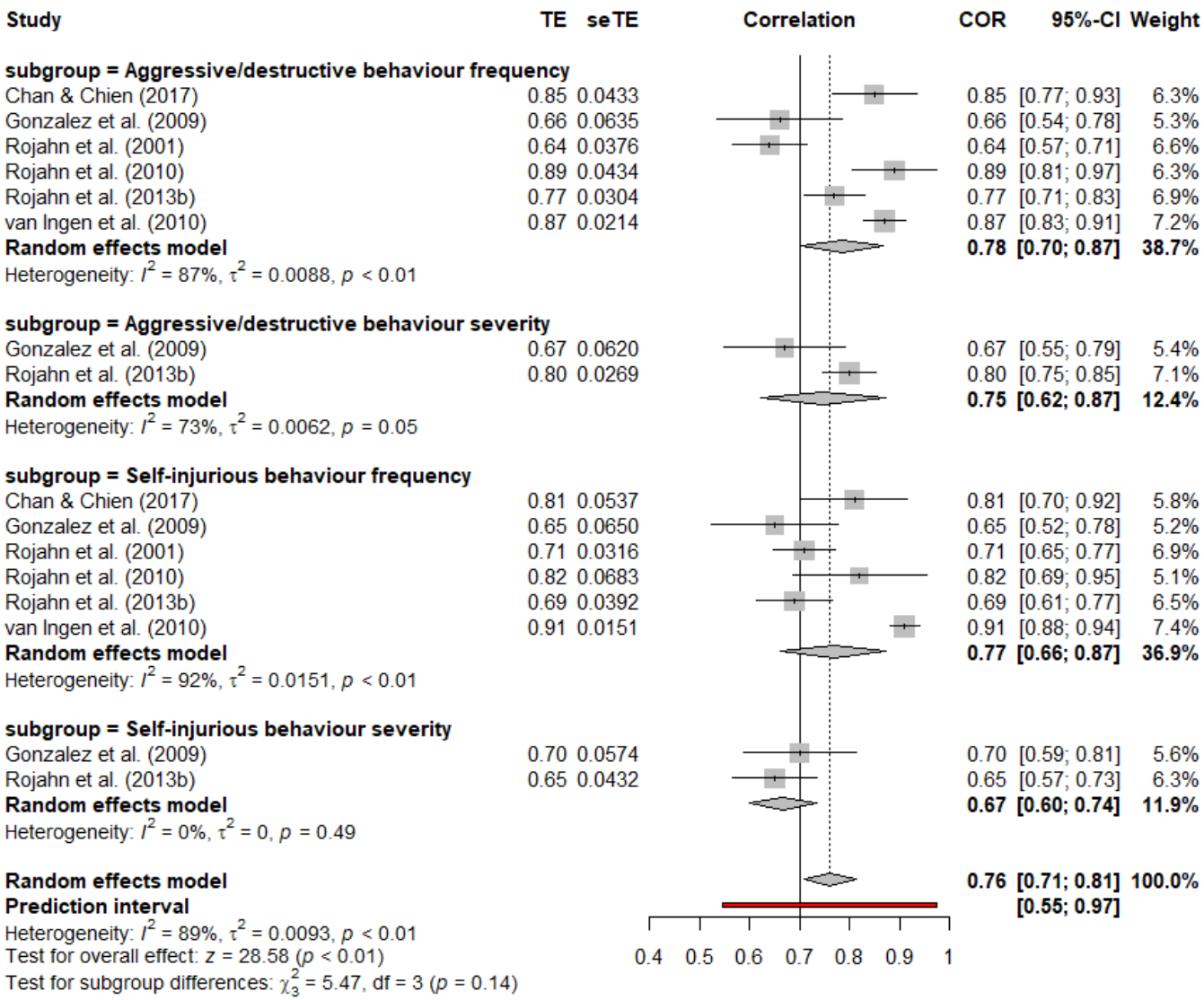
Behavior Problems Inventory – 01 (BPI-01)



Appendices Figure 12. Forest plot for the internal consistency of the BPI-01 using a random-effects model.

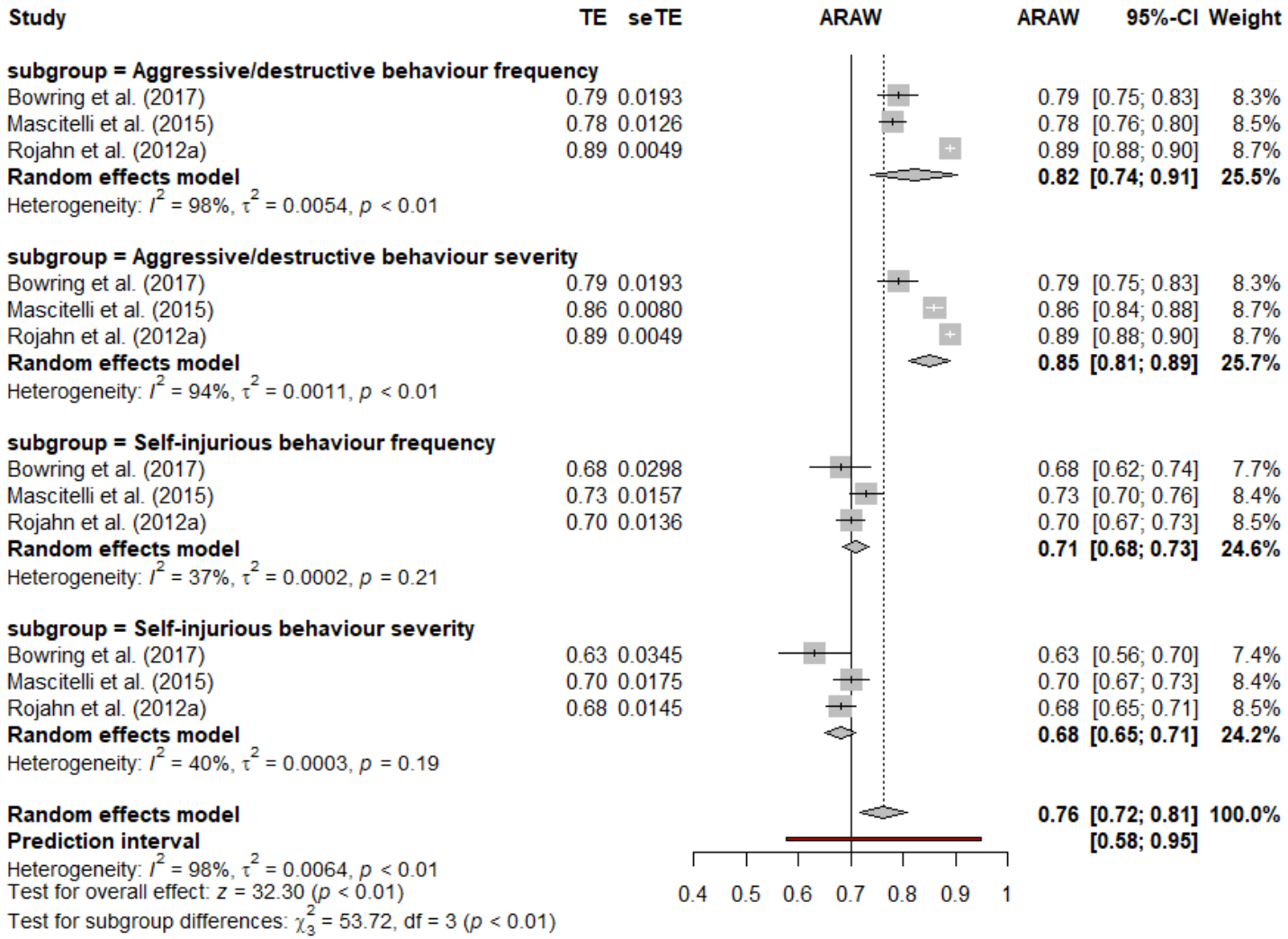


Appendices Figure 13. Forest plot for the inter-rater reliability of the BPI-01 using a random-effects model.

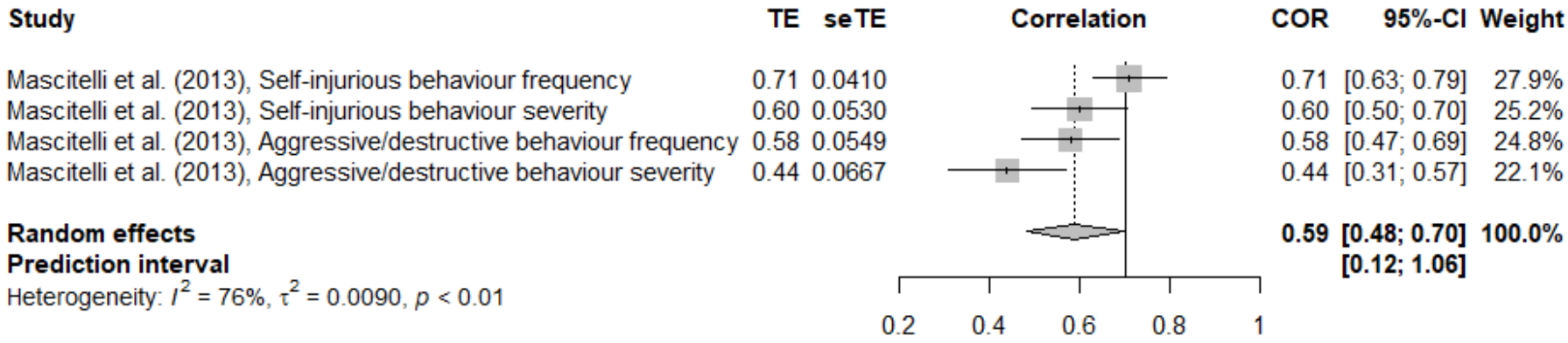


Appendices Figure 14. Forest plot for the test-retest reliability of the BPI-01 using a random-effects model.

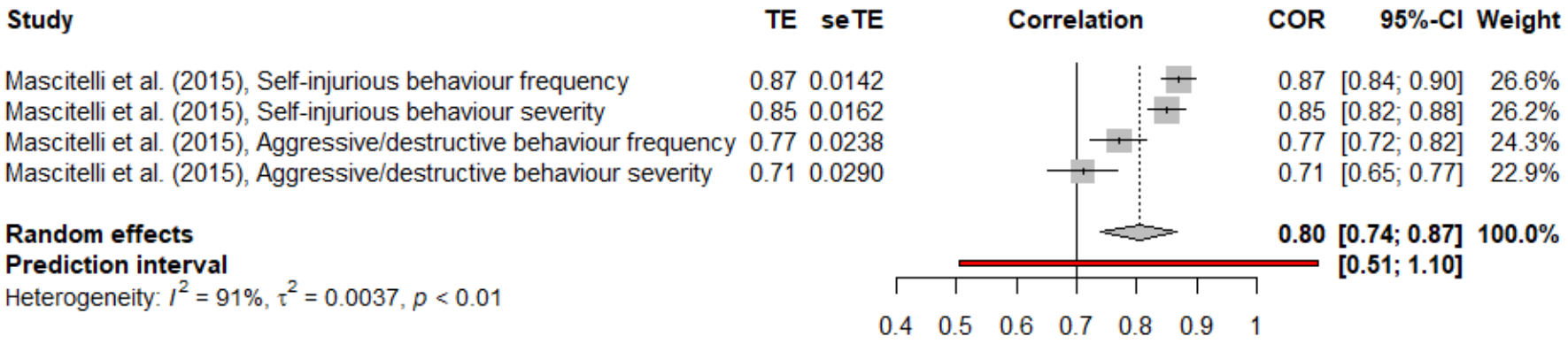
Behavior Problems Inventory – Short Form (BPI-Short Form)



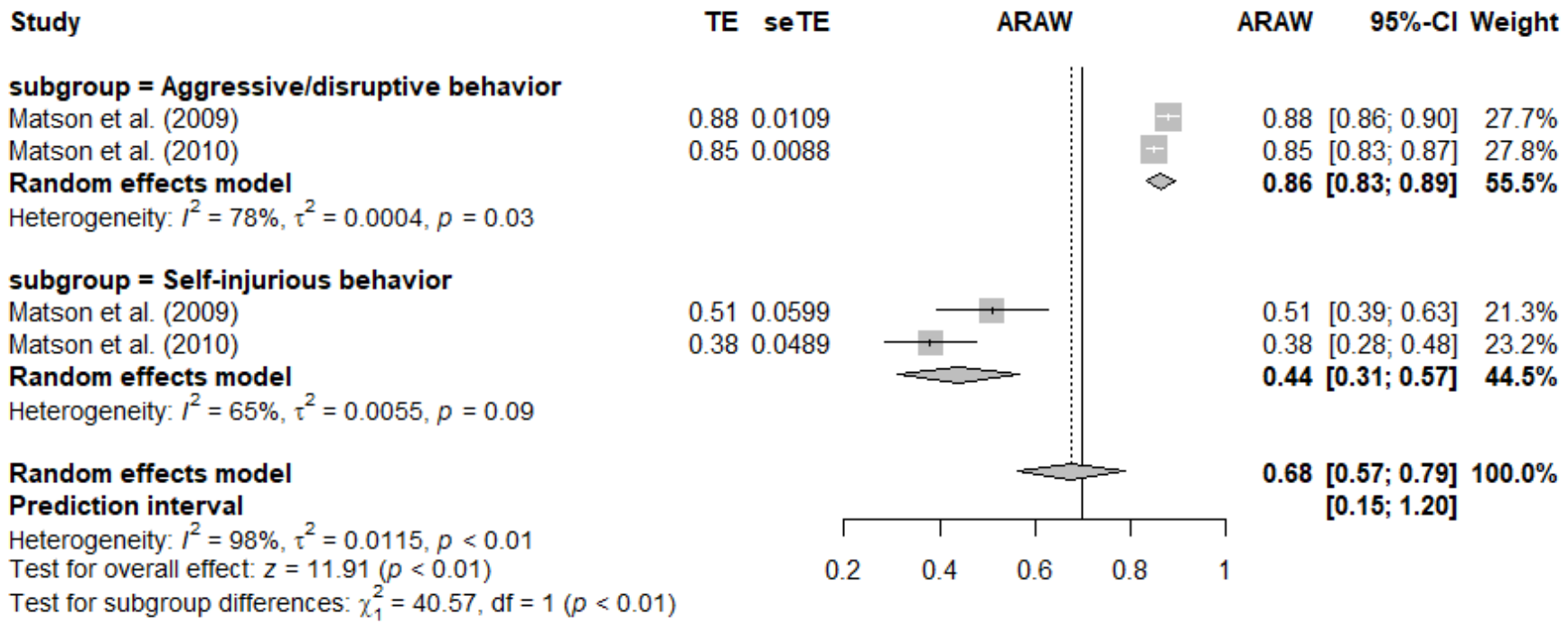
Appendices Figure 15. Forest plot for the internal consistency of the BPI-Short Form using a random-effects model.



Appendices Figure 16. Forest plot for the inter-rater reliability of the BPI-Short Form using a random-effects model.

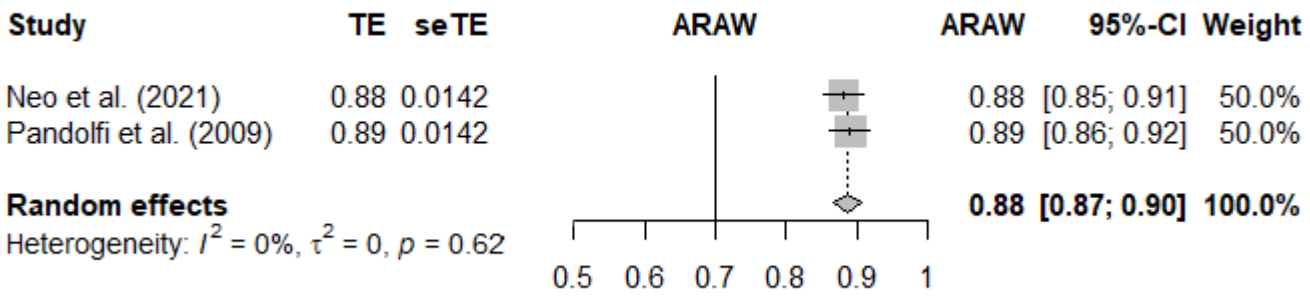


Appendices Figure 17. Forest plot for the test-retest reliability of the BPI-Short Form using a random-effects model.



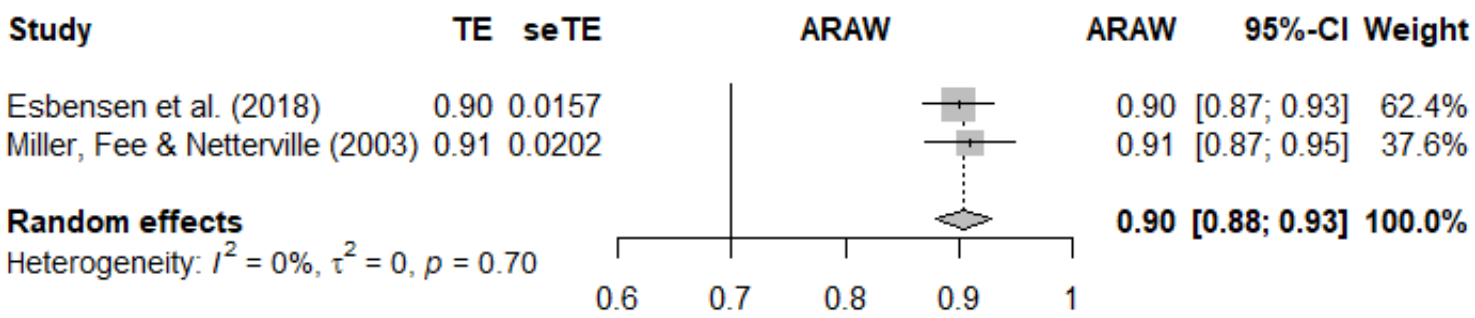
Appendices Figure 18. Forest plot for the internal consistency of the BISCUITT-Part 3 using a random-effects model.

Child Behavior Checklist 1.5-5 (CBCL 1.5-5) *aggressive behaviour* scale

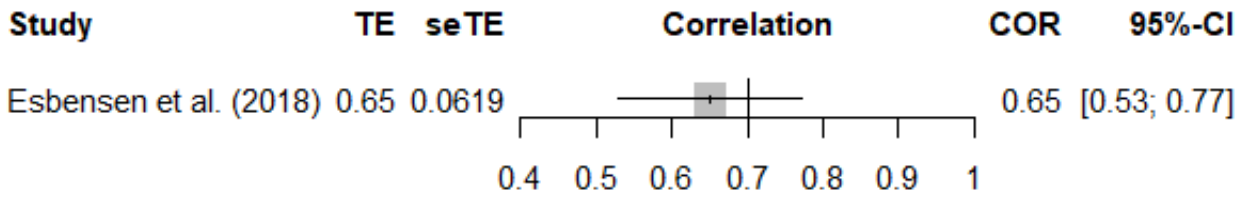


Appendices Figure 19. Forest plot for the internal consistency of the CBCL 1.5-5 *aggressive behaviour* scale using a random-effects model.

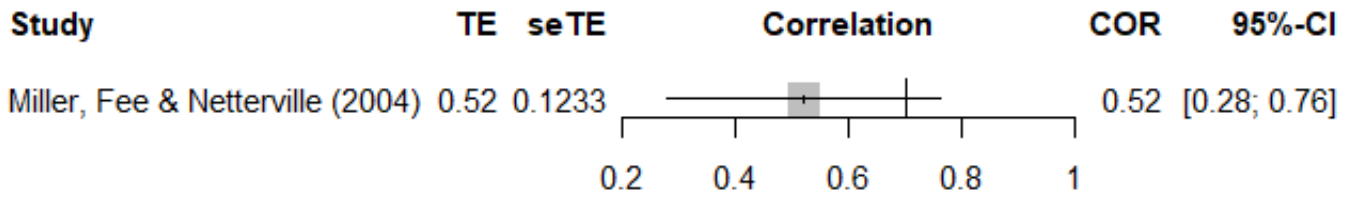
Child Behavior Checklist 6-18 (CBCL 6-18) *aggressive behaviour* scale



Appendices Figure 20. Forest plot for the internal consistency of the CBCL 6-18 *aggressive behaviour* scale using a random-effects model.

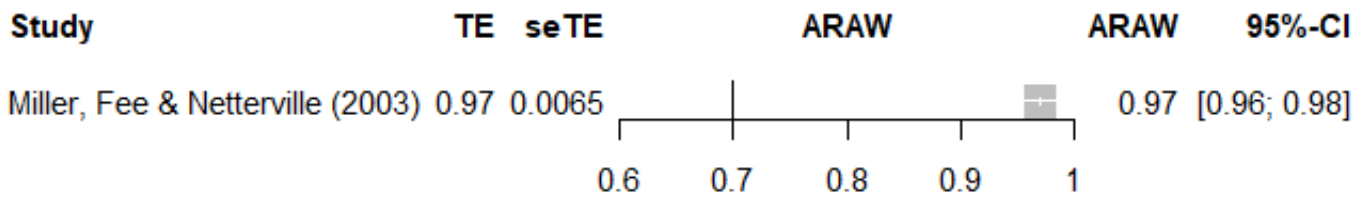


Appendices Figure 21. Forest plot for the inter-rater reliability of the CBCL 6-18 *aggressive behaviour* scale using a random-effects model.

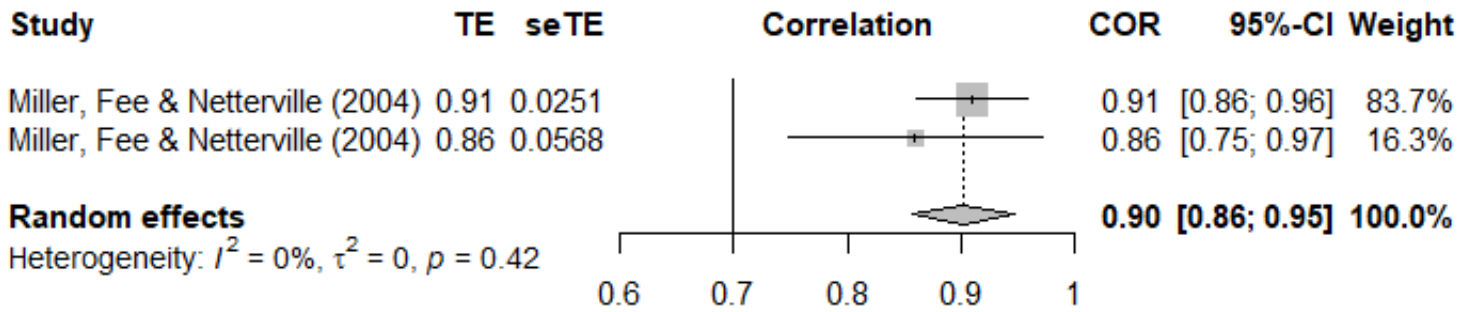


Appendices Figure 22. Forest plot for the test-retest reliability of the CBCL 6-18 *aggressive behaviour* scale using a random-effects model.

Child Behavior Checklist – Teacher Report Form (CBCL-TRF) *aggressive behaviour* scale

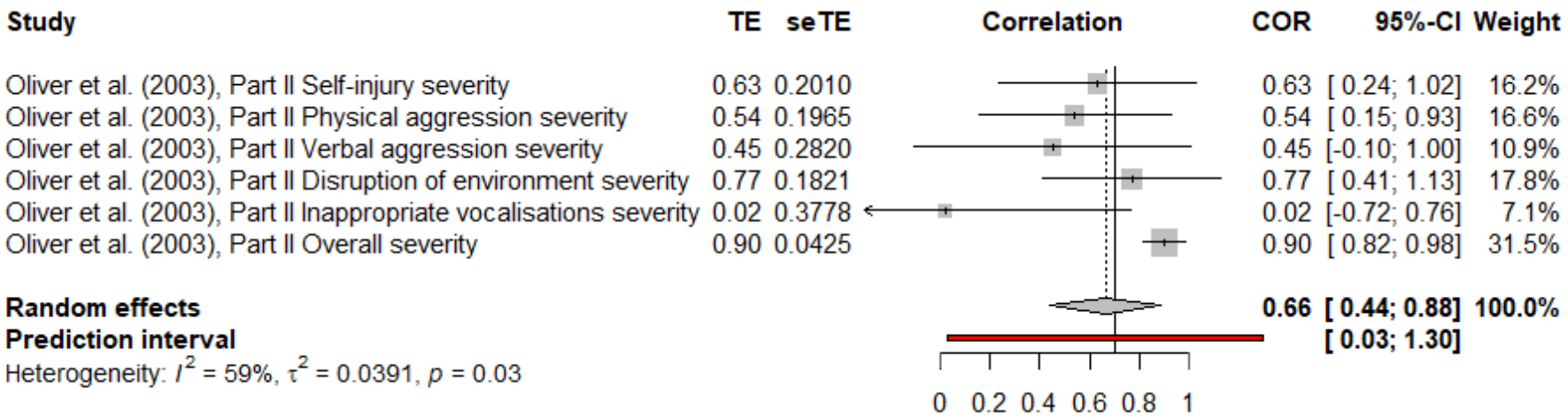


Appendices Figure 23. Forest plot for the internal consistency of the CBCL-TRF *aggressive behaviour* scale using a random-effects model.

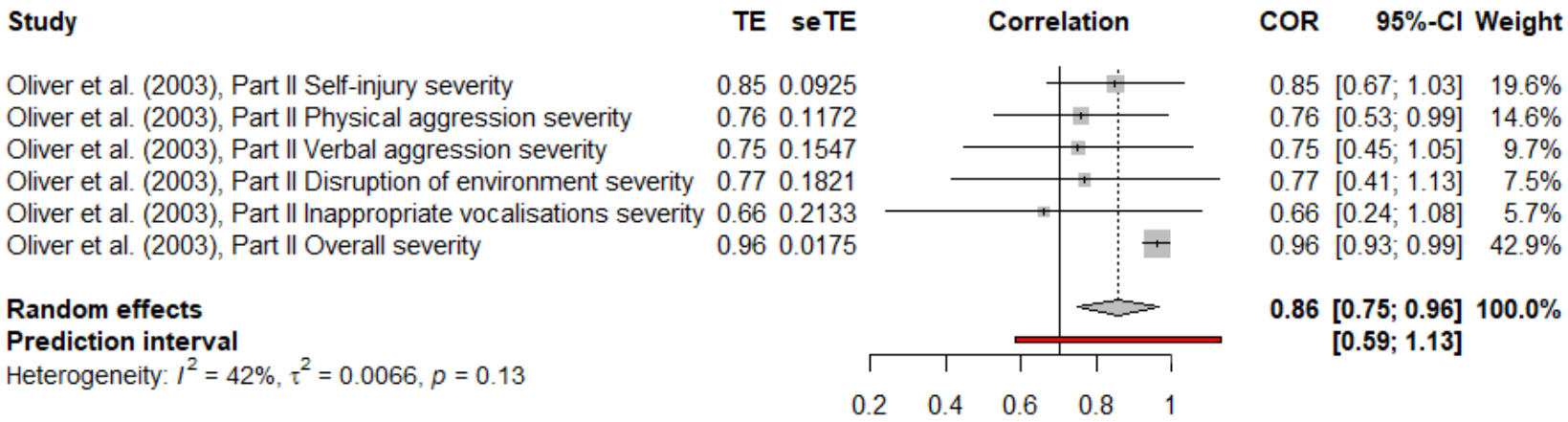


Appendices Figure 24. Forest plot for the test-retest reliability of the CBCL-TRF *aggressive behaviour* scale using a random-effects model.

Challenging Behaviour Interview (CBI)

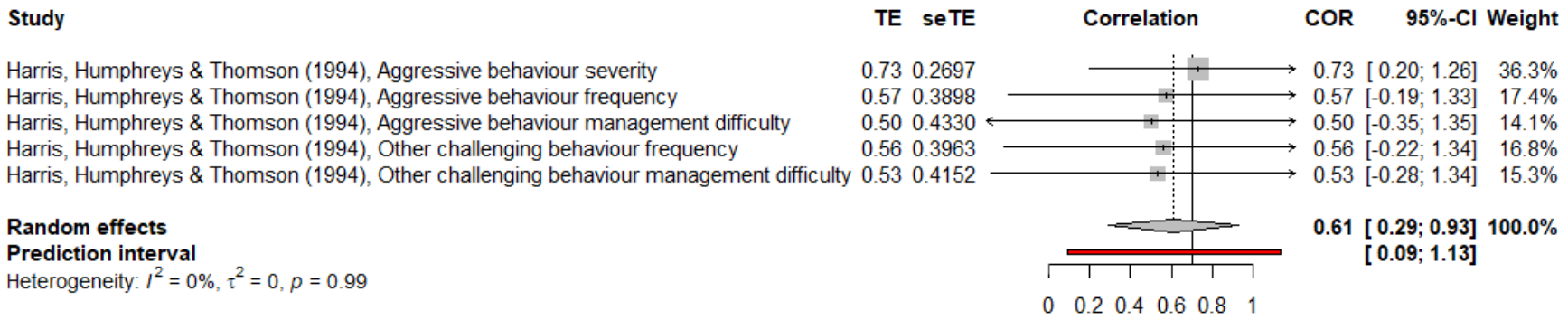


Appendices Figure 25. Forest plot for the inter-rater reliability of the CBI using a random-effects model.

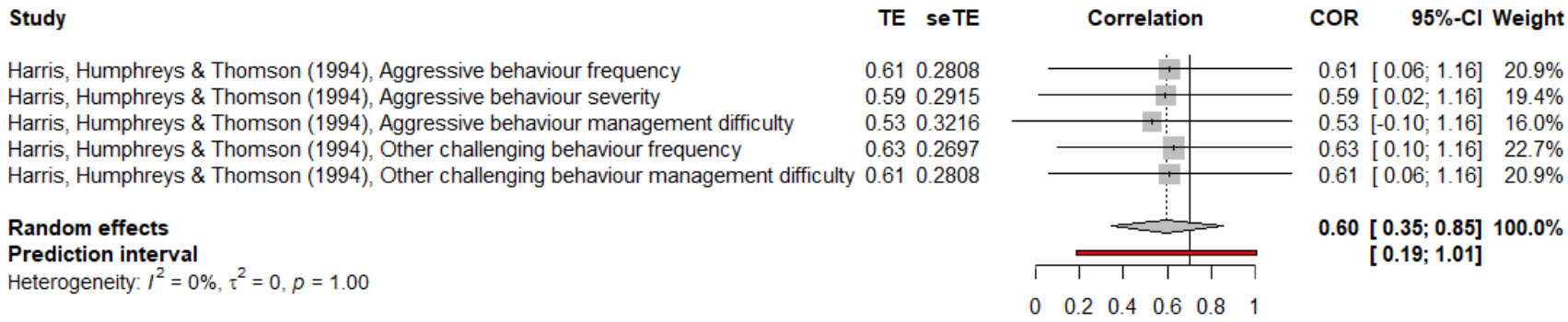


Appendices Figure 26. Forest plot for the test-retest reliability of the CBI using a random-effects model.

Checklist of Challenging Behaviour (CCB)

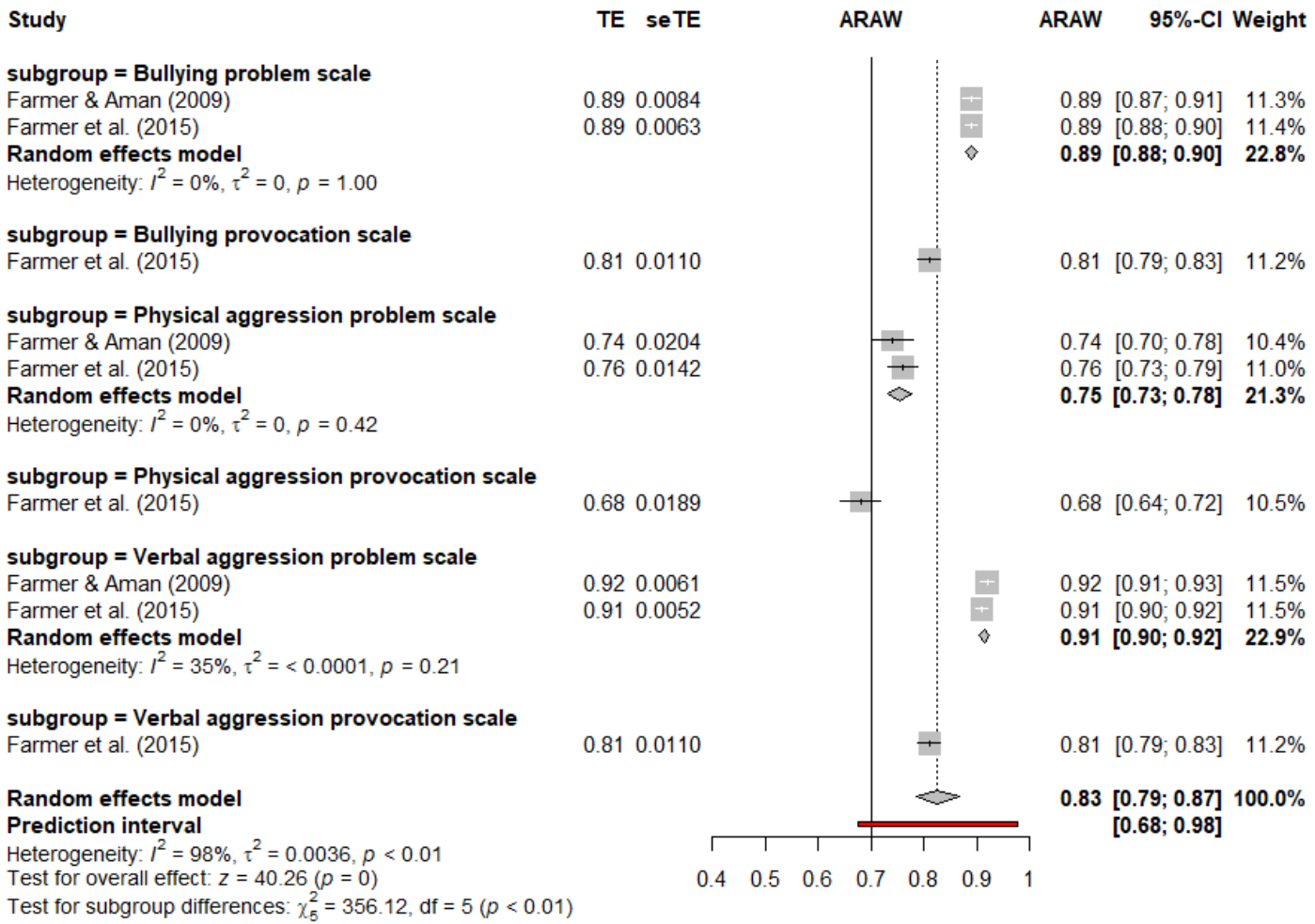


Appendices Figure 27. Forest plot for the inter-rater reliability of the CCB using a random-effects model.

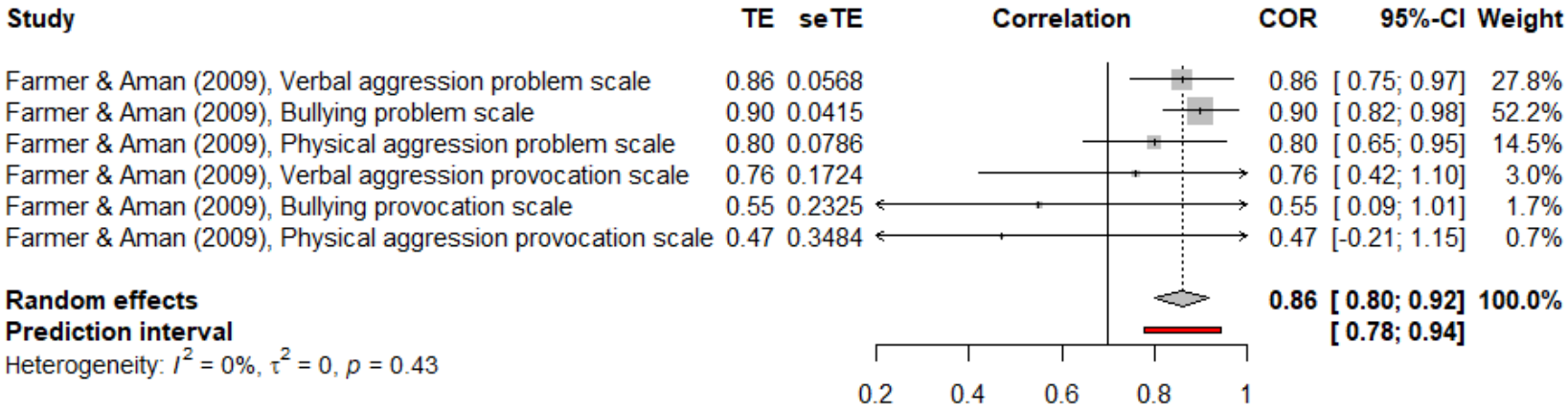


Appendices Figure 28. Forest plot for the test-retest reliability of the CCB using a random-effects model.

Children's Scale of Hostility and Aggression (C-SHARP)

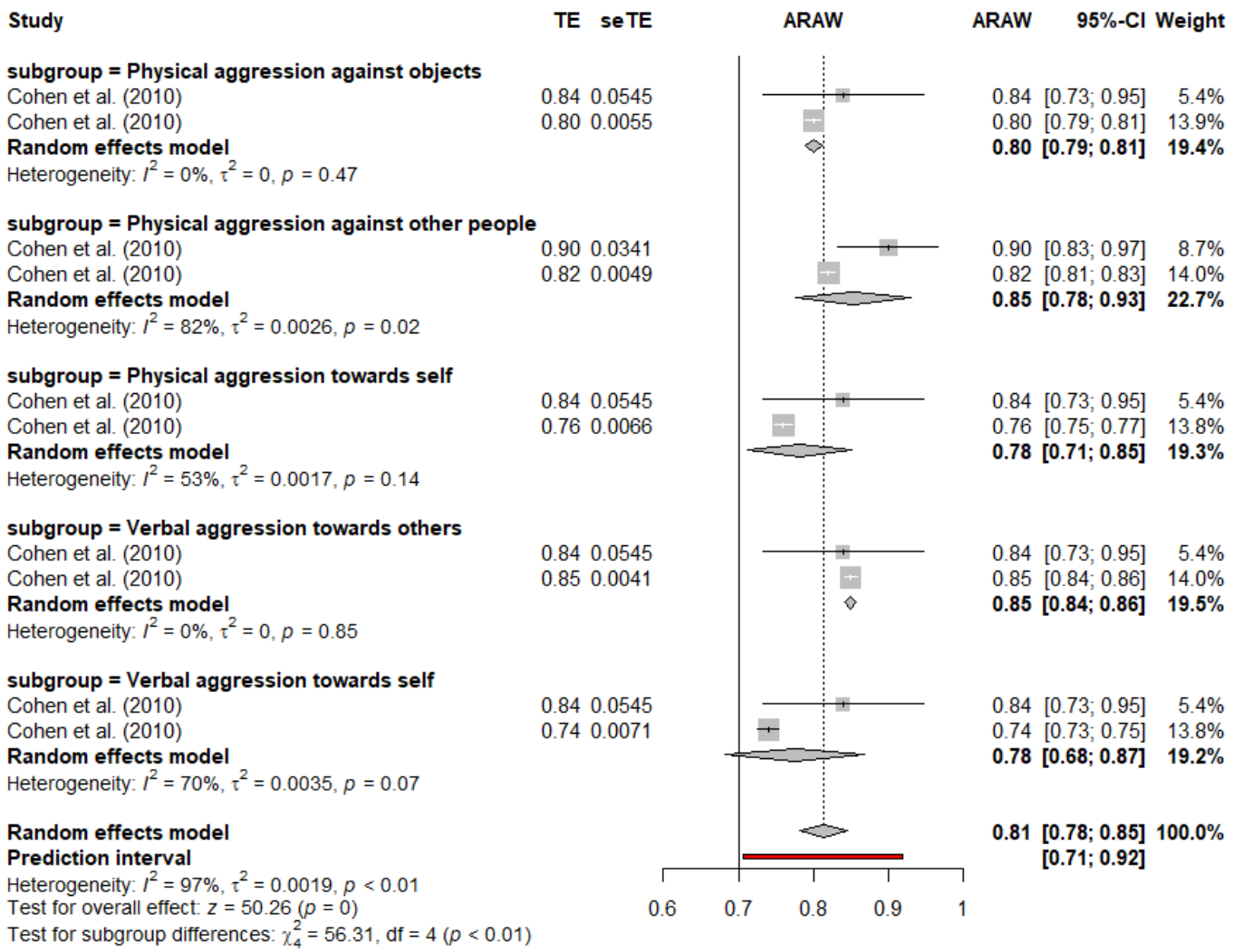


Appendices Figure 29. Forest plot for the internal consistency of the C-SHARP using a random-effects model.

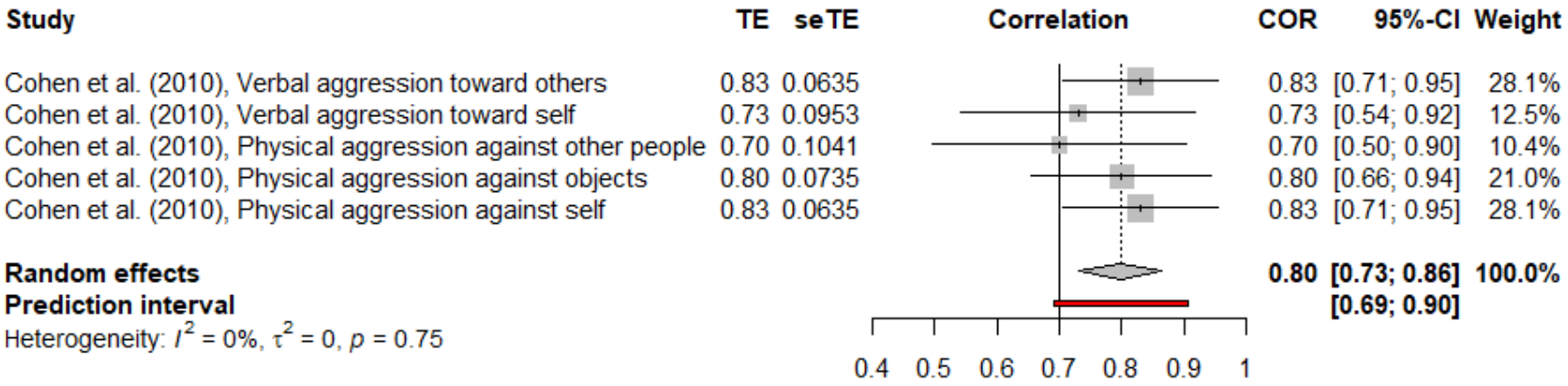


Appendices Figure 30. Forest plot for the inter-rater reliability of the C-SHARP using a random-effects model.

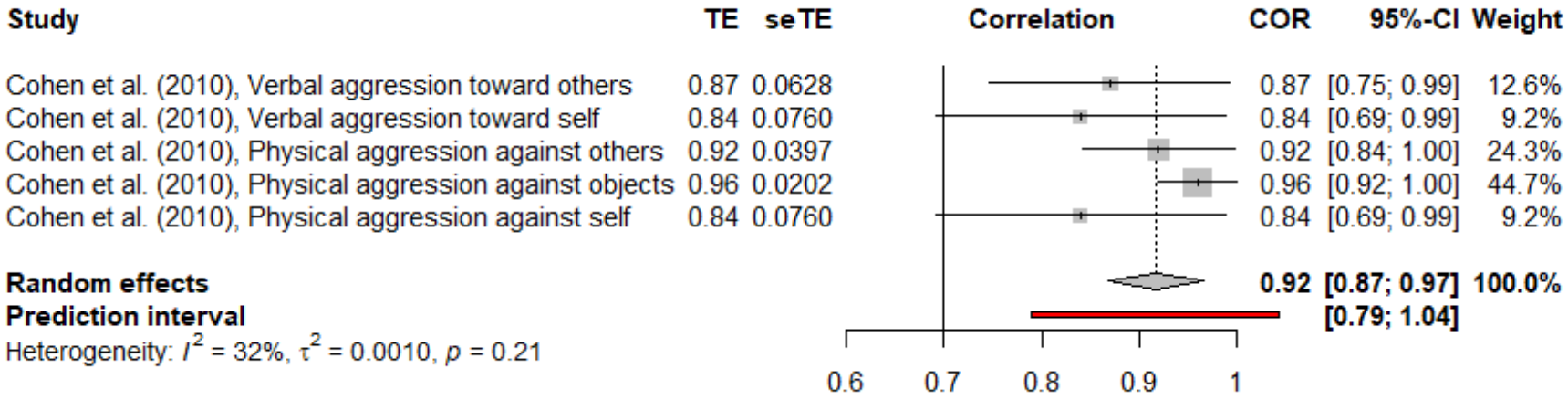
Institute for Basic Research-Modified Overt Aggression Scale (IBR-MOAS)



Appendices Figure 31. Forest plot for the internal consistency of the IBR-MOAS using a random-effects model.

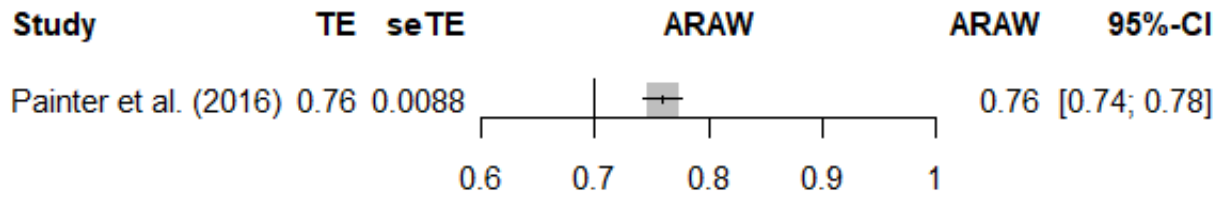


Appendices Figure 32. Forest plot for the inter-rater reliability of the IBR-MOAS using a random-effects model.

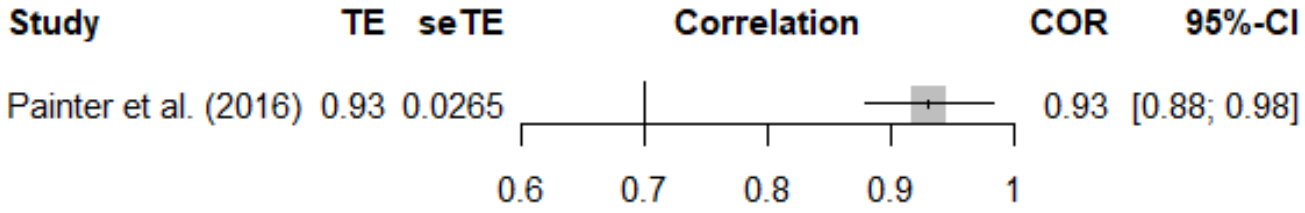


Appendices Figure 33. Forest plot for the test-retest reliability of the IBR-MOAS using a random-effects model.

Learning Disability Needs Assessment Tool (LDNAT) challenging behaviour scale

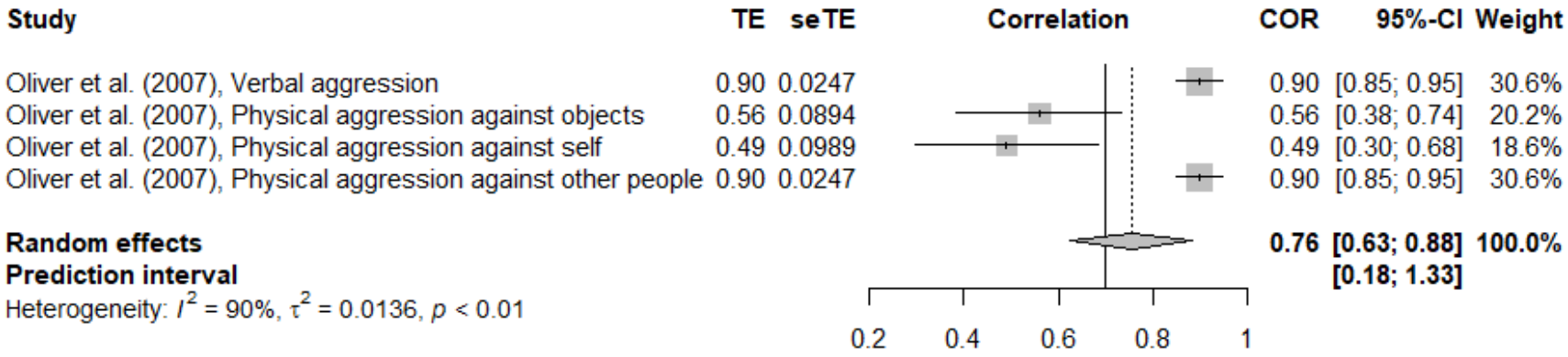


Appendices Figure 34. Forest plot for the internal consistency of the LDNAT *challenging behaviour* scale using a random-effects model.



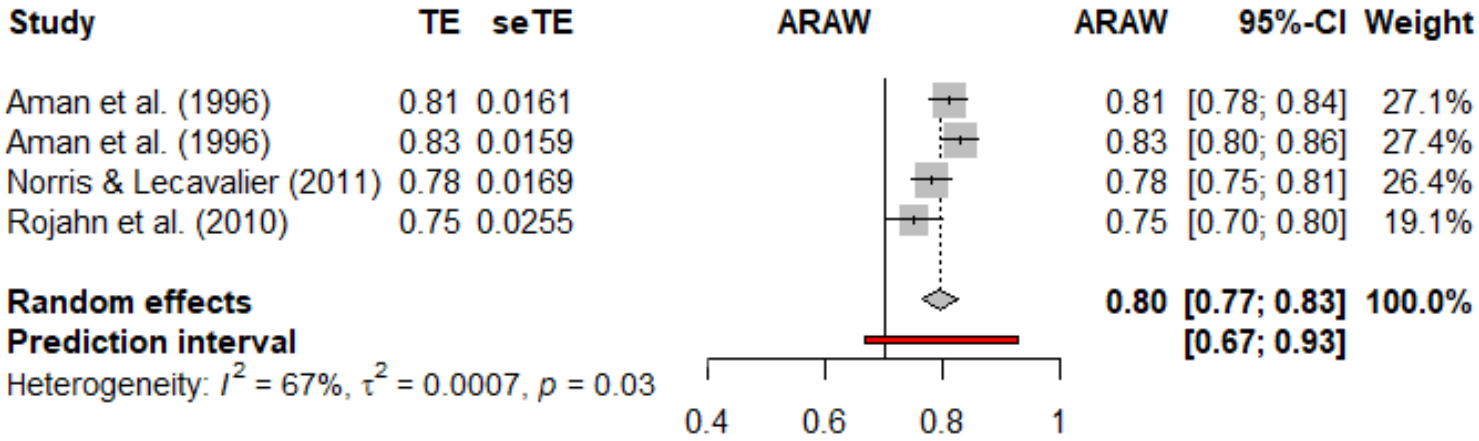
Appendices Figure 35. Forest plot for the test-retest reliability of the LDNAT *challenging behaviour* scale using a random-effects model.

Modified Overt Aggression Scale (MOAS)

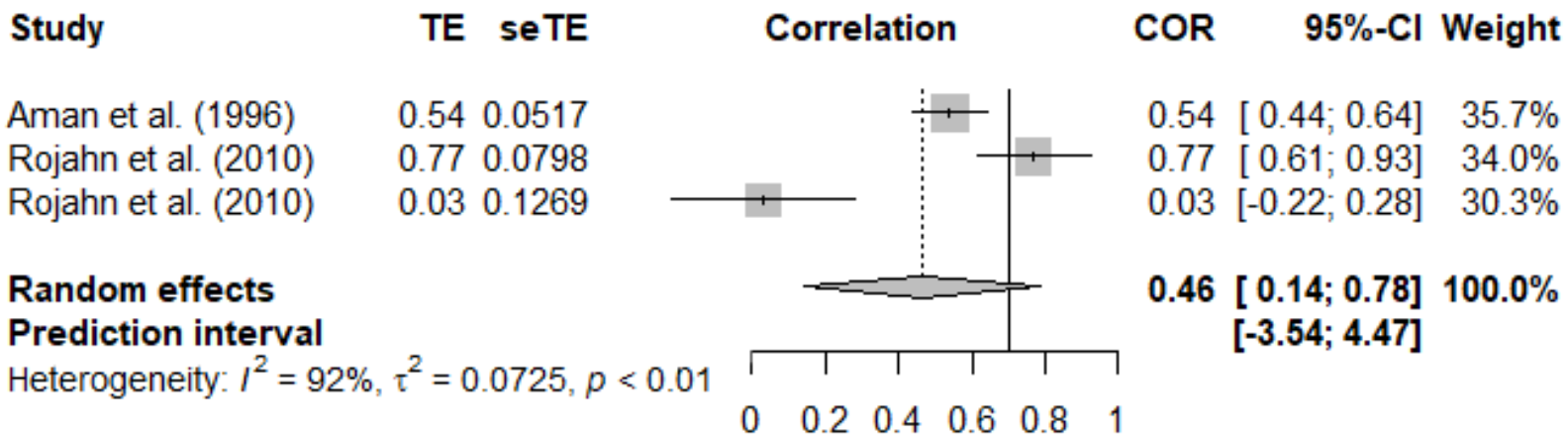


Appendices Figure 36. Forest plot for the inter-rater reliability of the MOAS using a random-effects model.

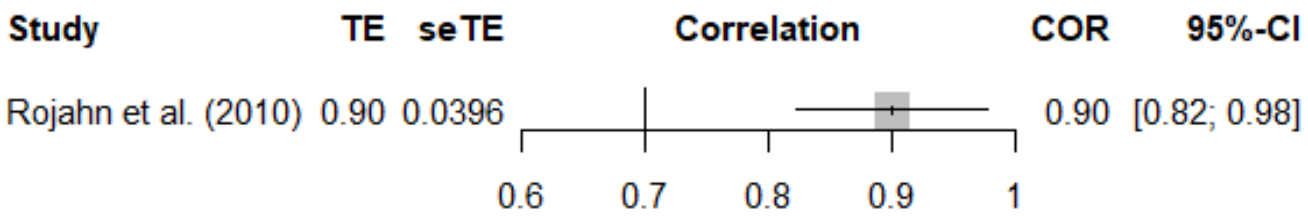
Nisonger Child Behavior Rating Form (NCBRF) self-injury/stereotypic subscale



Appendices Figure 37. Forest plot for the internal consistency of the NCBRF *self-injury/stereotypic sub*scale using a random-effects model.

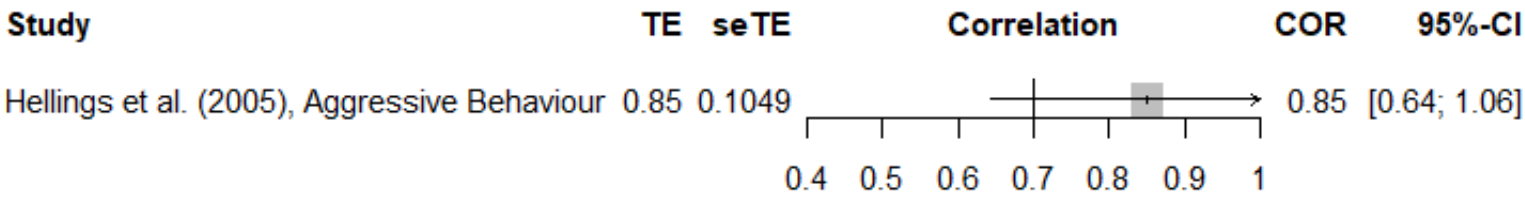


Appendices Figure 38. Forest plot for the inter-rater reliability of the NCBRF *self-injury/stereotypic* subscale using a random-effects model.



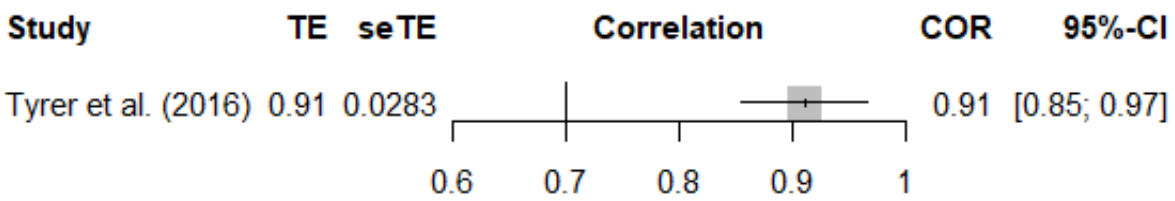
Appendices Figure 39. Forest plot for the test-retest reliability of the NCBRF *self-injury/stereotypic* subscale using a random-effects model.

Overt Aggression Scale (OAS)



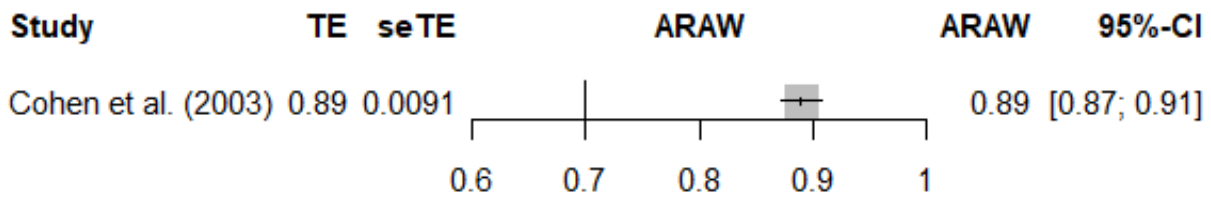
Appendices Figure 40. Forest plot for the inter-rater reliability of the OAS using a random-effects model.

Problem Behavior Checklist (PBCL)

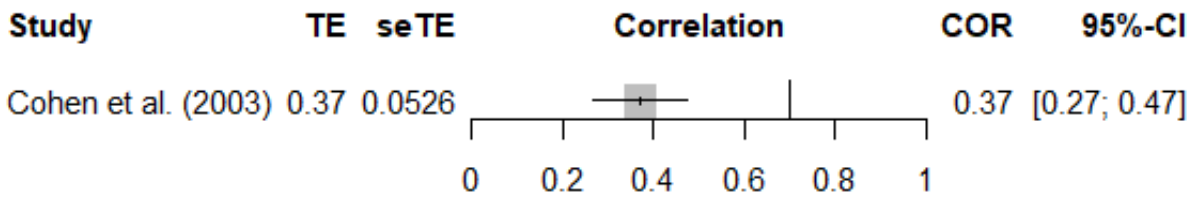


Appendices Figure 41. Forest plot for the inter-rater reliability of the PBCL using a random-effects model.

Pervasive Developmental Disorder Behavior Inventory-Parent Version (PDDBI-Parent) *aggression* scale

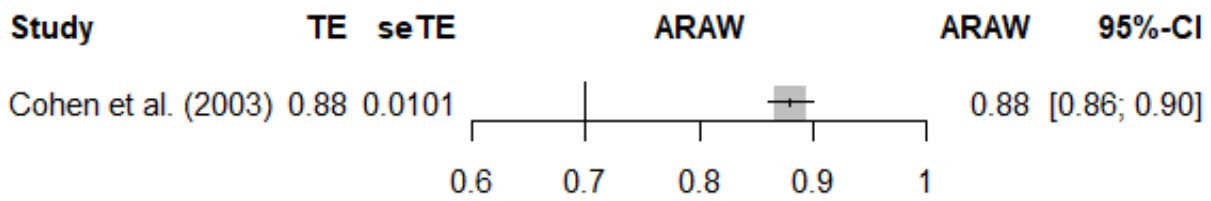


Appendices Figure 42. Forest plot for the internal consistency of the PDDBI-Parent *aggression* scale using a random-effects model.

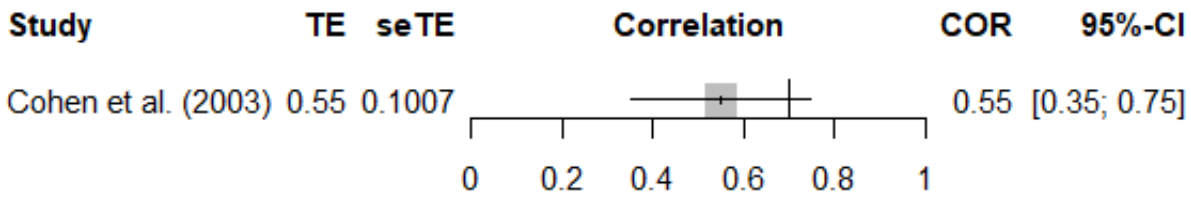


Appendices Figure 43. Forest plot for the inter-rater reliability of the PDDBI-Parent *aggression* scale using a random-effects model.

Pervasive Developmental Disorder Behavior Inventory-Parent Version (PDDBI-Teacher) *aggression* scale

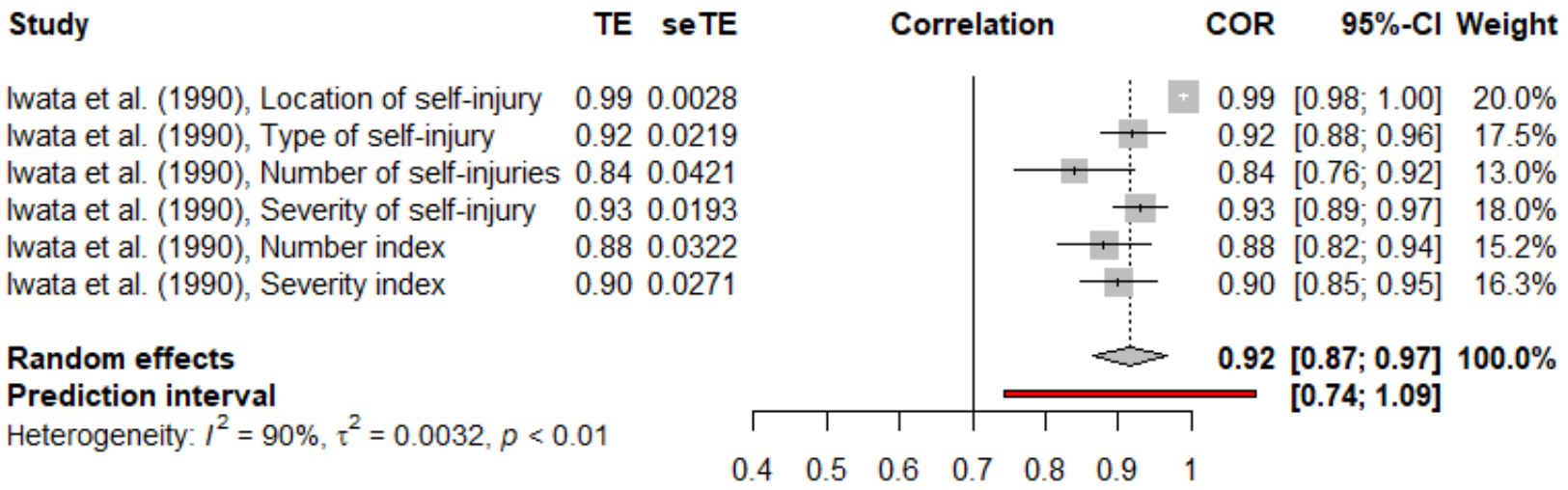


Appendices Figure 44. Forest plot for the internal consistency of the PDDBI-Teacher *aggression* scale using a random-effects model.



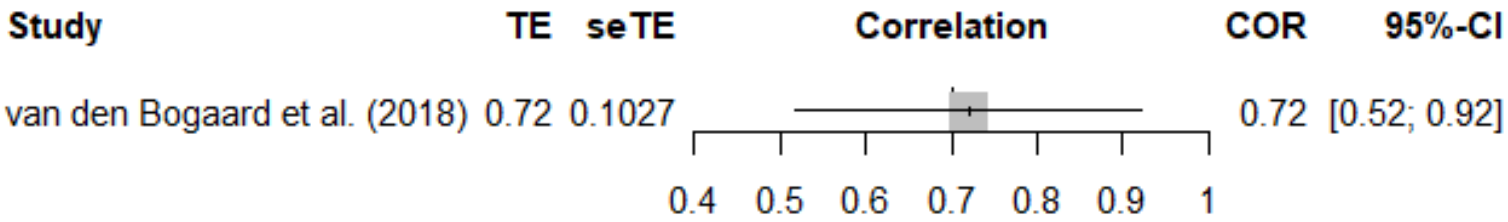
Appendices Figure 45. Forest plot for the inter-rater reliability of the PDDBI-Teacher *aggression* scale using a random-effects model.

Self-Injury Trauma Scale (SIT)



Appendices Figure 46. Forest plot for the inter-rater reliability of the SIT using a random-effects model.

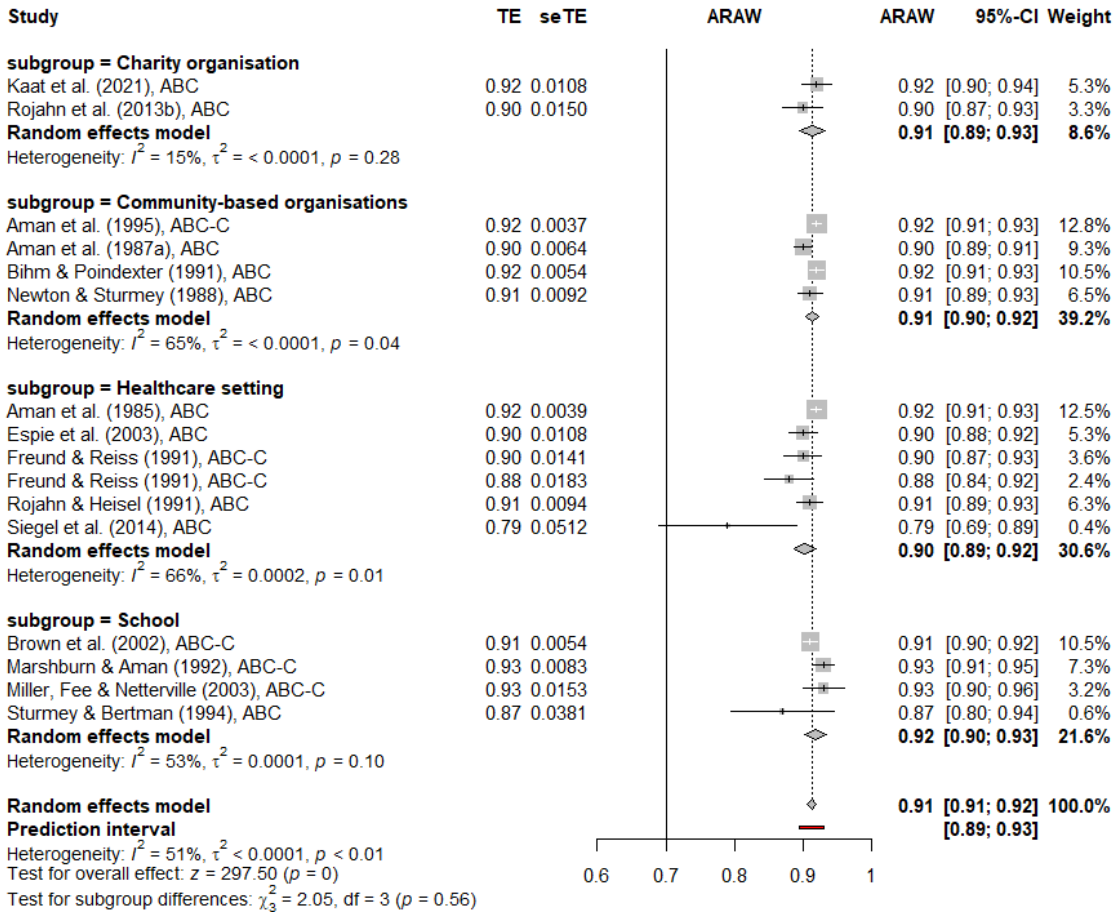
Staff Observation Aggression Scale – Intellectual Disability – Revised (SOAS-ID-R)



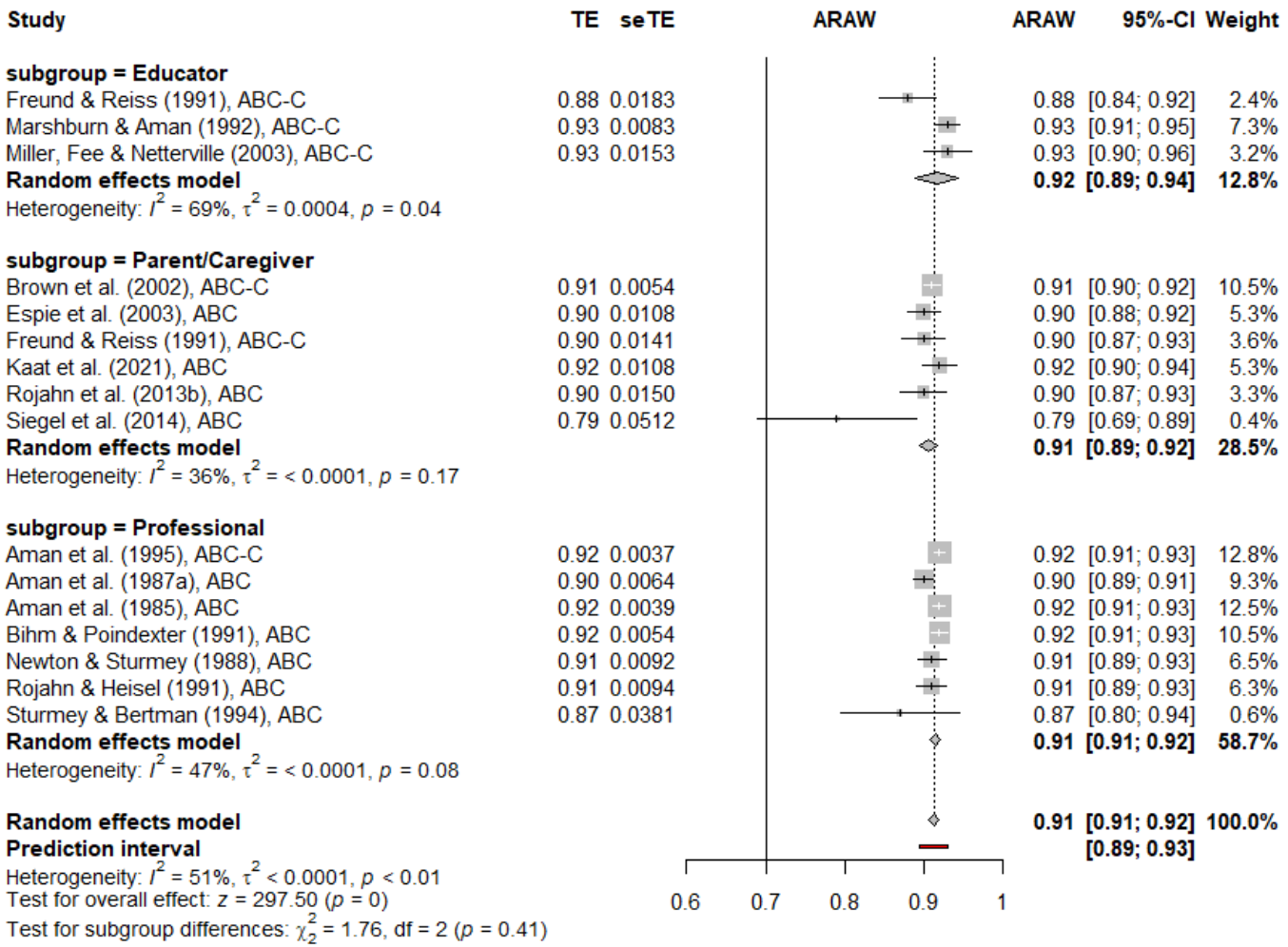
Appendices Figure 47. Forest plot for the inter-rater reliability of the SOAS-ID-R using a random-effects model.

Appendix 18: Chapter Three subgroup analysis forest plots per measure of behaviours that challenge

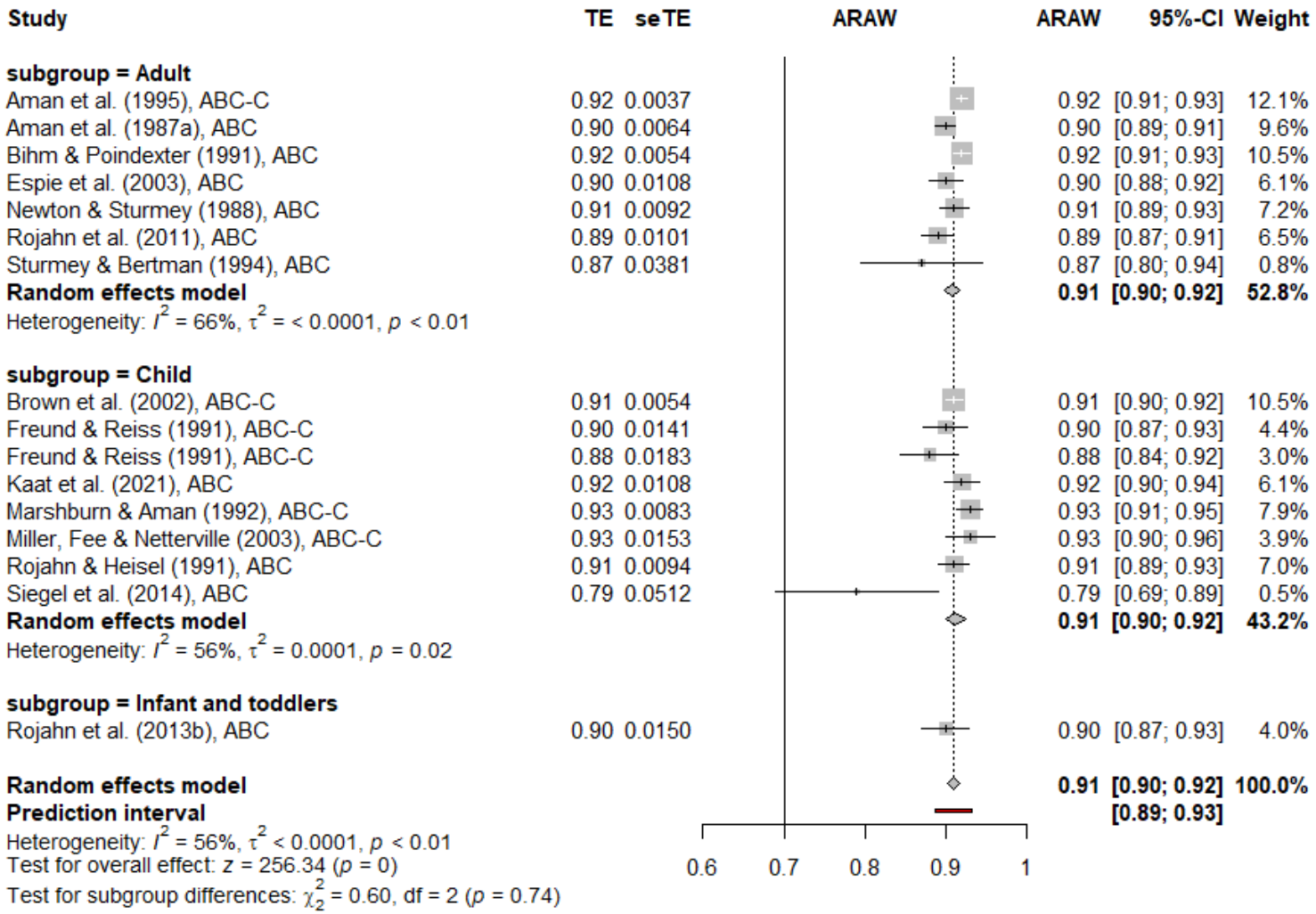
Aberrant Behavior Checklist (ABC) *irritability* subscale: internal consistency



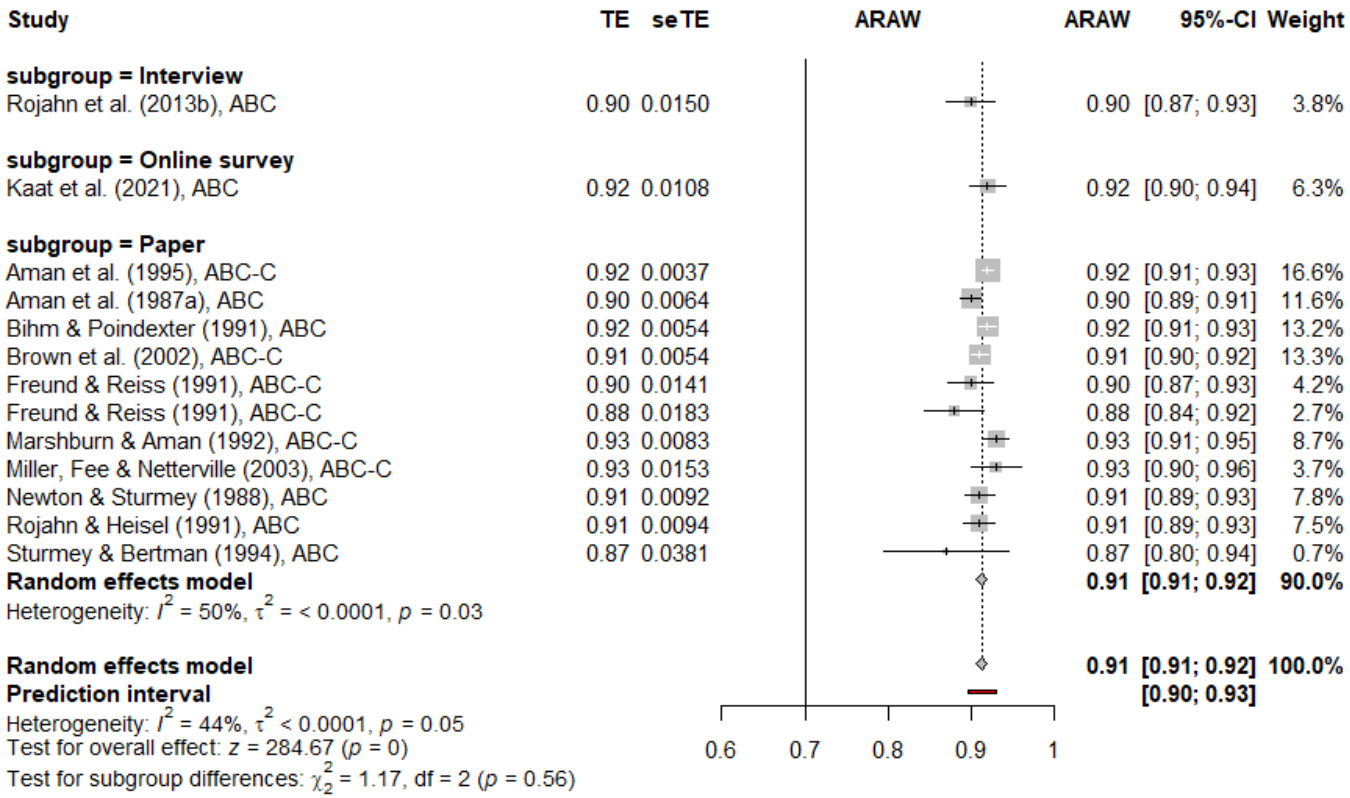
Appendices Figure 48. Subgroup forest plot assessing differences in the internal consistency of the ABC *irritability* subscale attributable to recruitment strategy.



Appendices Figure 49. Subgroup forest plot assessing differences in the internal consistency of the ABC *irritability* subscale attributable to informant completing the measure.

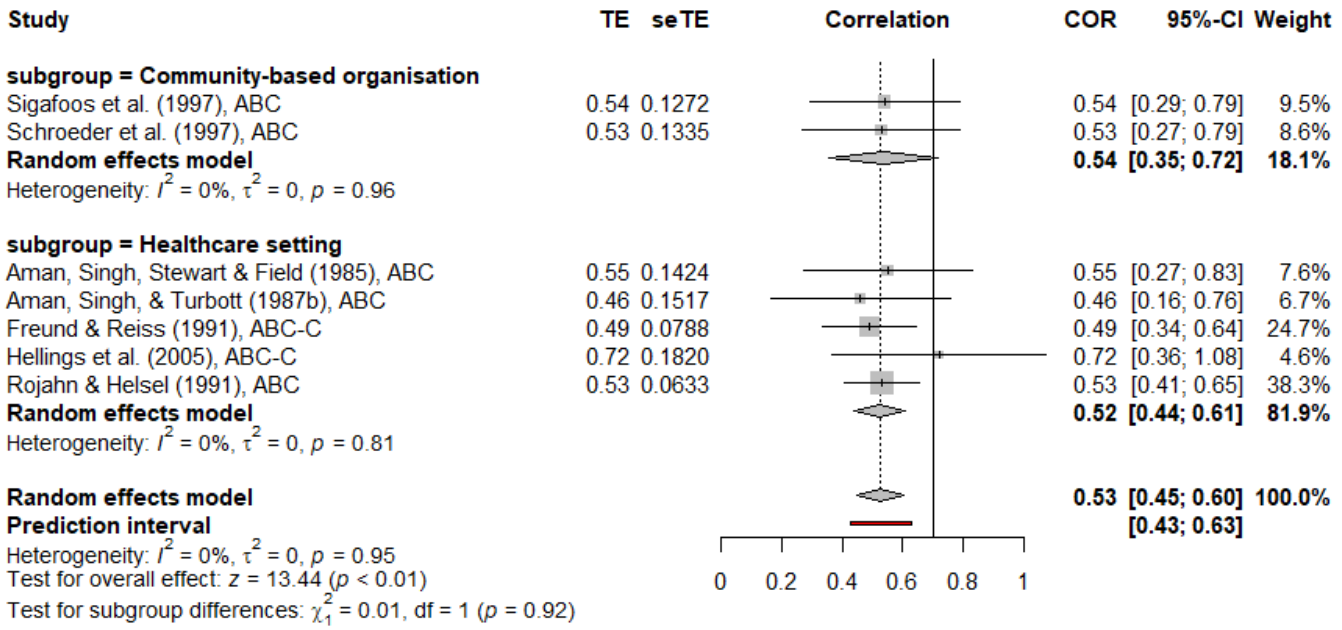


Appendices Figure 50. Subgroup forest plot assessing differences in the internal consistency of the ABC *irritability* subscale attributable to ratings for children or adults.

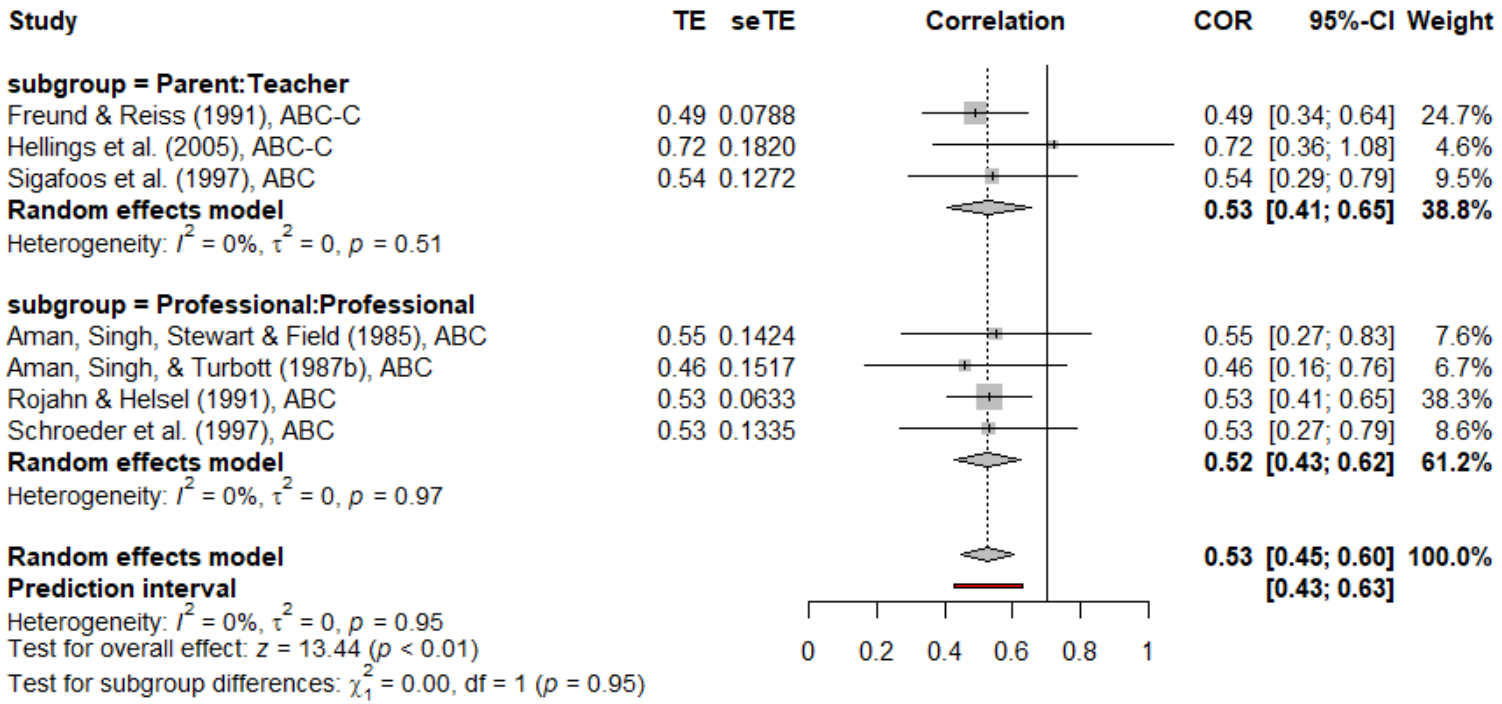


Appendices Figure 51. Subgroup forest plot assessing differences in the internal consistency of the ABC *irritability* subscale attributable to method of administration.

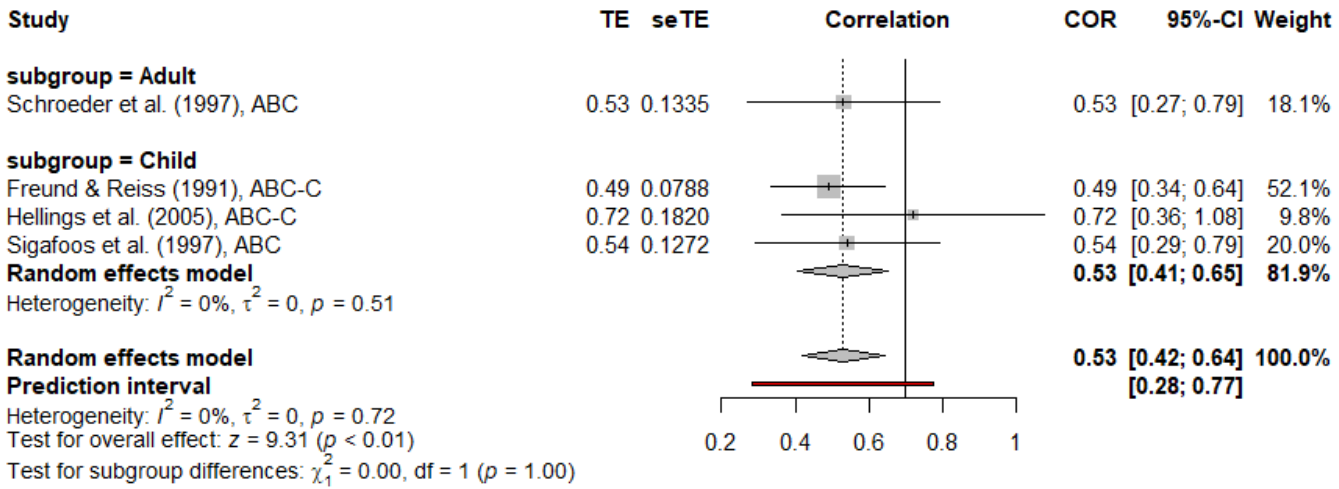
Aberrant Behavior Checklist (ABC) *irritability* subscale: inter-rater reliability



Appendices Figure 52. Subgroup forest plot assessing differences in the inter-rater reliability of the ABC *irritability* subscale attributable to recruitment strategy.

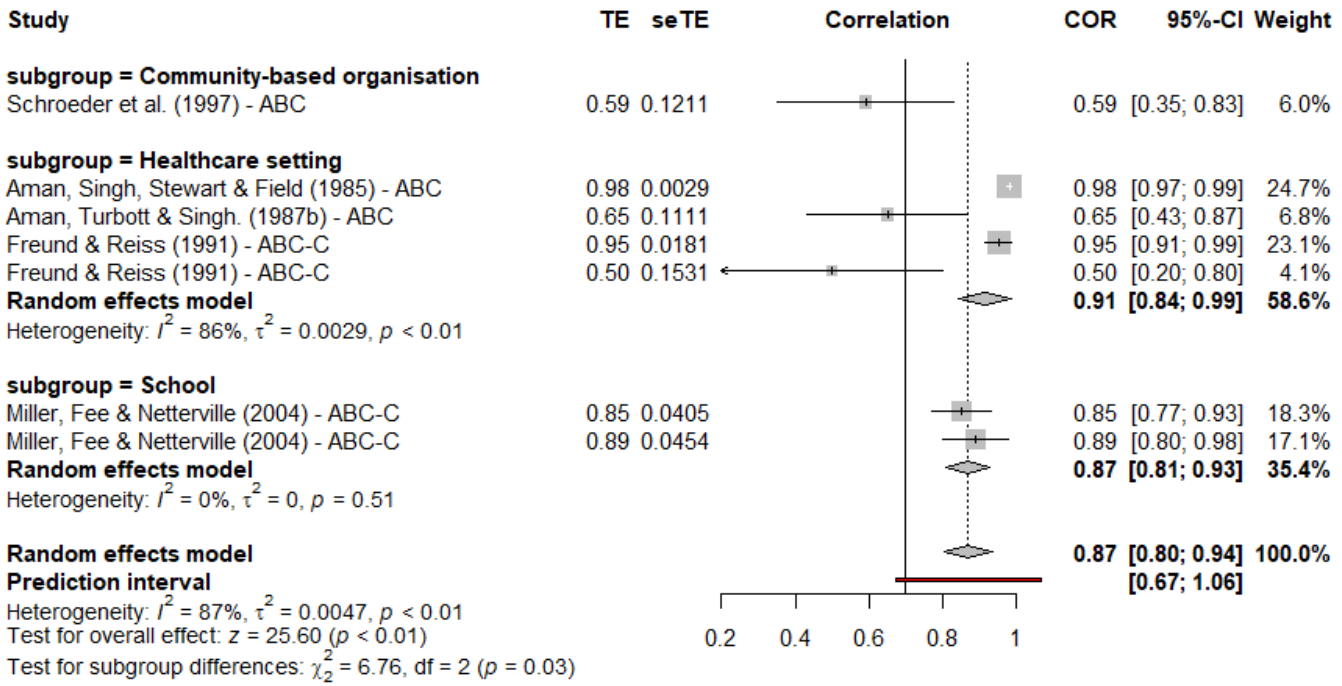


Appendices Figure 53. Subgroup forest plot assessing differences in the inter-rater reliability of the ABC *irritability* subscale attributable to informant pairs completing the measure.

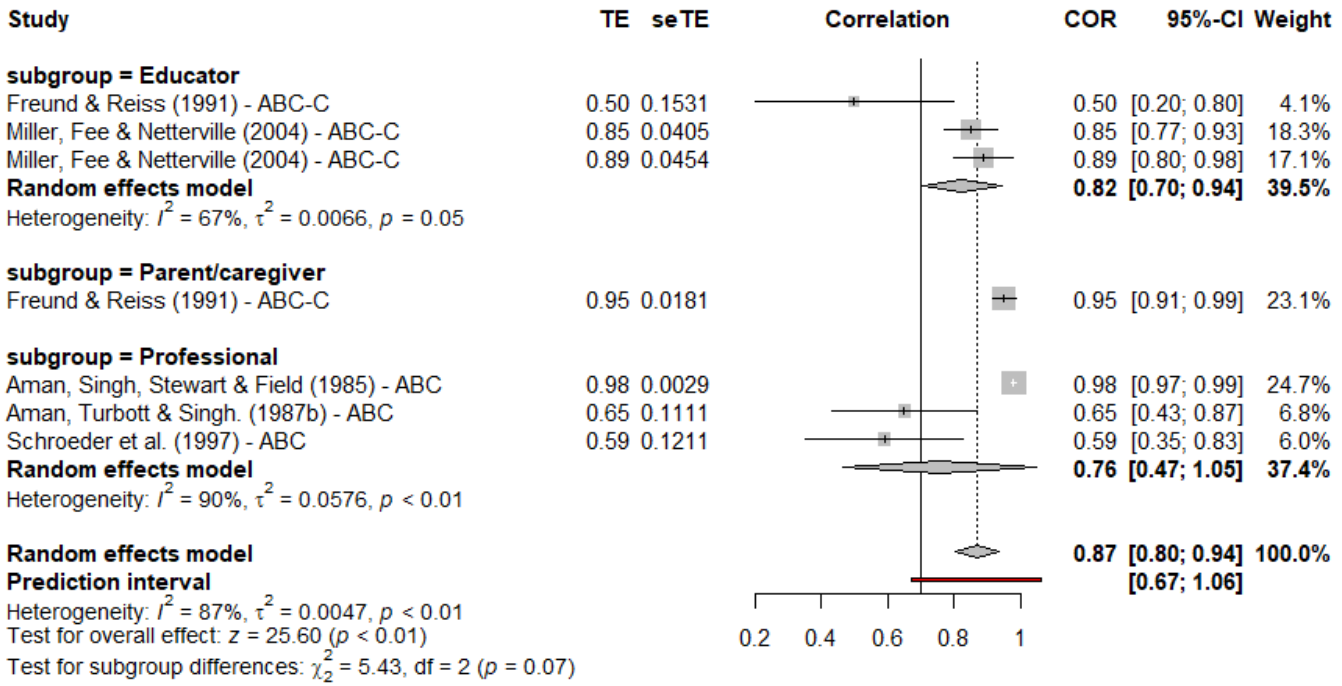


Appendices Figure 54. Subgroup forest plot assessing differences in the inter-rater reliability of the ABC *irritability* subscale attributable to ratings for children or adults.

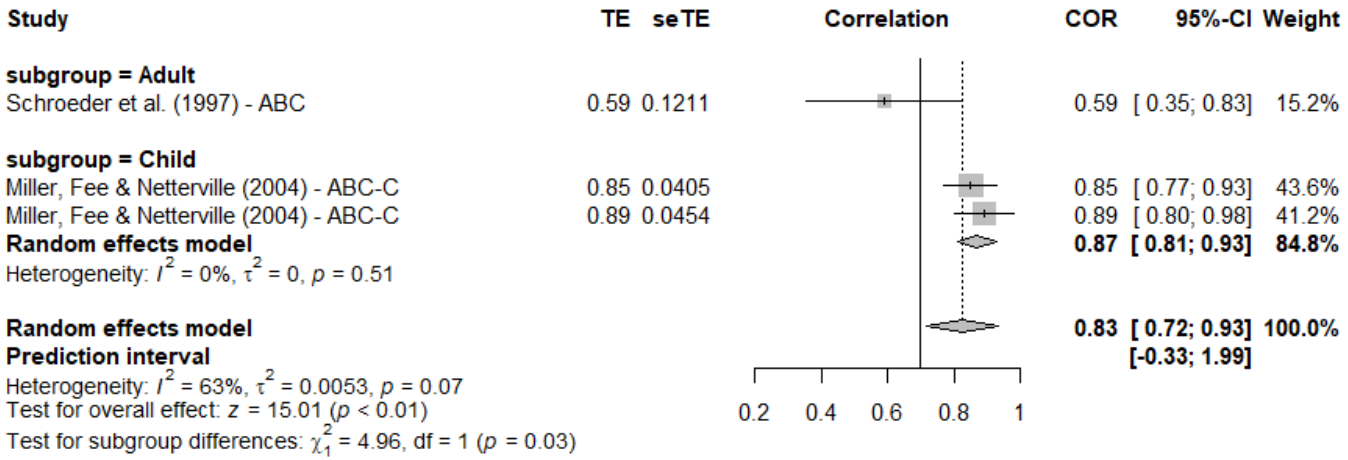
Aberrant Behavior Checklist (ABC) irritability subscale: test-retest reliability



Appendices Figure 55. Subgroup forest plot assessing differences in the test-retest reliability of the ABC *irritability* subscale attributable to recruitment strategy.

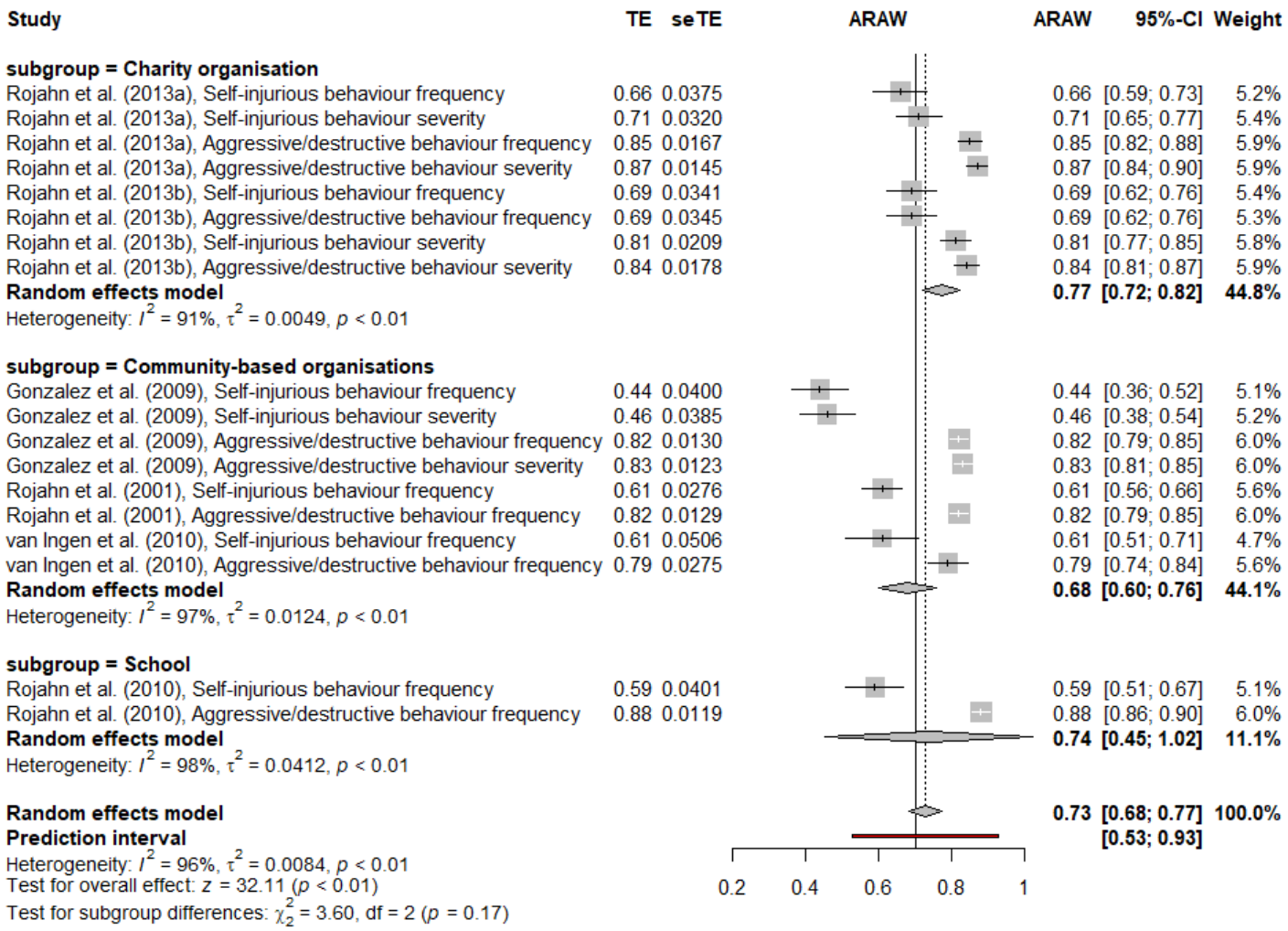


Appendices Figure 56. Subgroup forest plot assessing differences in the test-retest reliability of the ABC *irritability* subscale attributable to informant completing the measure.

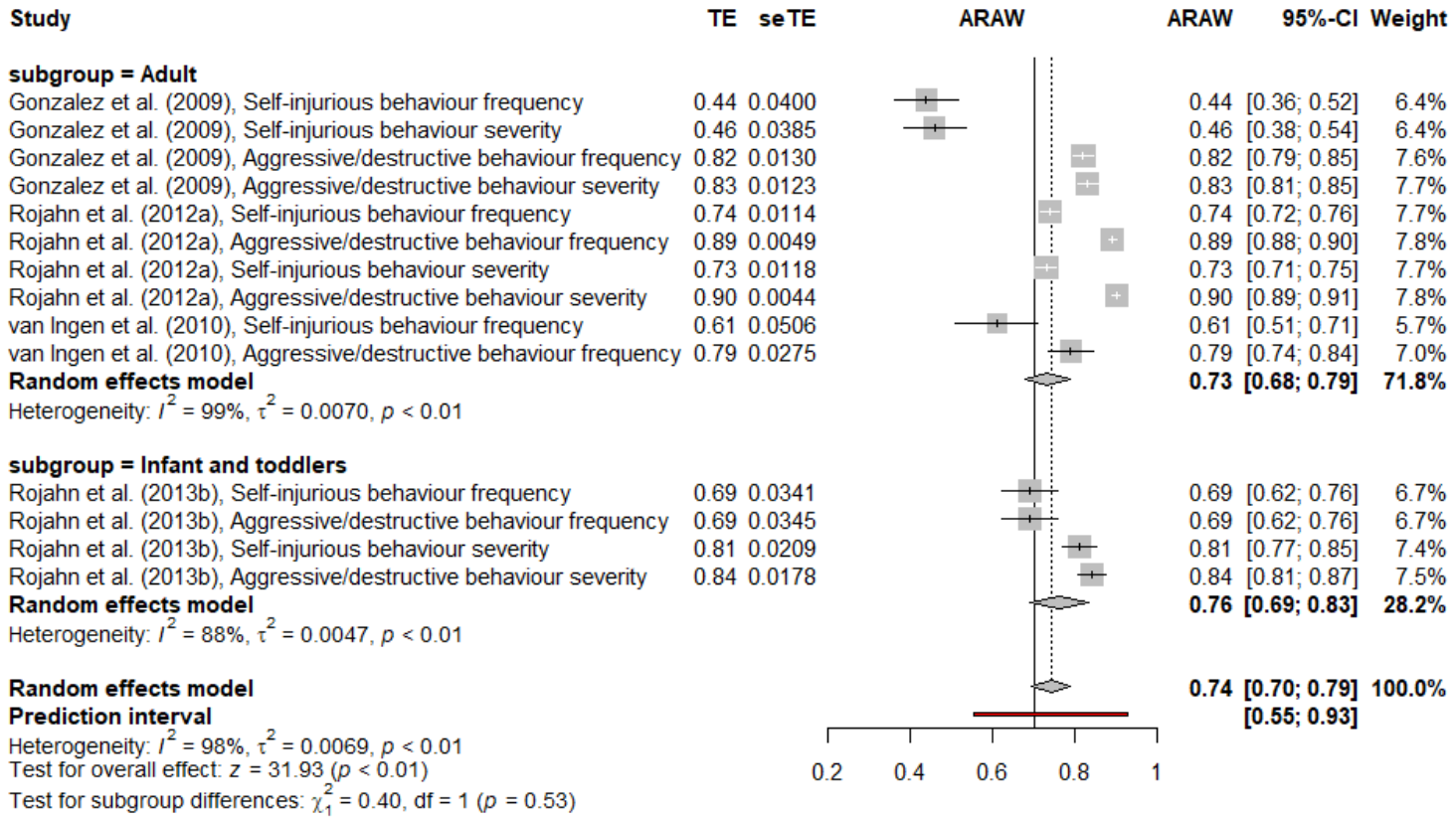


Appendices Figure 57. Subgroup forest plot assessing differences in the test-retest reliability of the ABC *irritability* subscale attributable to ratings for children or adults.

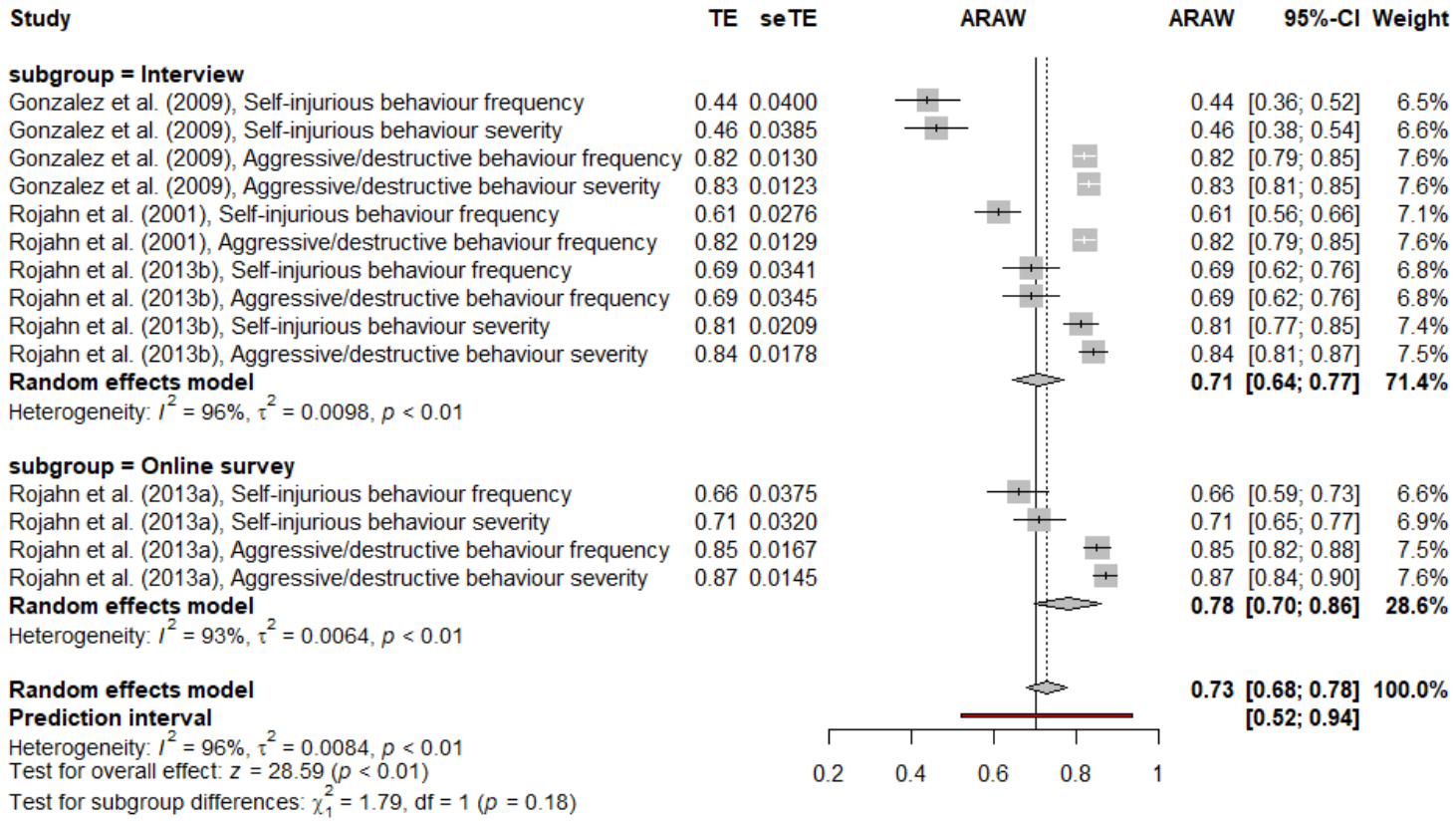
Behavior Problems Inventory – 01 (BPI-01): internal consistency



Appendices Figure 58. Subgroup forest plot assessing differences in the internal consistency of the BPI-01 attributable to recruitment strategy.

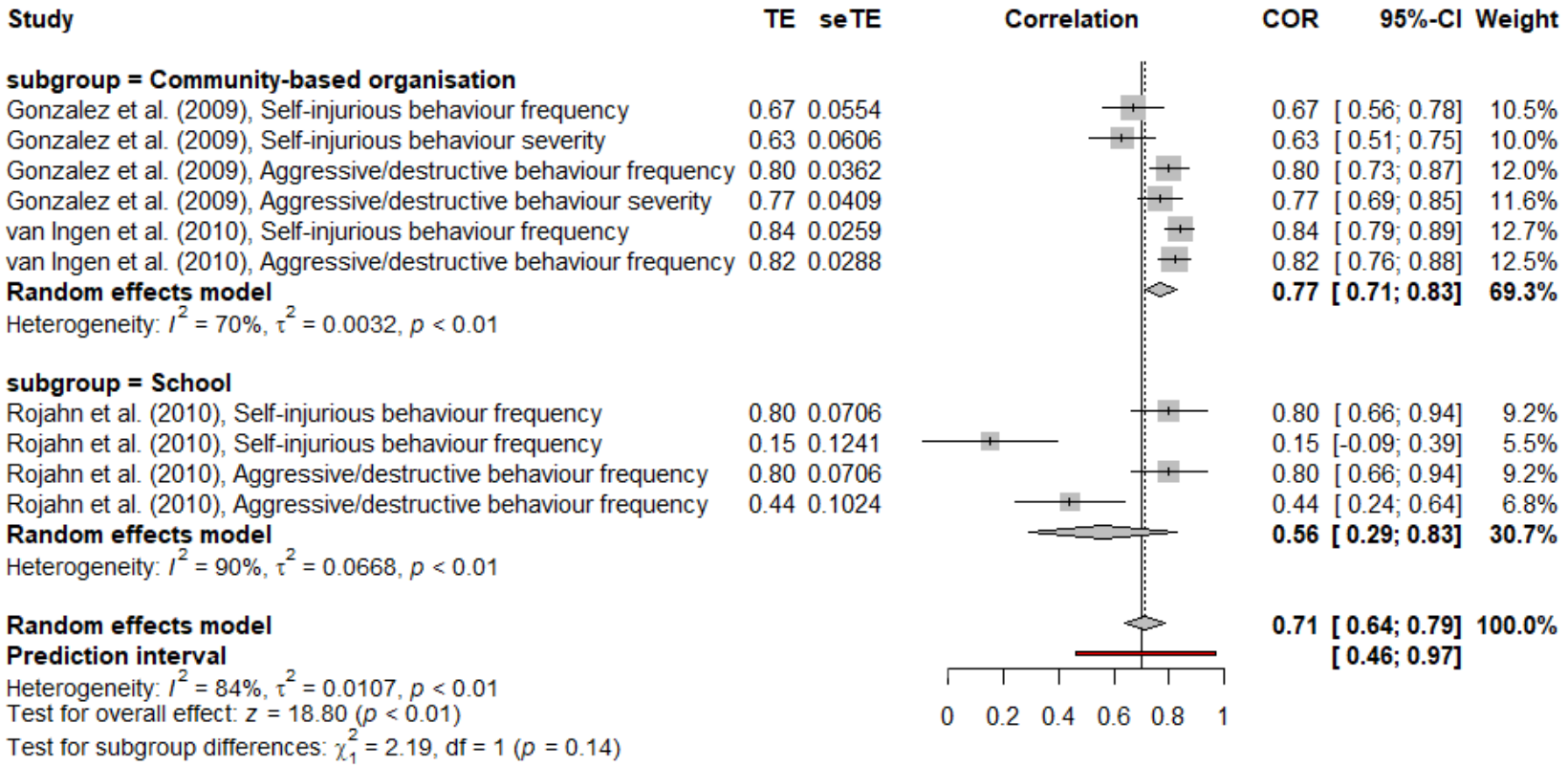


Appendices Figure 59. Subgroup forest plot assessing differences in the internal consistency of the BPI-01 attributable to ratings for children or adults.

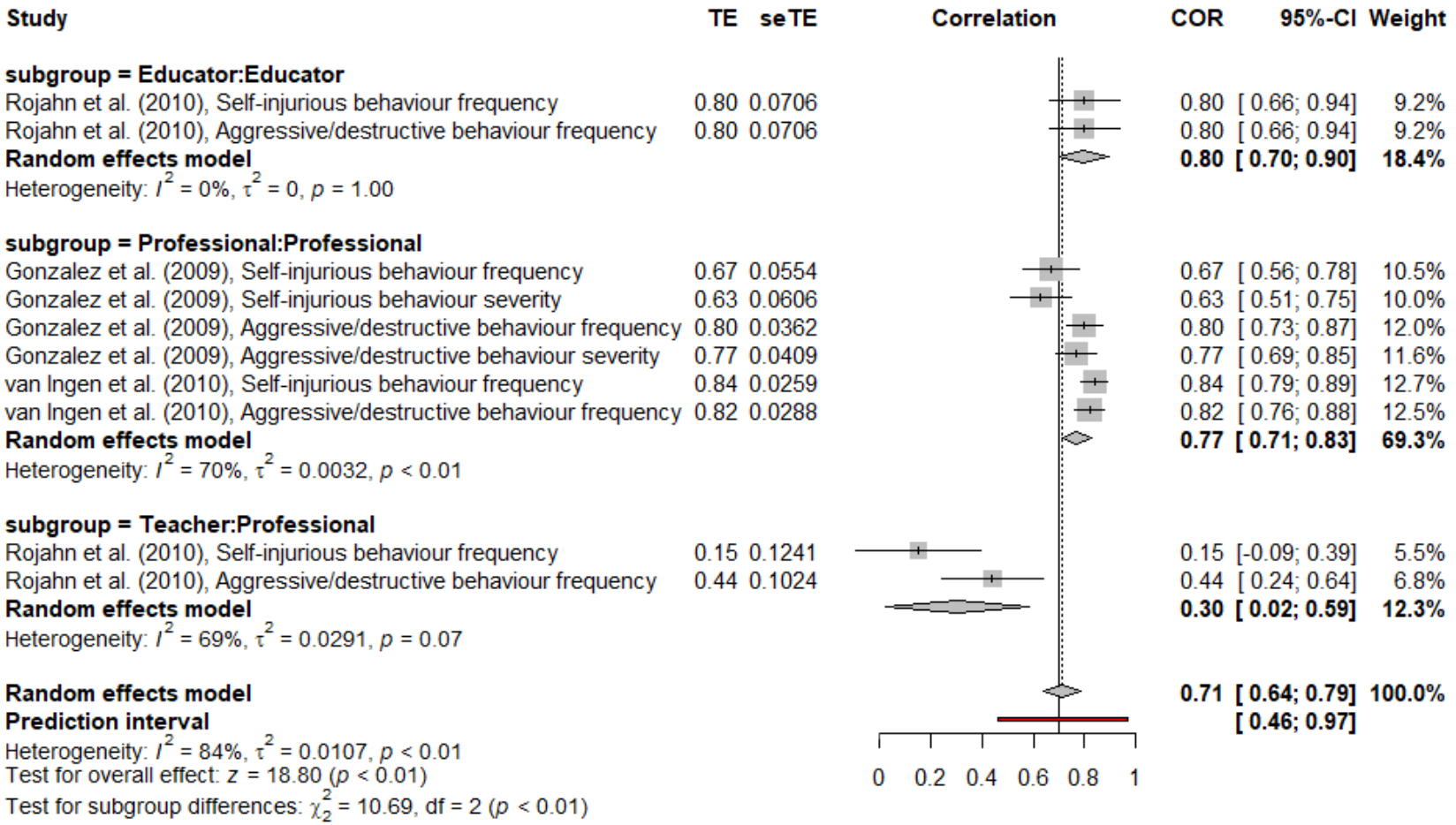


Appendices Figure 60. Subgroup forest plot assessing differences in the internal consistency of the BPI-01 attributable to method of administration.

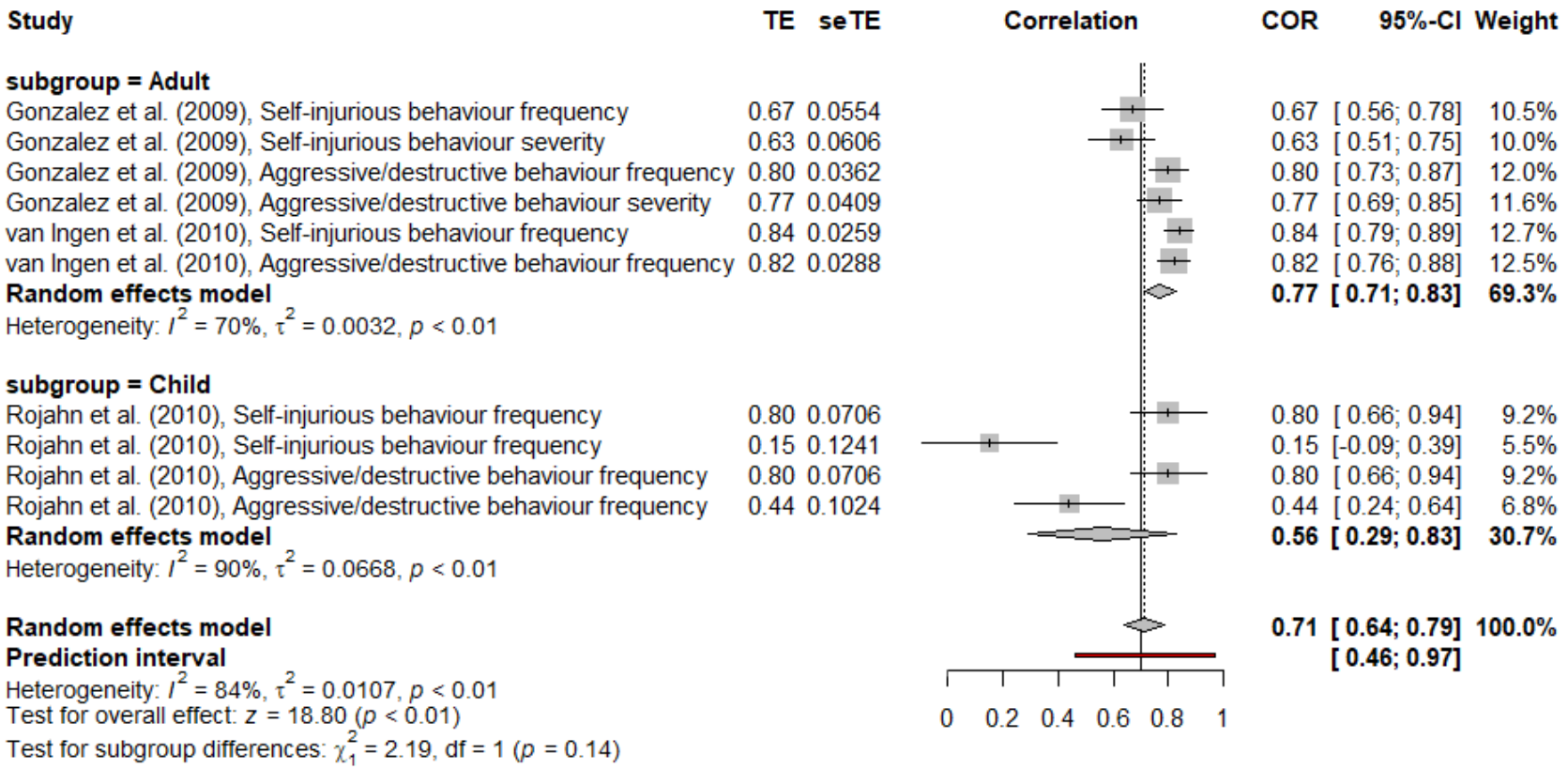
Behavior Problems Inventory – 01 (BPI-01): inter-rater reliability



Appendices Figure 61. Subgroup forest plot assessing differences in the inter-rater reliability of the BPI-01 attributable to recruitment strategy.

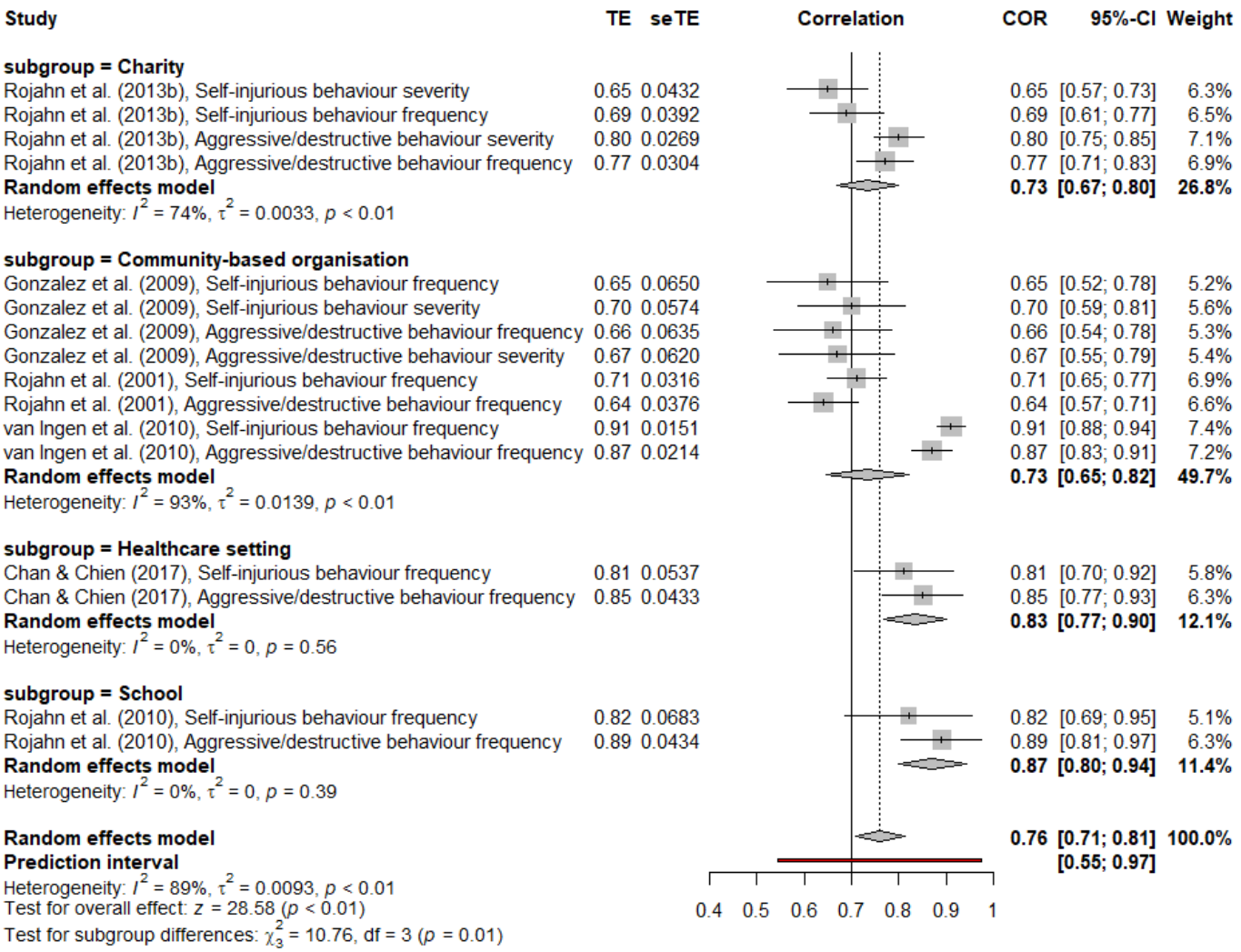


Appendices Figure 62. Subgroup forest plot assessing differences in the inter-rater reliability of the BPI-01 attributable to informant pairs completing the measure.

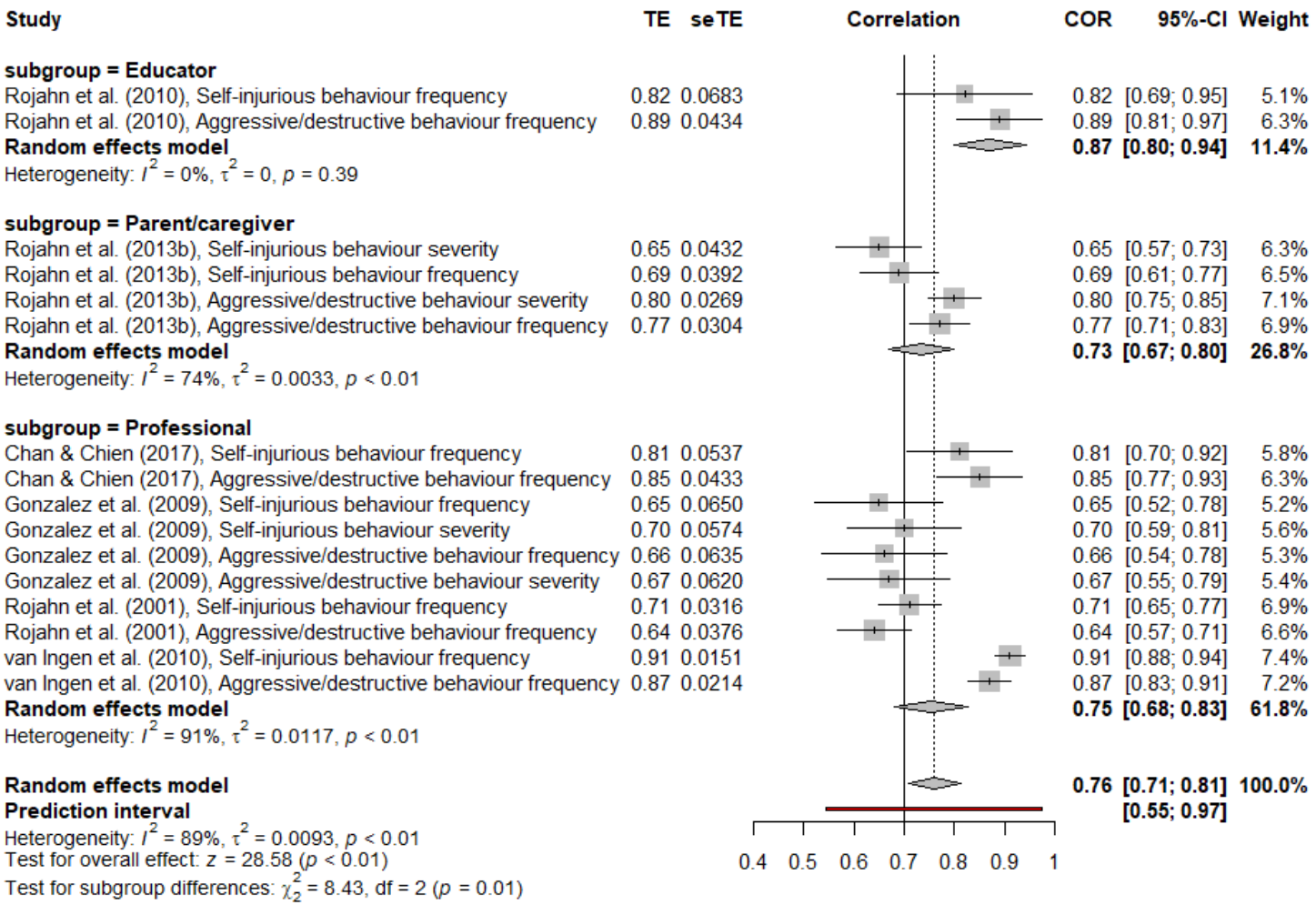


Appendices Figure 63. Subgroup forest plot assessing differences in the inter-rater reliability of the BPI-01 attributable to ratings for children or adults.

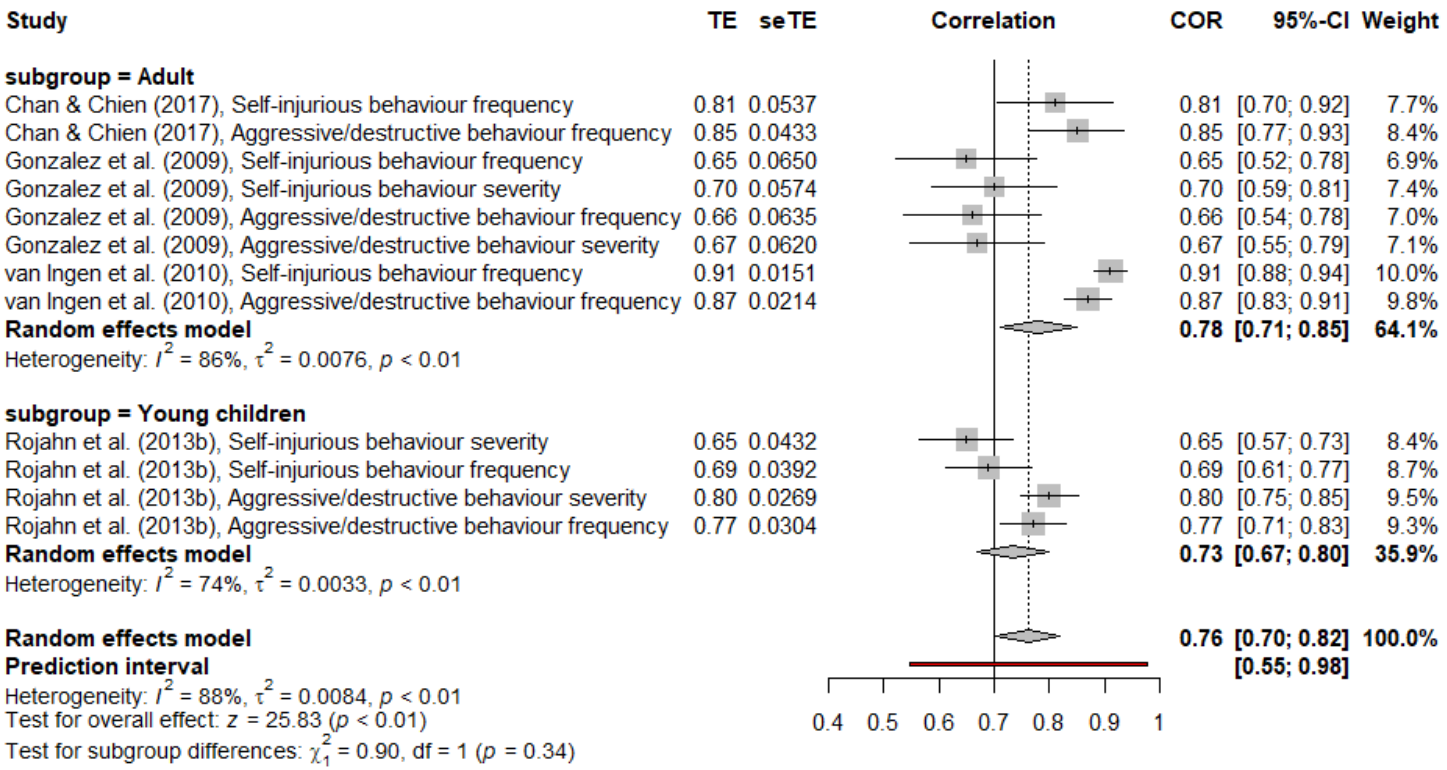
Behavior Problems Inventory – 01 (BPI-01): Test-retest reliability



Appendices Figure 64. Subgroup forest plot assessing differences in the test-retest reliability of the BPI-01 attributable to recruitment strategy.

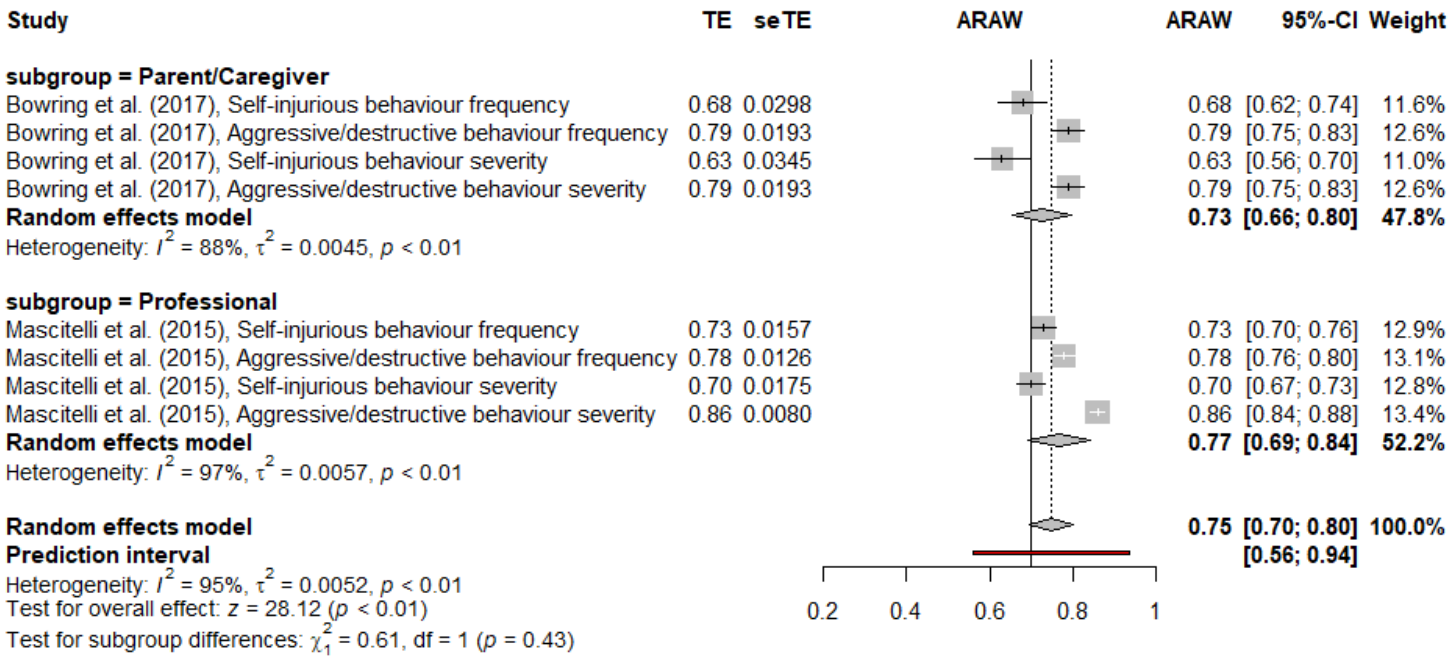


Appendices Figure 65. Subgroup forest plot assessing differences in the test-retest reliability of the BPI-01 attributable to informant completing the measure.



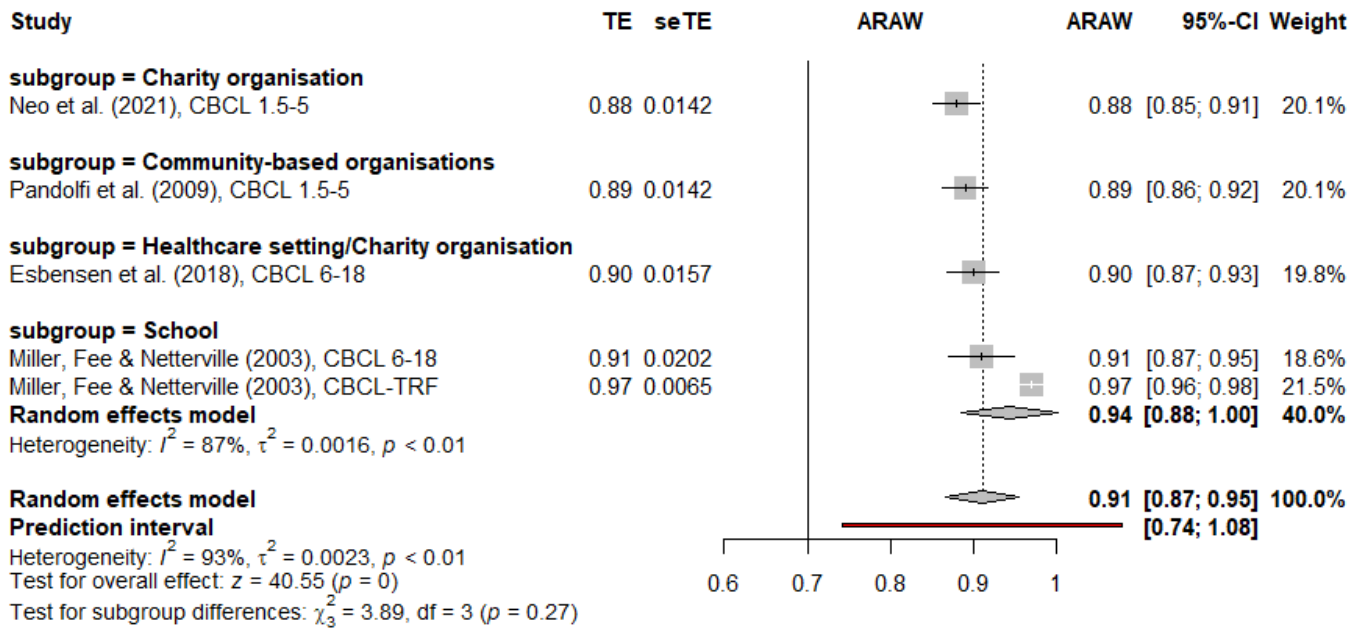
Appendices Figure 66. Subgroup forest plot assessing differences in the test-retest reliability of the BPI-01 attributable to ratings for children or adults.

Behavior Problems Inventory – Short Form (BPI-Short Form): internal consistency

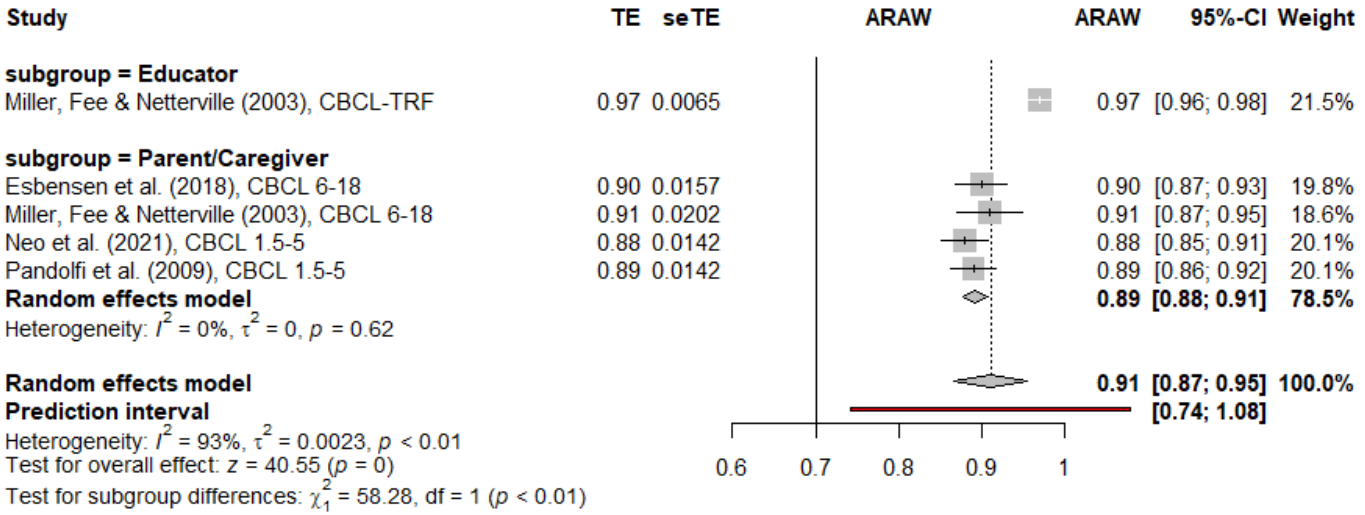


Appendices Figure 67. Subgroup forest plot assessing differences in the internal consistency of the BPI-Short form attributable to informant completing the measure.

Child Behavior Checklist (CBCL) *aggressive behaviour* scale: internal consistency

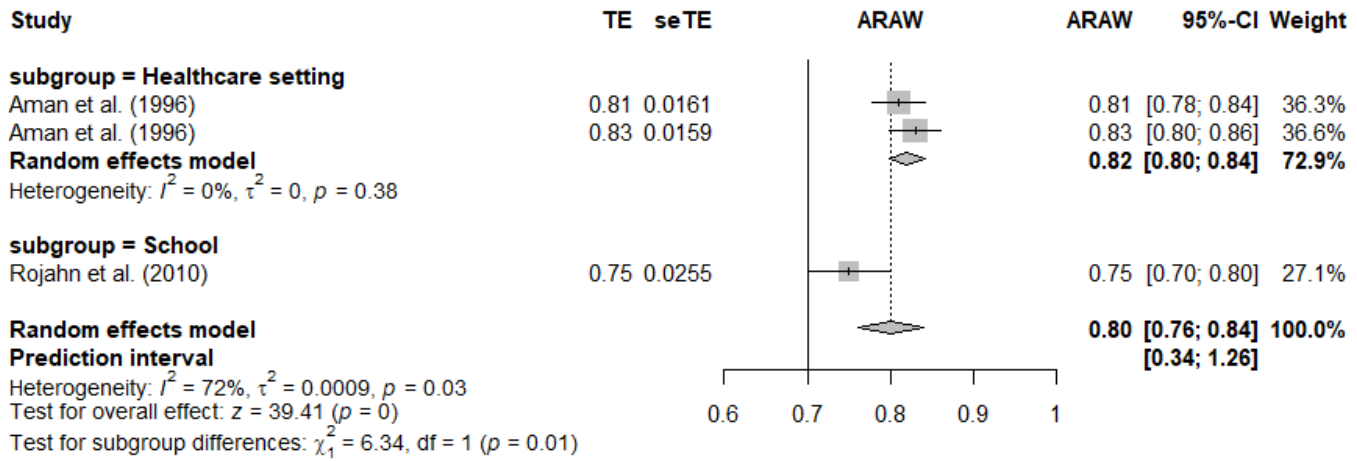


Appendices Figure 68. Subgroup forest plot assessing differences in the internal consistency of the versions of the CBCL attributable to recruitment strategy.

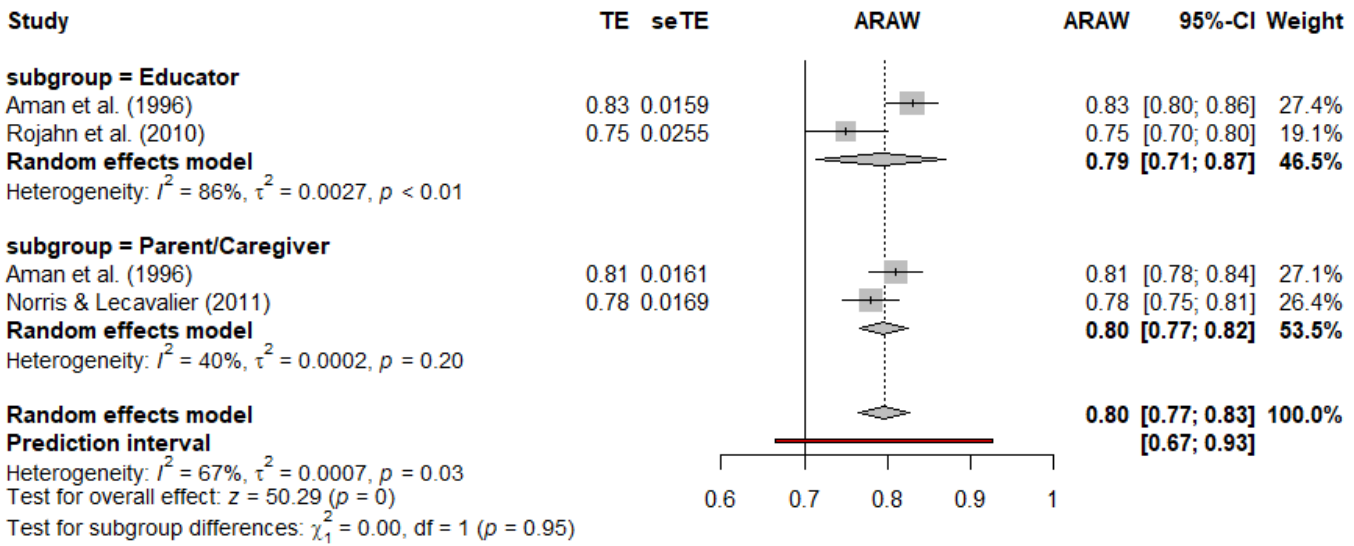


Appendices Figure 69. Subgroup forest plot assessing differences in the internal consistency of the versions of the CBCL attributable to informant completing the measure.

Nisonger Child Behavior Rating Form (NCBRF) *Self-injury/stereotypic behaviour* subscale: internal consistency

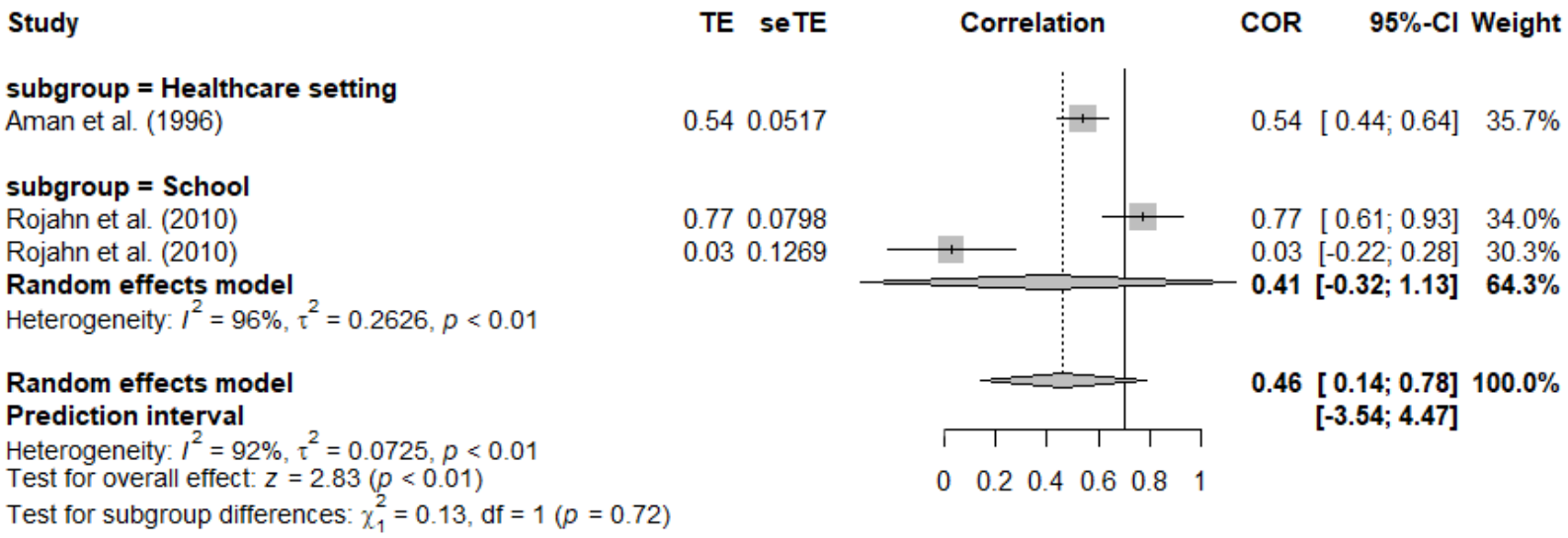


Appendices Figure 70. Subgroup forest plot assessing differences in the internal consistency of the NCBRF *self-injury/stereotypic* behaviour subscale attributable to recruitment strategy.

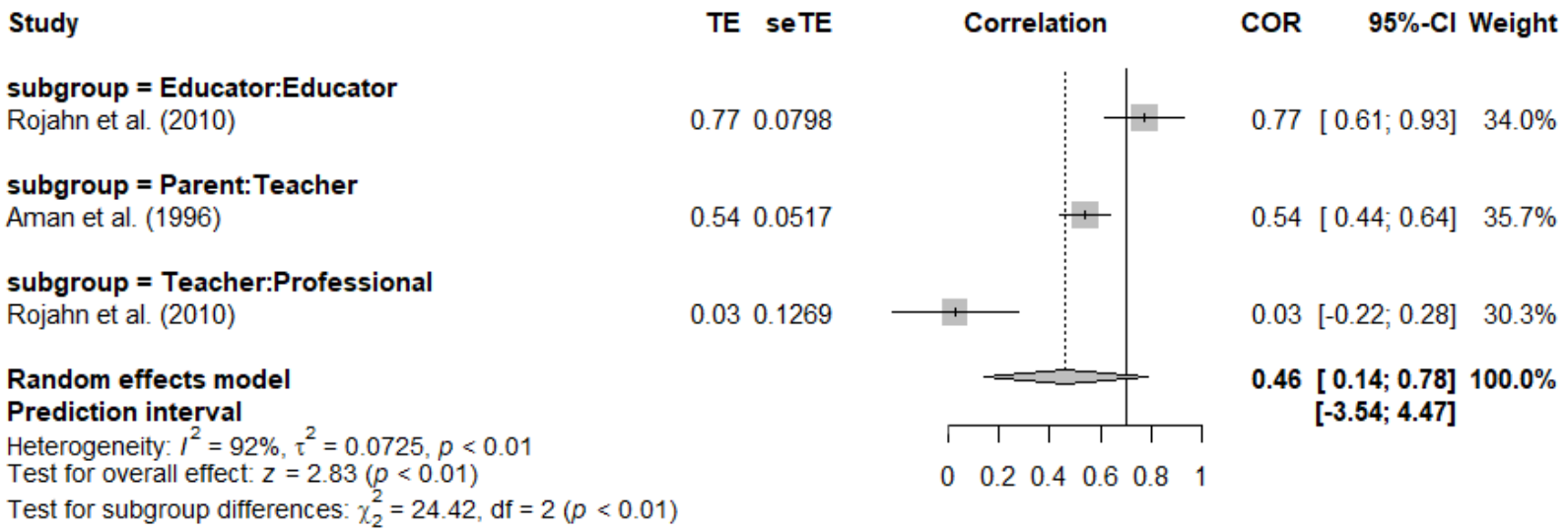


Appendices Figure 71. Subgroup forest plot assessing differences in the internal consistency of the NCBRF *self-injury/stereotypic* behaviour subscale attributable to informant completing the measure.

Nisonger Child Behavior Rating Form (NCBRF) *Self-injury/stereotypic behaviour* subscale: inter-rater reliability



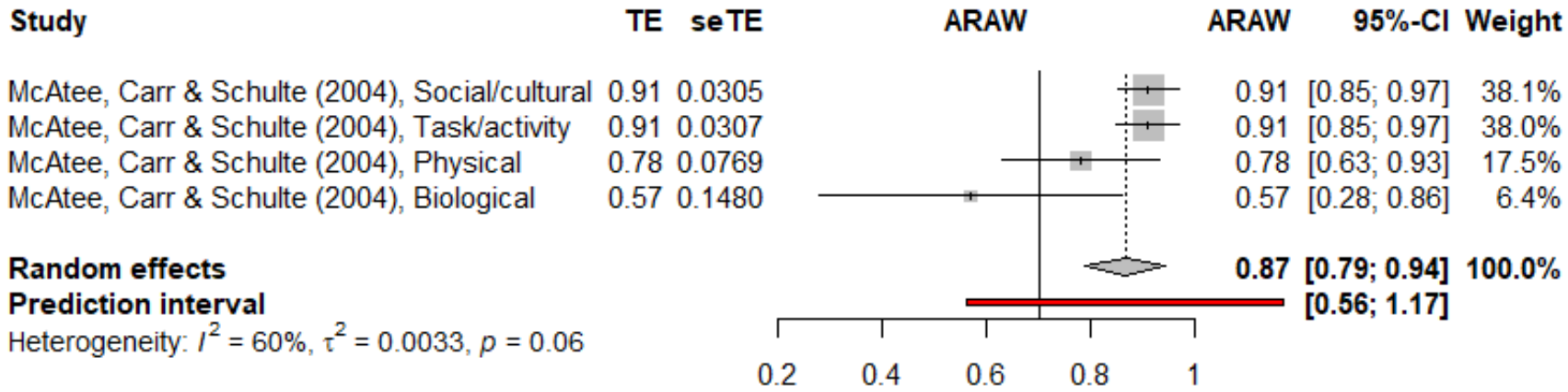
Appendices Figure 72. Subgroup forest plot assessing differences in the inter-rater reliability of the NCBRF *self-injury/stereotypic* behaviour subscale attributable to recruitment strategy.



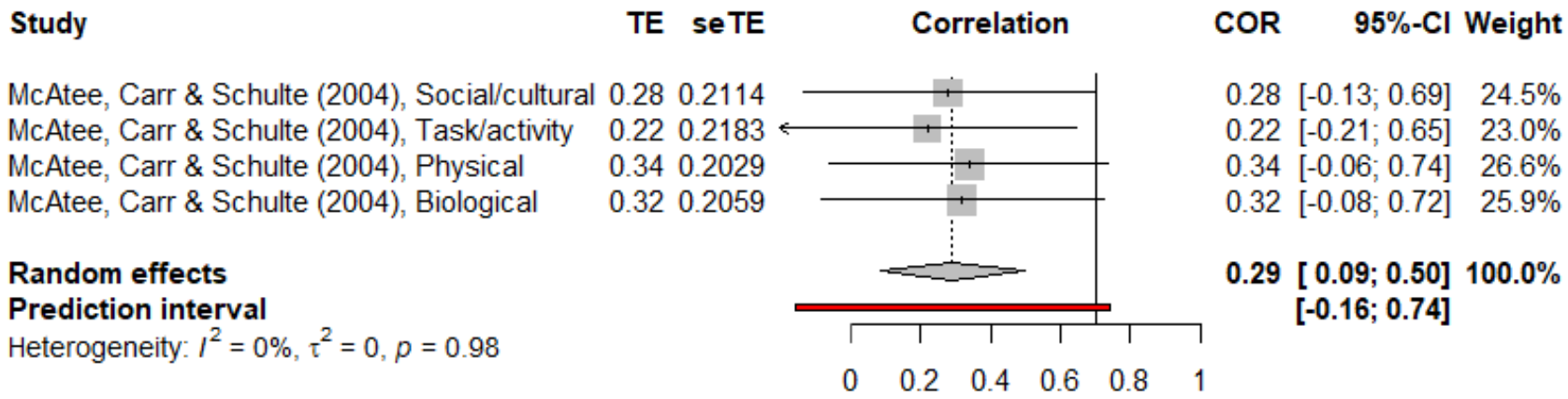
Appendices Figure 73. Subgroup forest plot assessing differences in the inter-rater reliability of the NCBRF *self-injury/stereotypic* behaviour subscale attributable to informant completing the measure.

Appendix 19: Chapter Three forest plots of IC, IRR and TRTR per measure of behavioural function

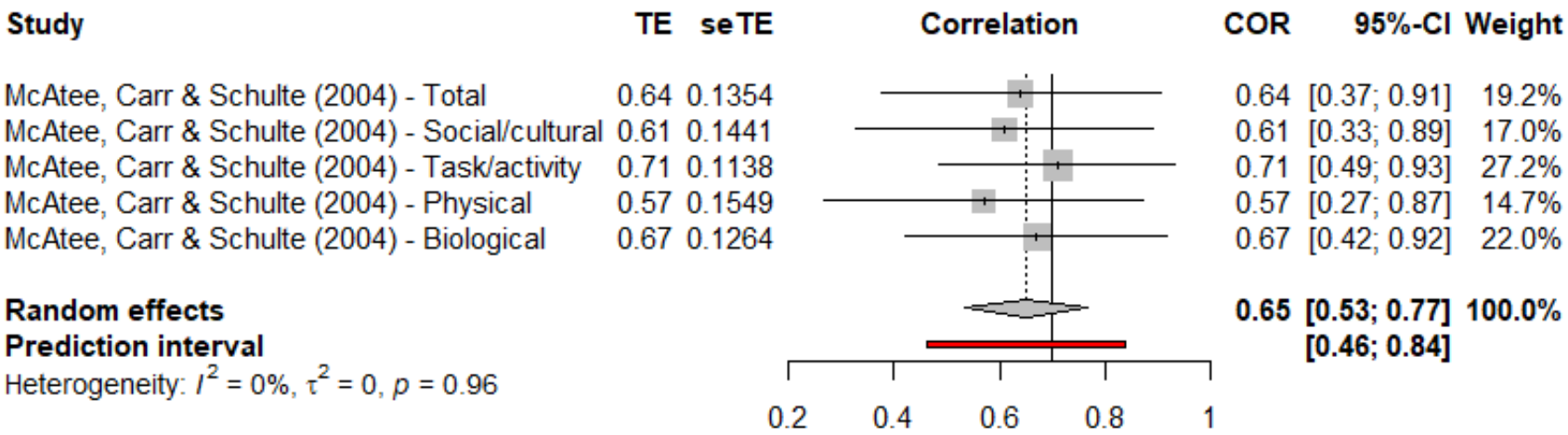
Contextual Assessment Inventory (CAI)



Appendices Figure 74. Forest plot for the internal consistency of the CAI using a random-effects model.

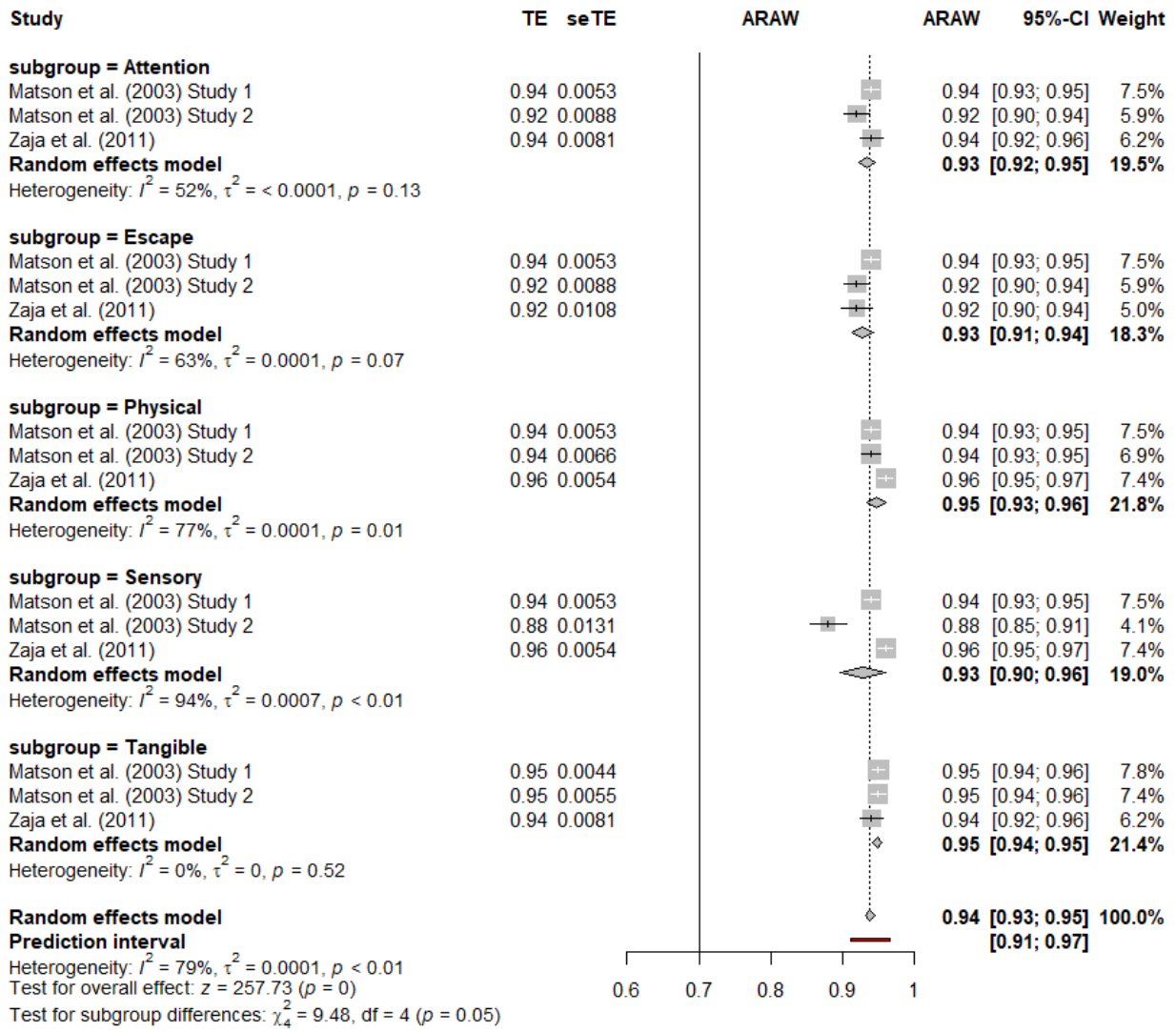


Appendices Figure 75. Forest plot for the inter-rater reliability of the CAI using a random-effects model.

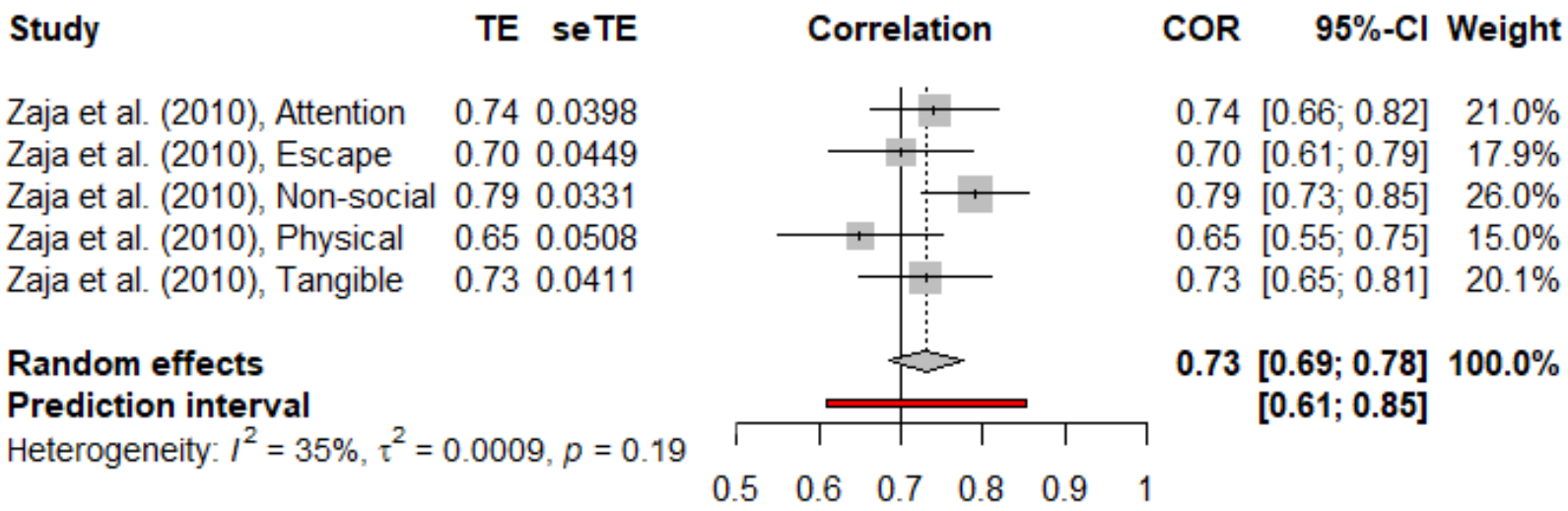


Appendices Figure 76. Forest plot for the test-retest reliability of the CAI using a random-effects model.

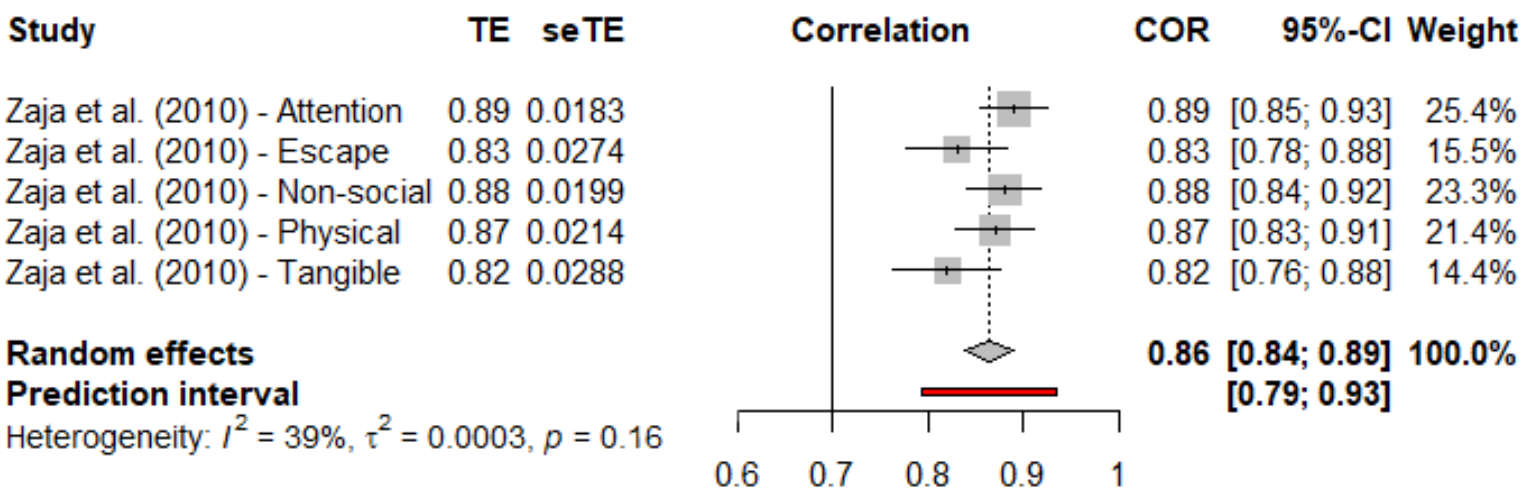
Functional Assessment for multiple CausaliTy (FACT)



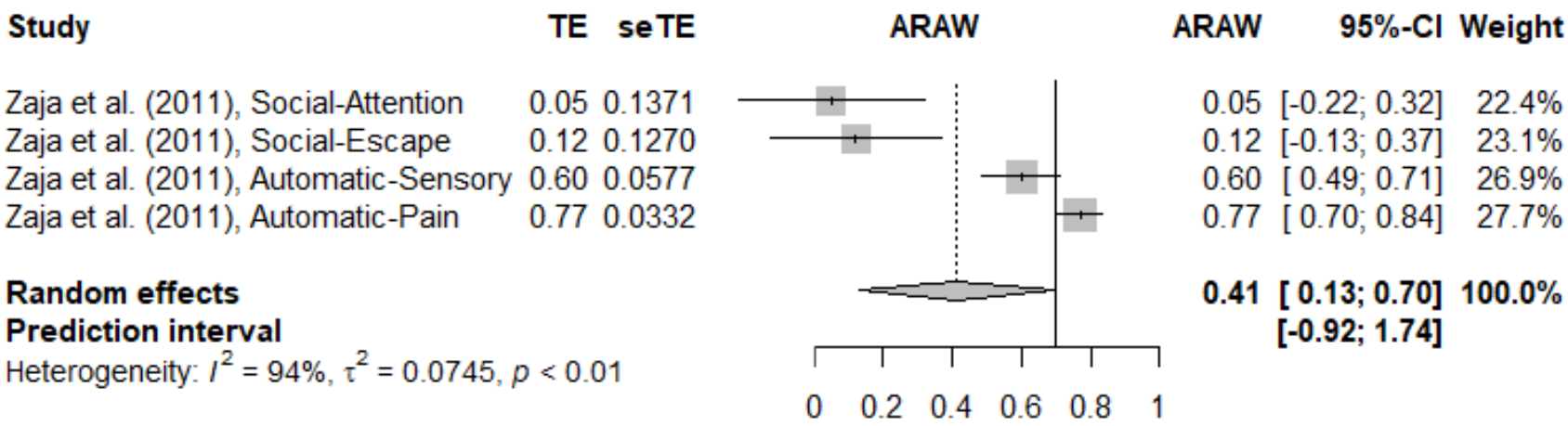
Appendices Figure 77. Forest plot for the internal consistency of the FACT using a random-effects model.



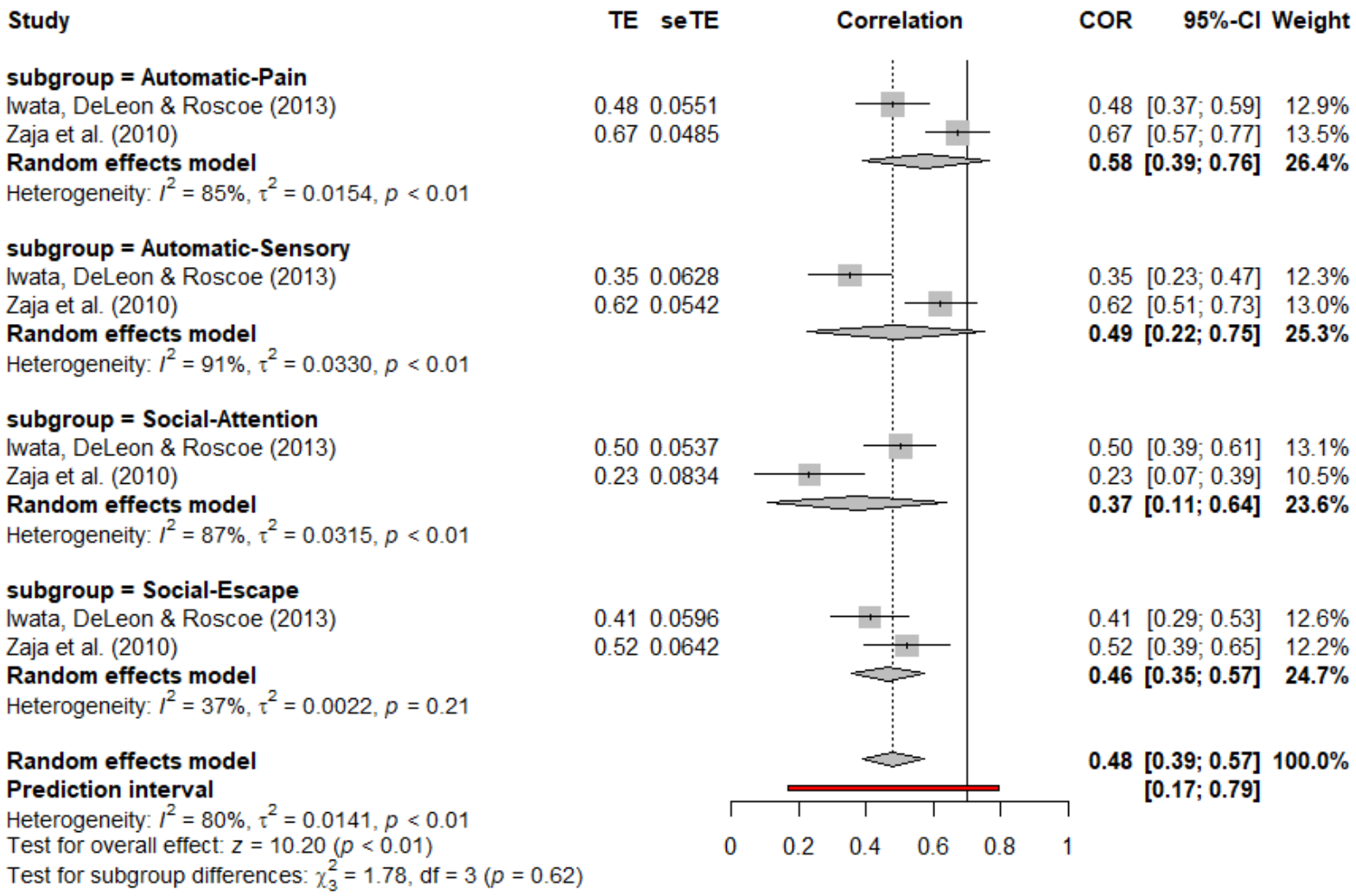
Appendices Figure 78. Forest plot for the inter-rater reliability of the FACT using a random-effects model.



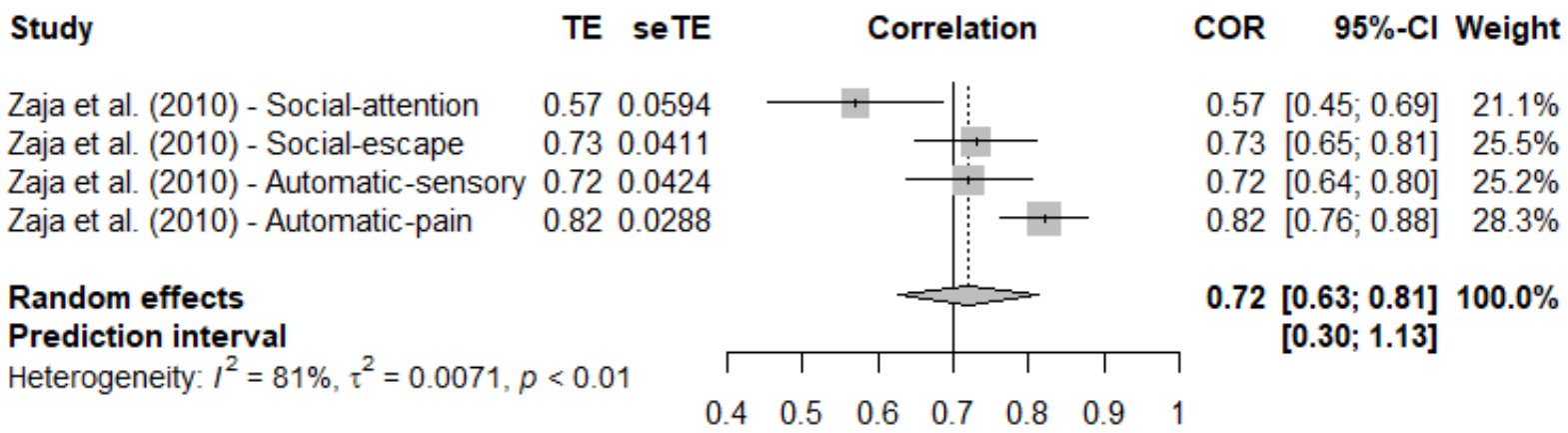
Appendices Figure 79. Forest plot for the test-retest reliability of the FACT using a random-effects model.



Appendices Figure 80. Forest plot for the internal consistency of the FAST using a random-effects model.

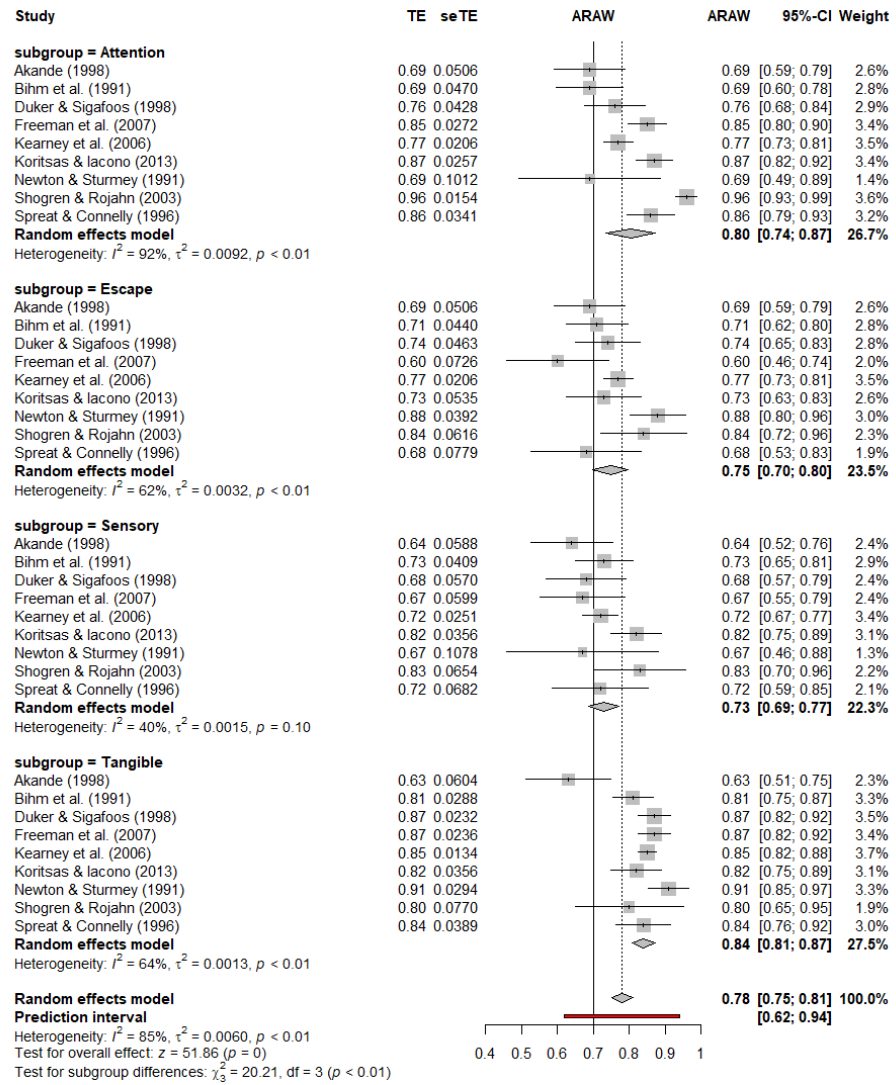


Appendices Figure 81. Forest plot for the inter-rater reliability of the FAST using a random-effects model.

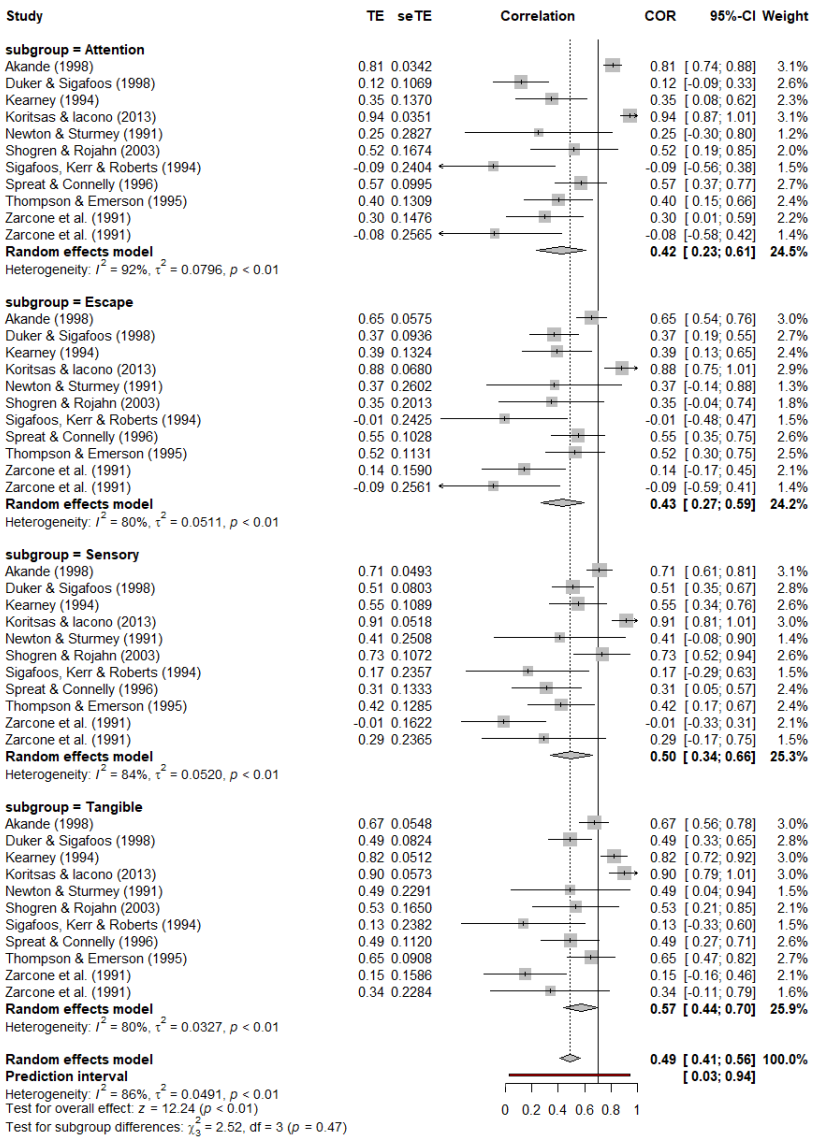


Appendices Figure 82. Forest plot for the test-retest reliability of the FAST using a random-effects model.

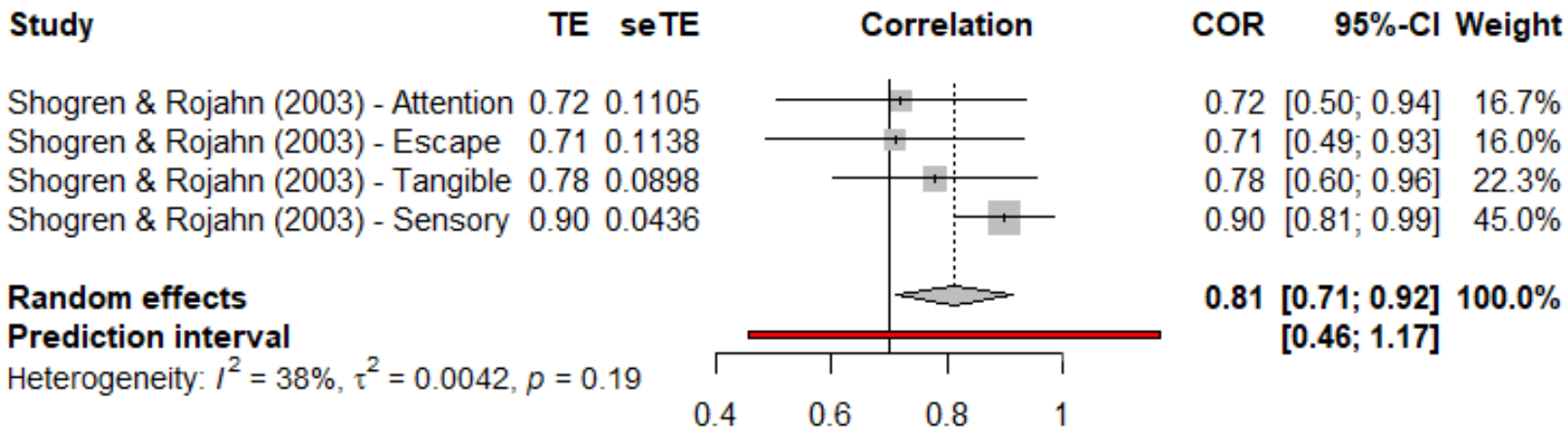
Motivation Assessment Scale (MAS)



Appendices Figure 83. Forest plot for the internal consistency of the MAS using a random-effects model.

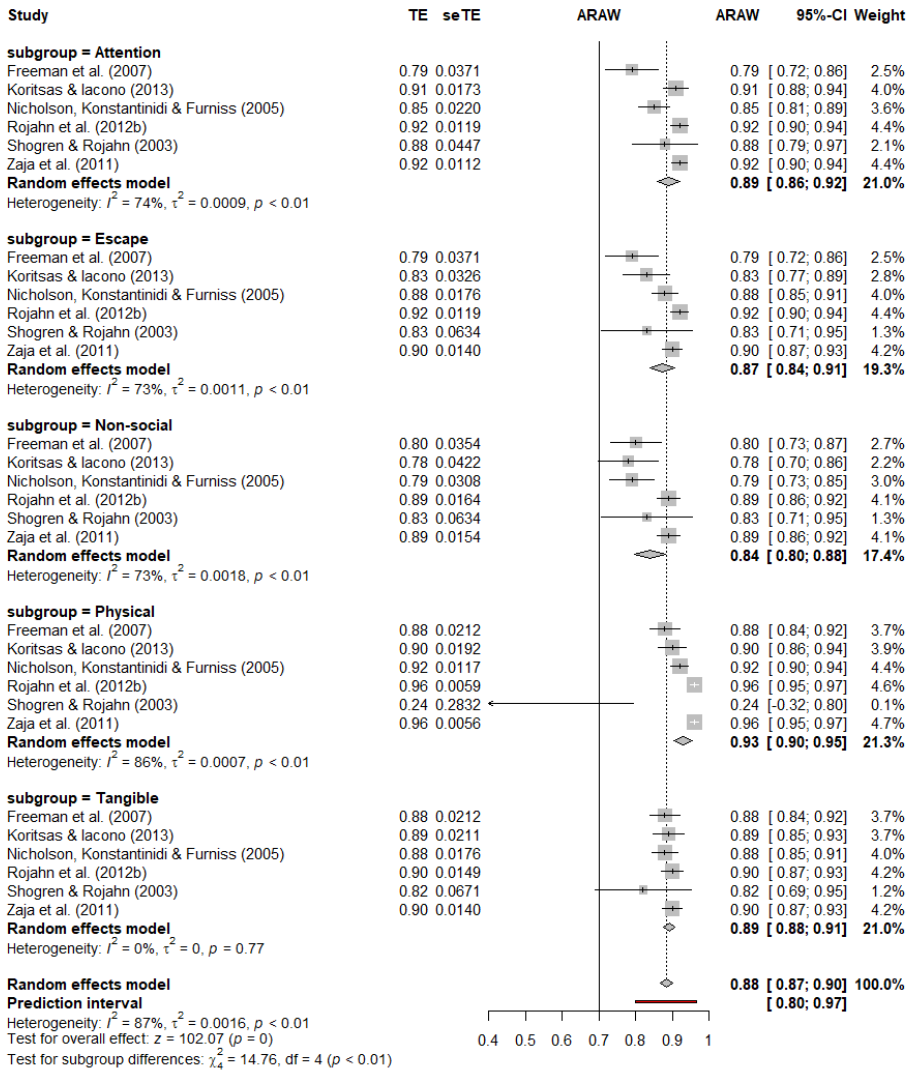


Appendices Figure 84. Forest plot for the inter-rater reliability of the MAS using a random-effects model.

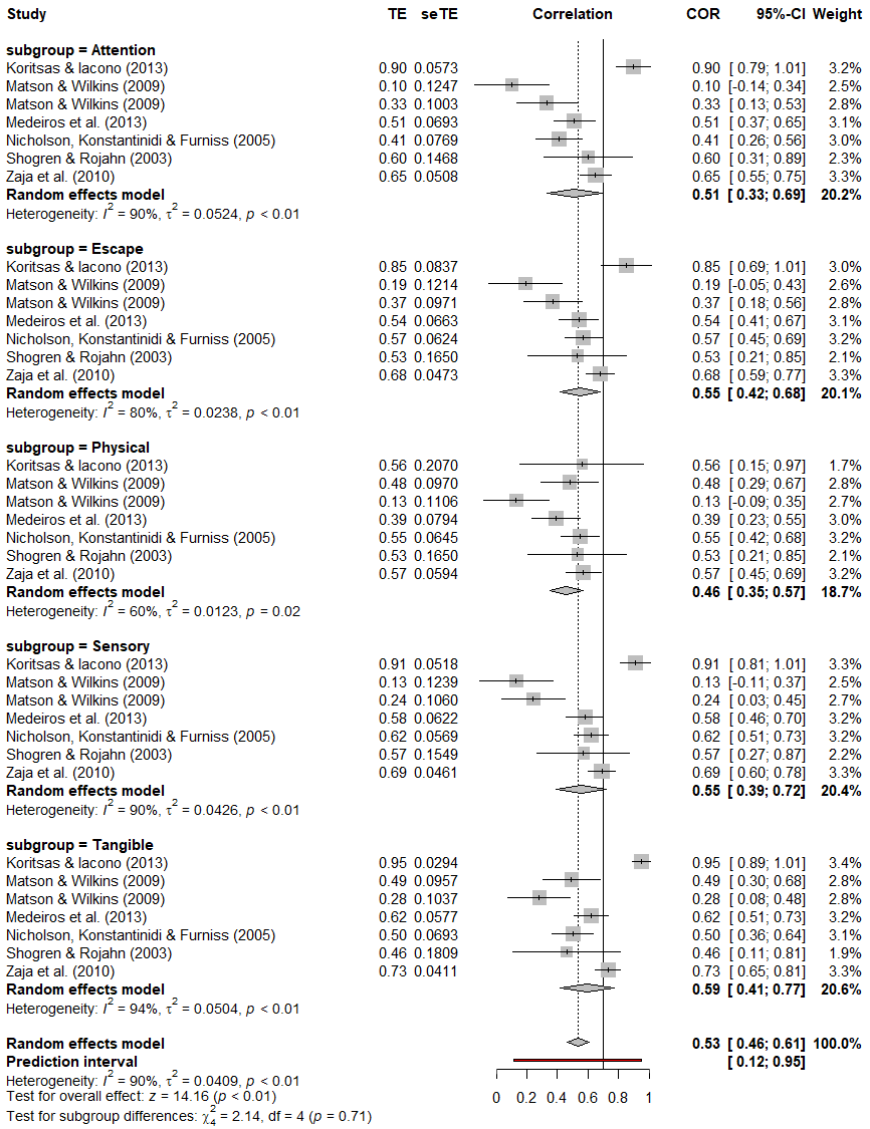


Appendices Figure 85. Forest plot for the test-retest reliability of the MAS using a random-effects model.

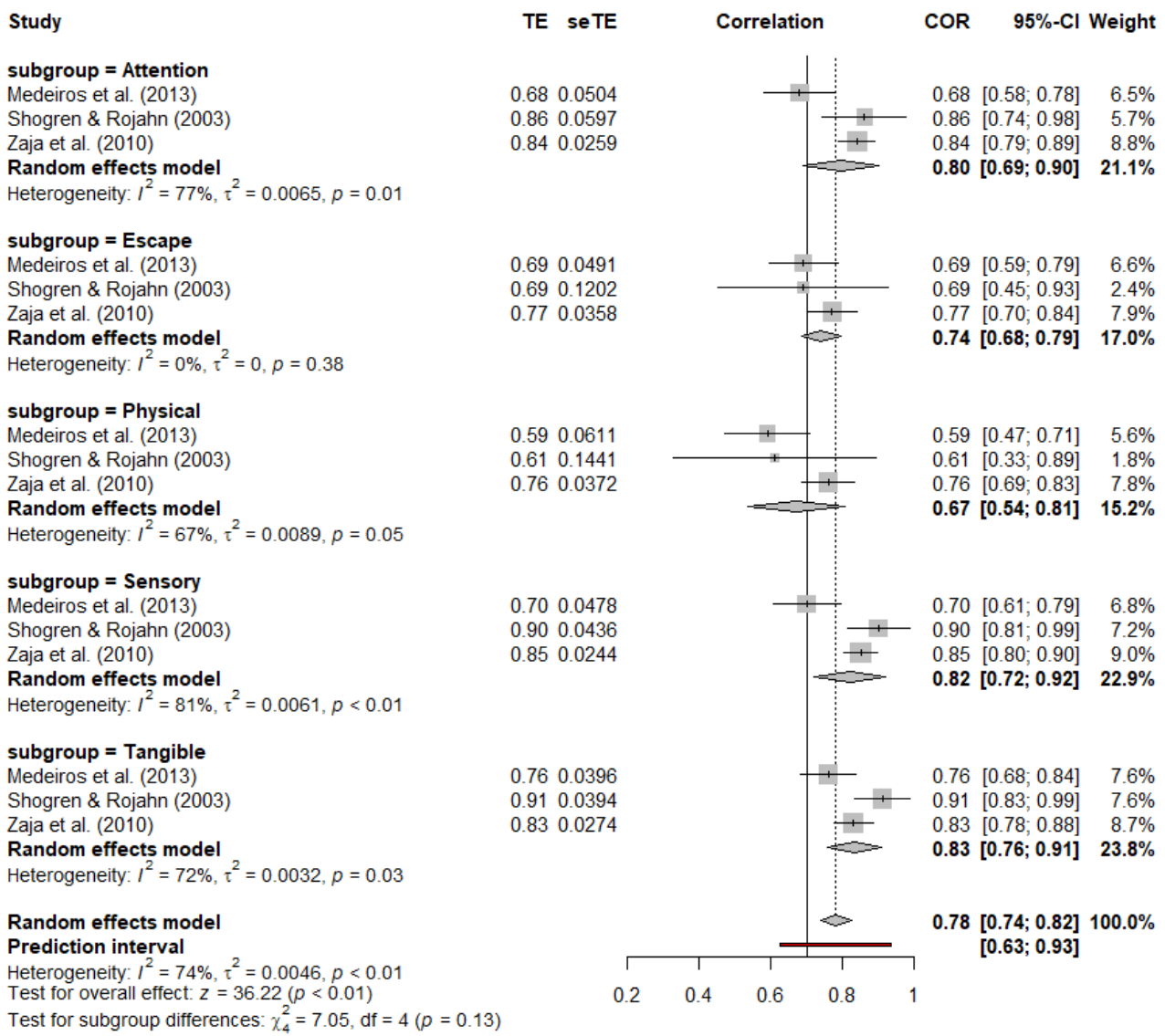
Questions About Behavioral Function (QABF)



Appendices Figure 86. Forest plot for the internal consistency of the QABF using a random-effects model.

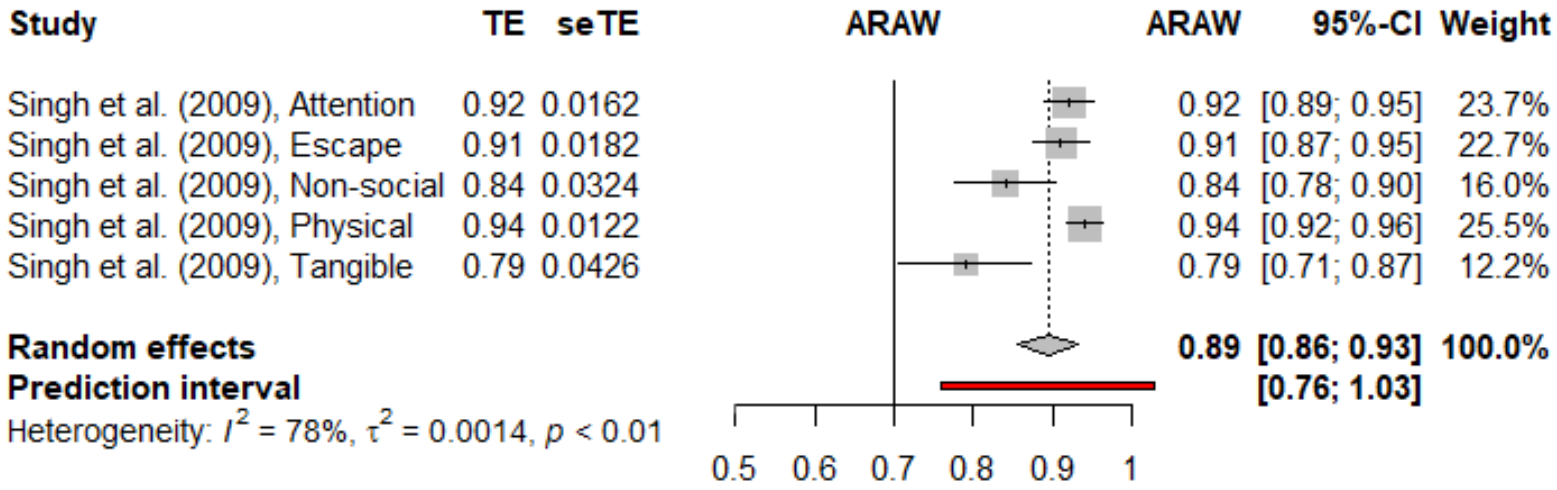


Appendices Figure 87. Forest plot for the inter-rater reliability of the QABF using a random-effects model.

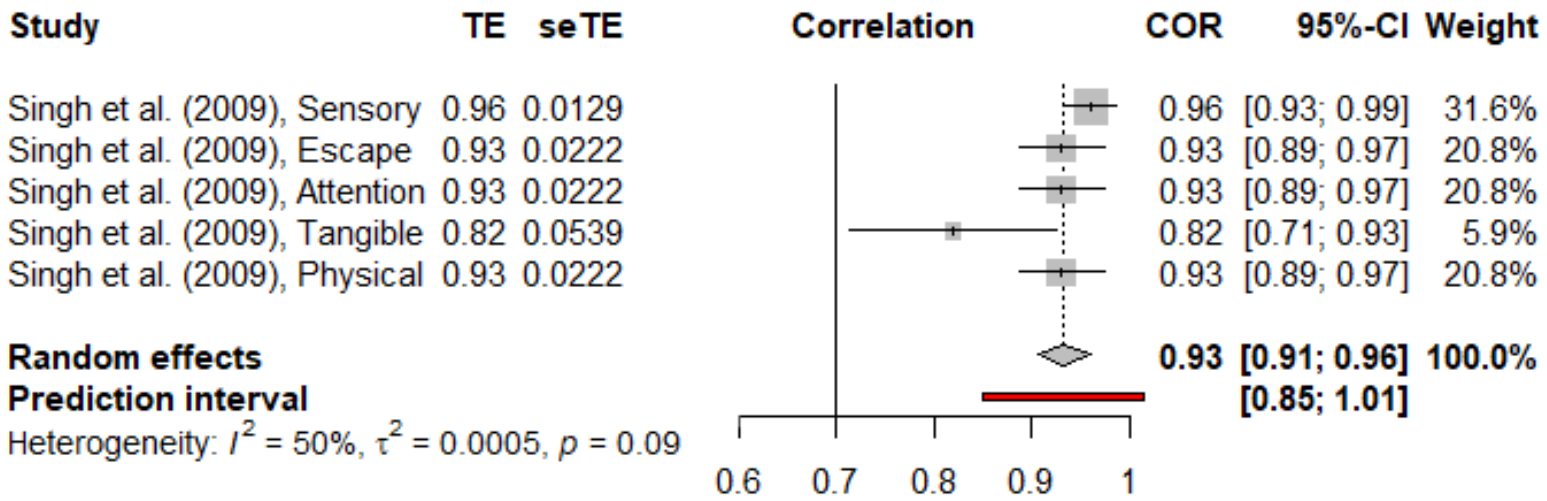


Appendices Figure 88. Forest plot for the test-retest reliability of the QABF using a random-effects model.

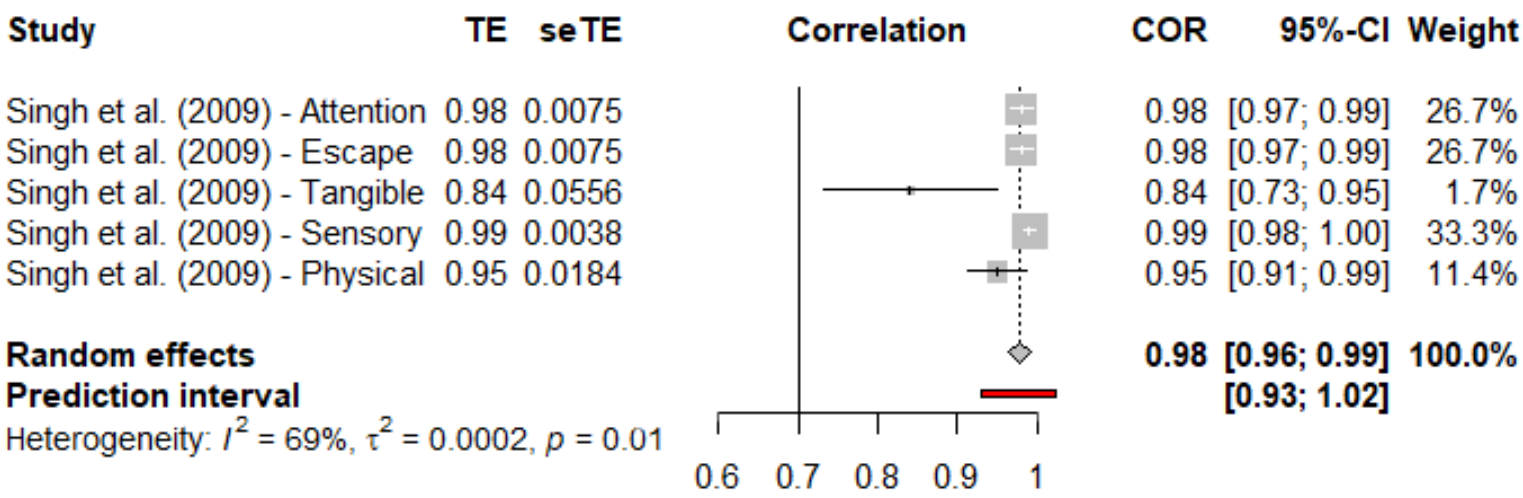
Questions About Behavioral Function – Short Form (QABF-Short Form)



Appendices Figure 89. Forest plot for the internal consistency of the QABF-Short Form using a random-effects model.



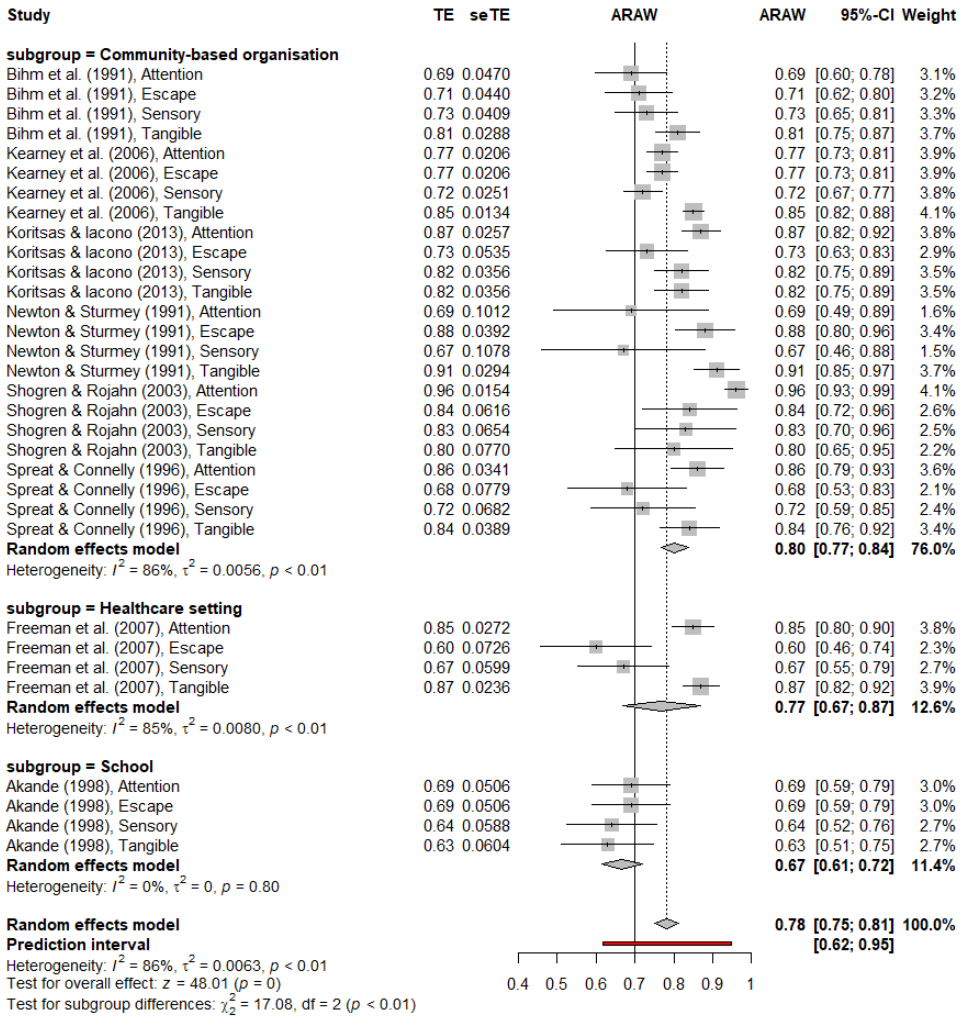
Appendices Figure 90. Forest plot for the inter-rater reliability of the QABF-Short Form using a random-effects model.



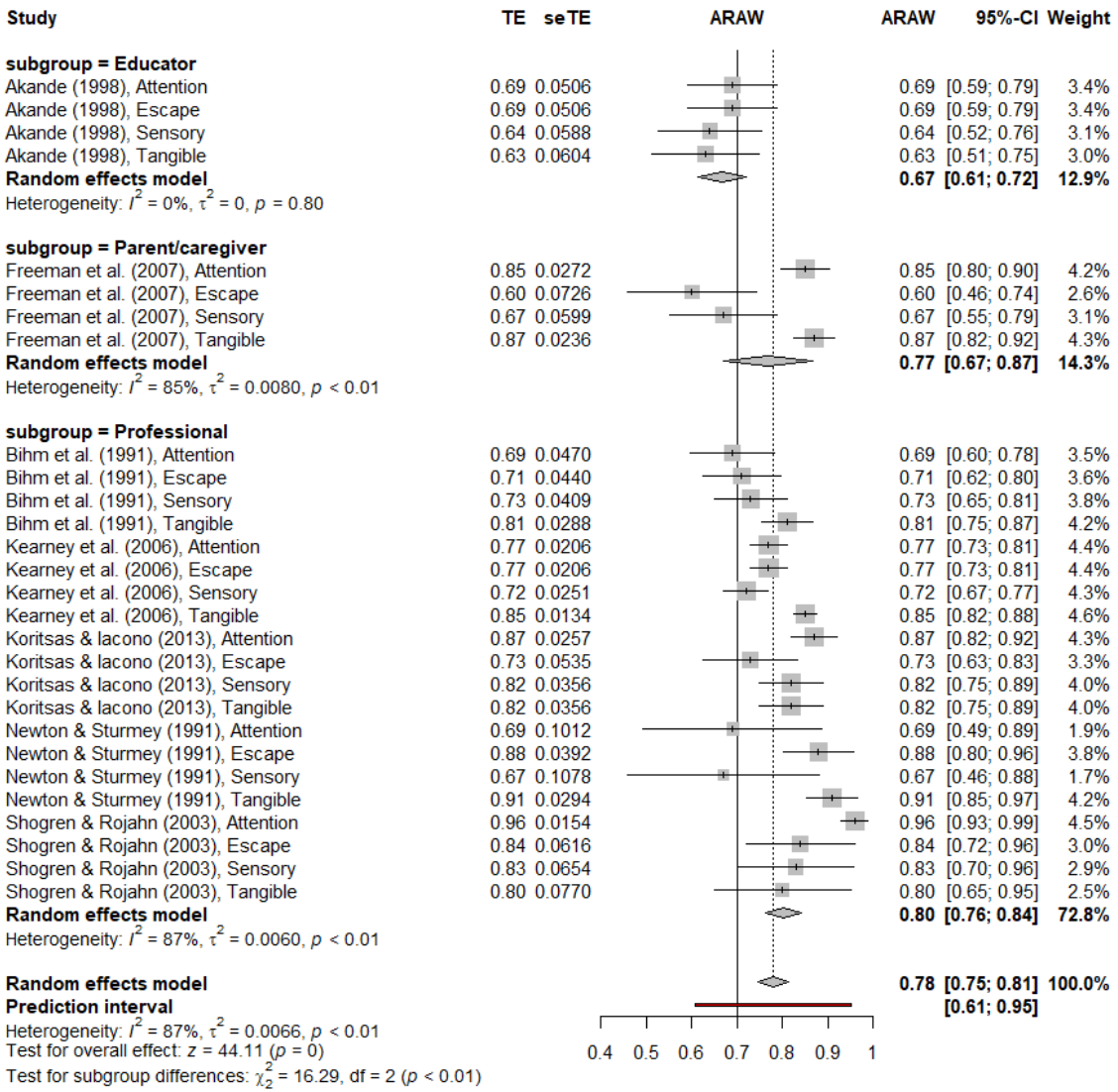
Appendices Figure 91. Forest plot for the test-retest reliability of the QABF-Short Form using a random-effects model.

Appendix 20: Chapter Three subgroup analysis forest plots per measure of behavioural function

Motivation Assessment Scale (MAS): Internal Consistency

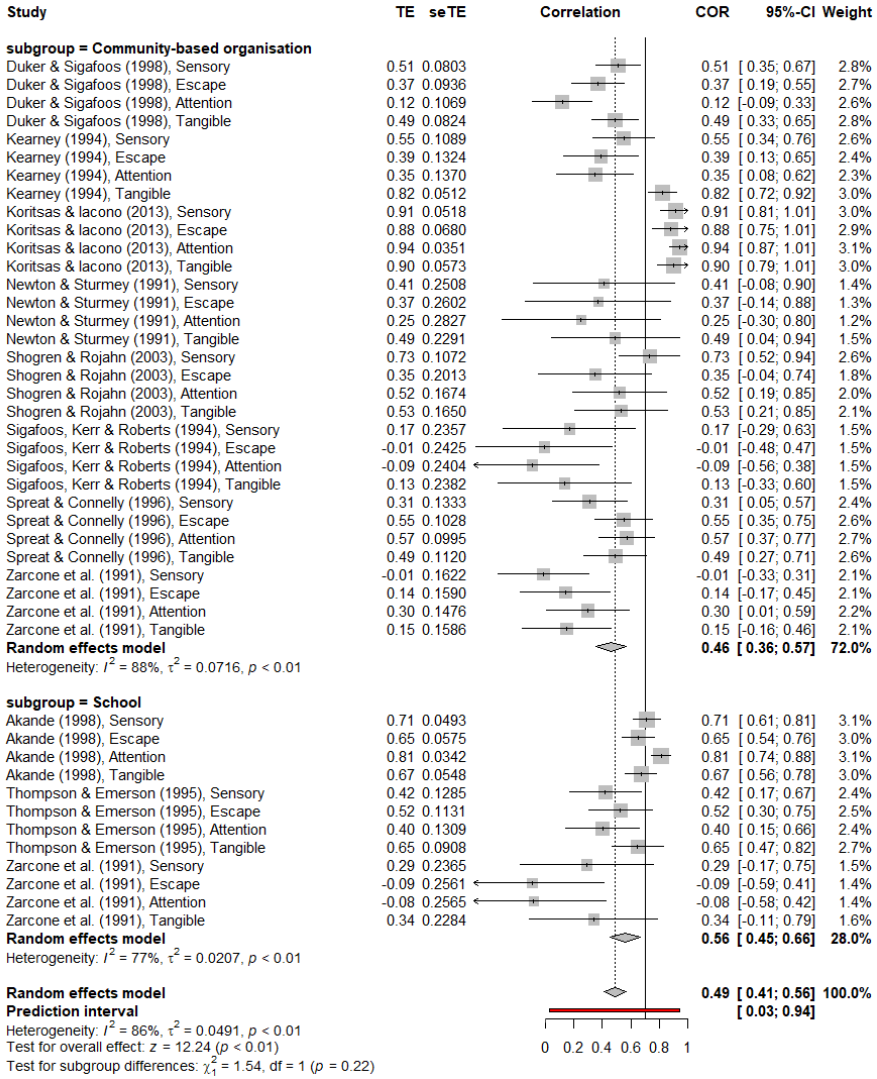


Appendices Figure 92. Subgroup forest plot assessing differences in the internal consistency of the MAS attributable to recruitment strategy.

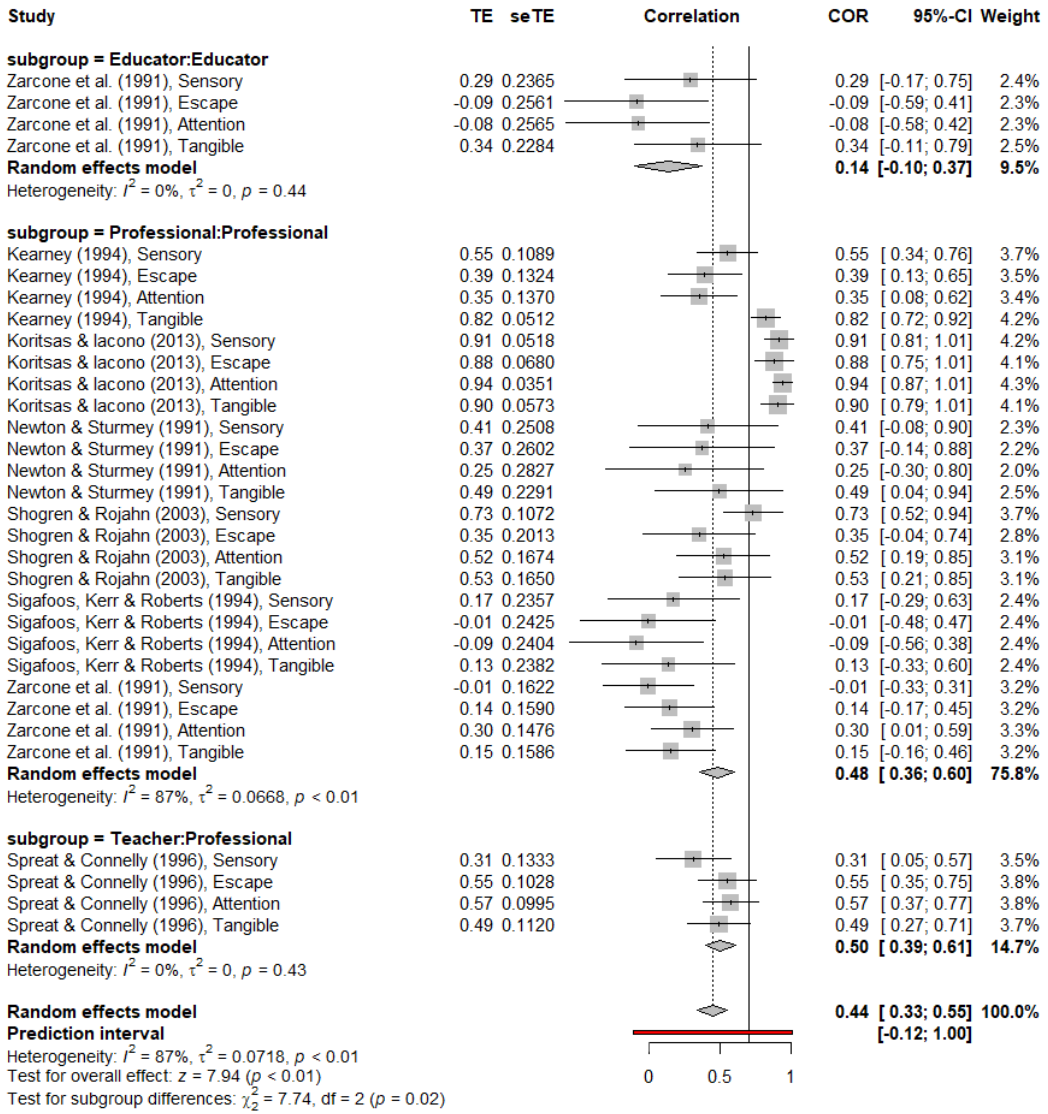


Appendices Figure 93. Subgroup forest plot assessing differences in the internal consistency of the MAS attributable to informant completing the measure.

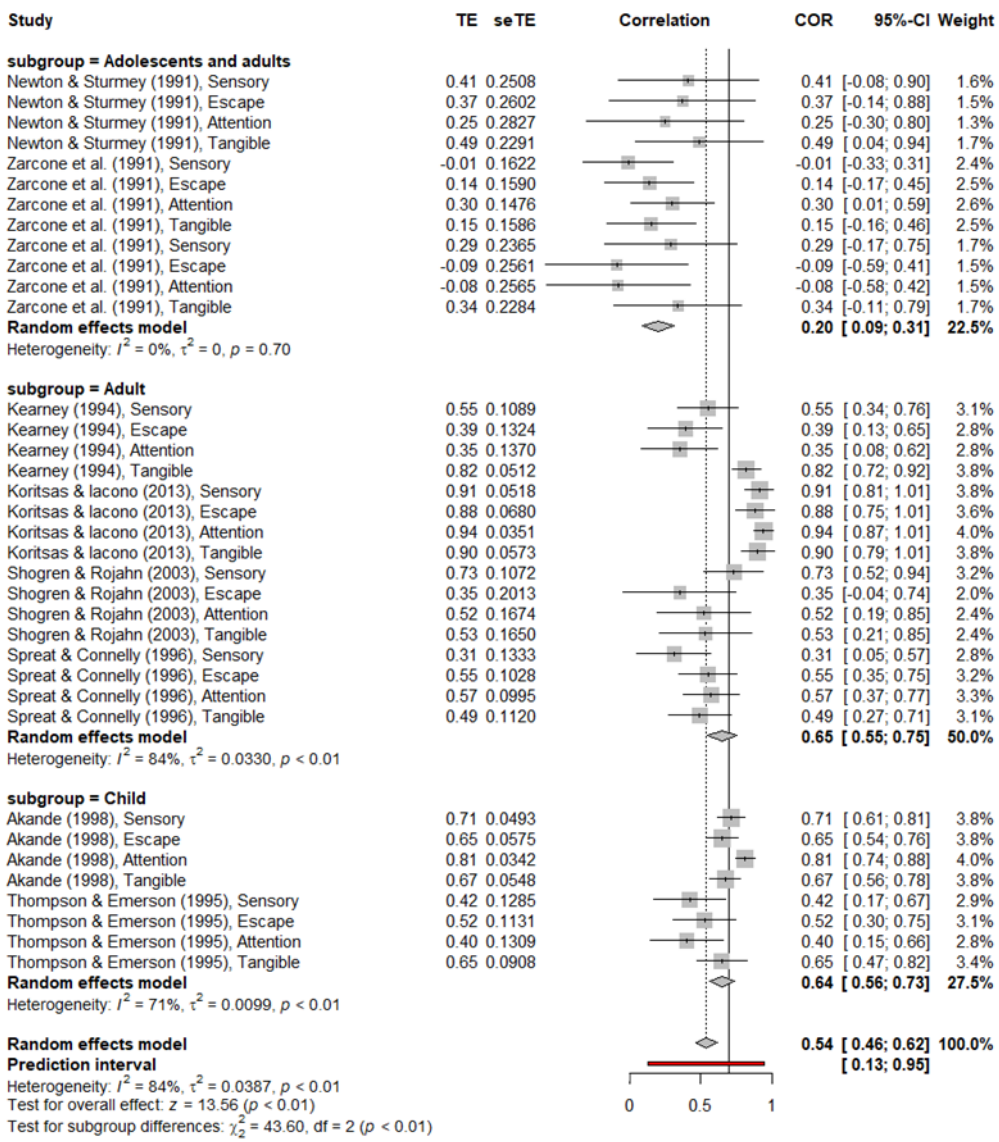
Motivation Assessment Scale (MAS): inter-rater reliability



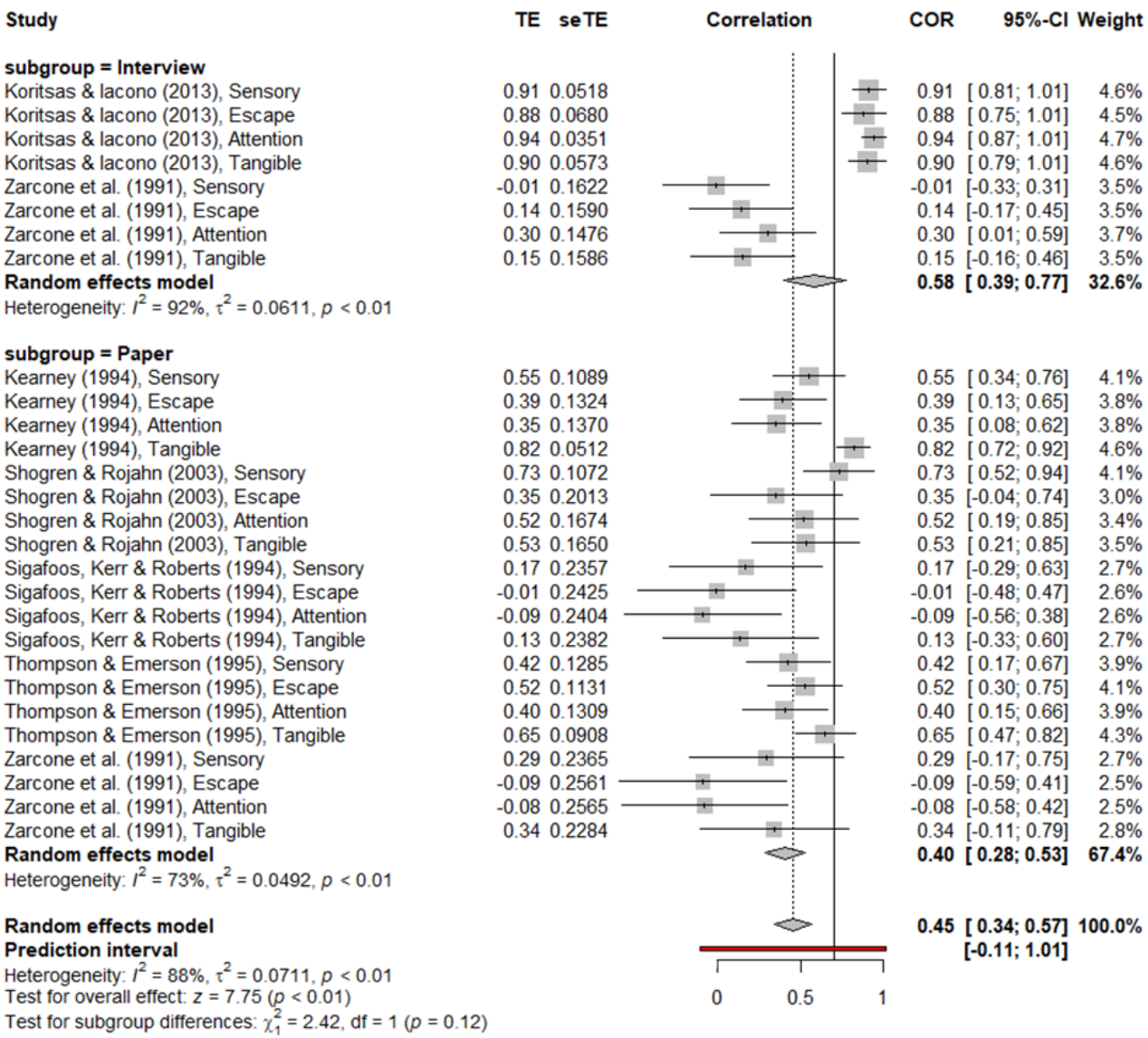
Appendices Figure 94. Subgroup forest plot assessing differences in the inter-rater reliability of the MAS attributable to recruitment strategy.



Appendices Figure 95. Subgroup forest plot assessing differences in the inter-rater reliability of the MAS attributable to informant pairs completing the measure.

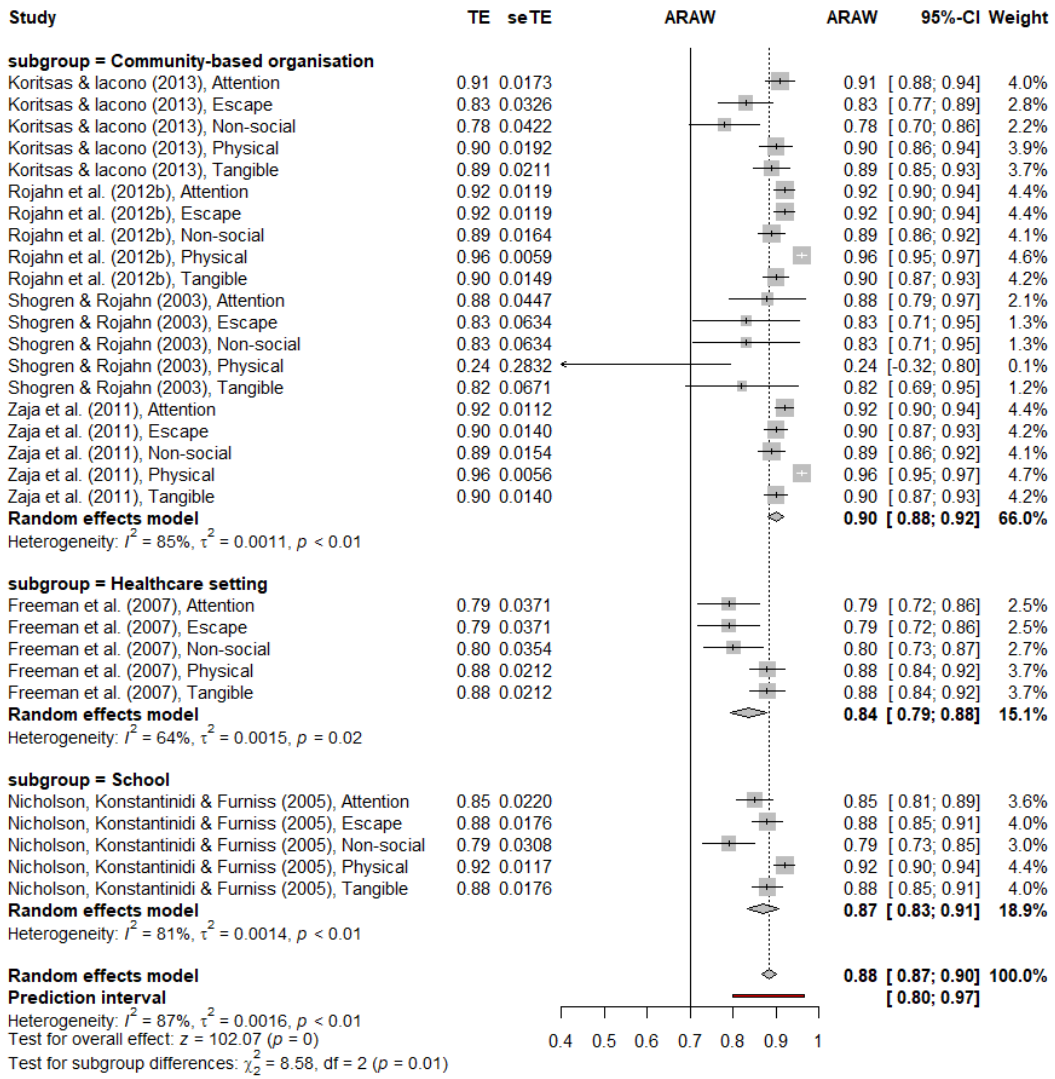


Appendices Figure 96. Subgroup forest plot assessing differences in the inter-rater reliability of the MAS attributable to ratings for children or adults.

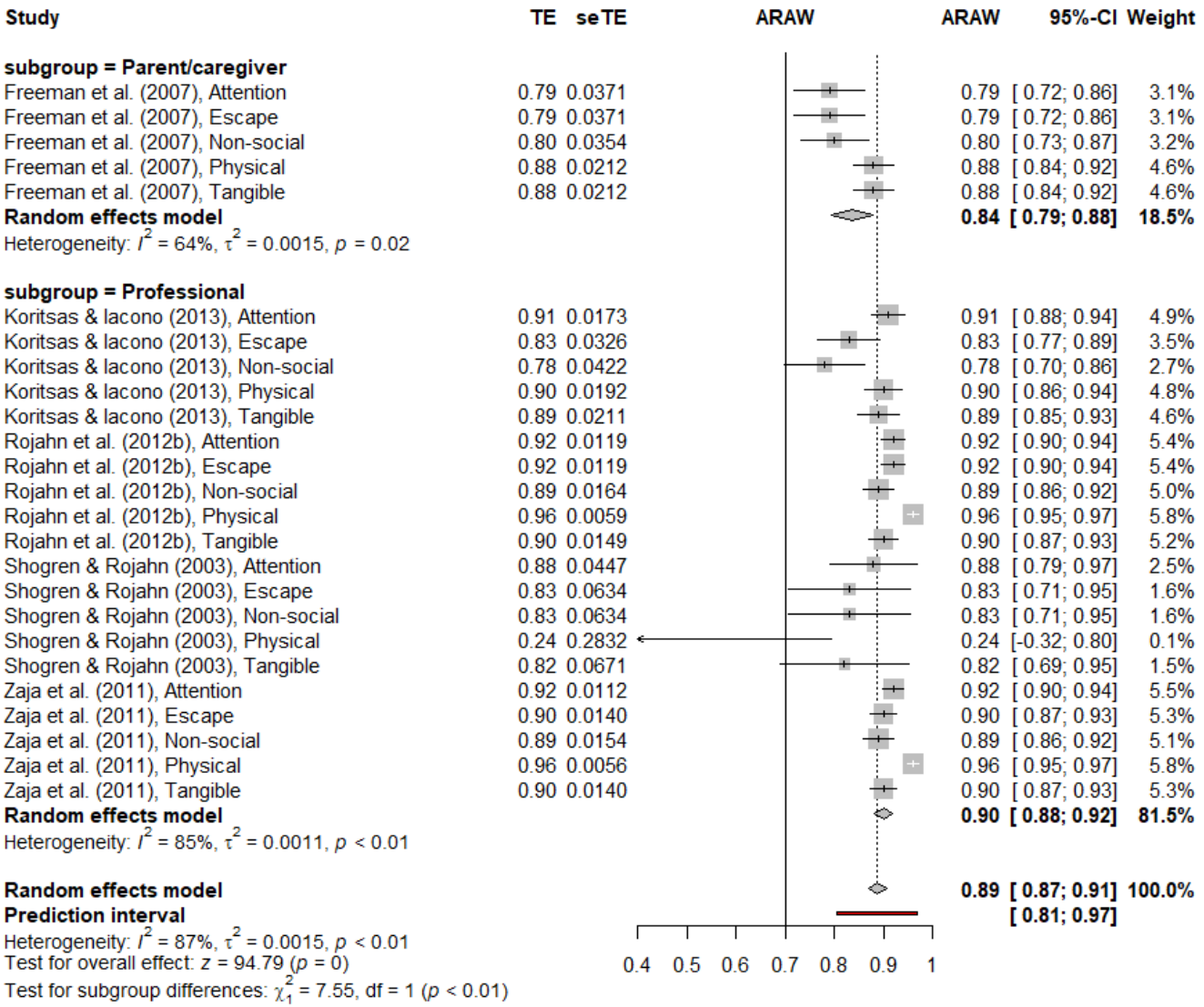


Appendices Figure 97. Subgroup forest plot assessing differences in the inter-rater reliability of the MAS attributable to method of administration.

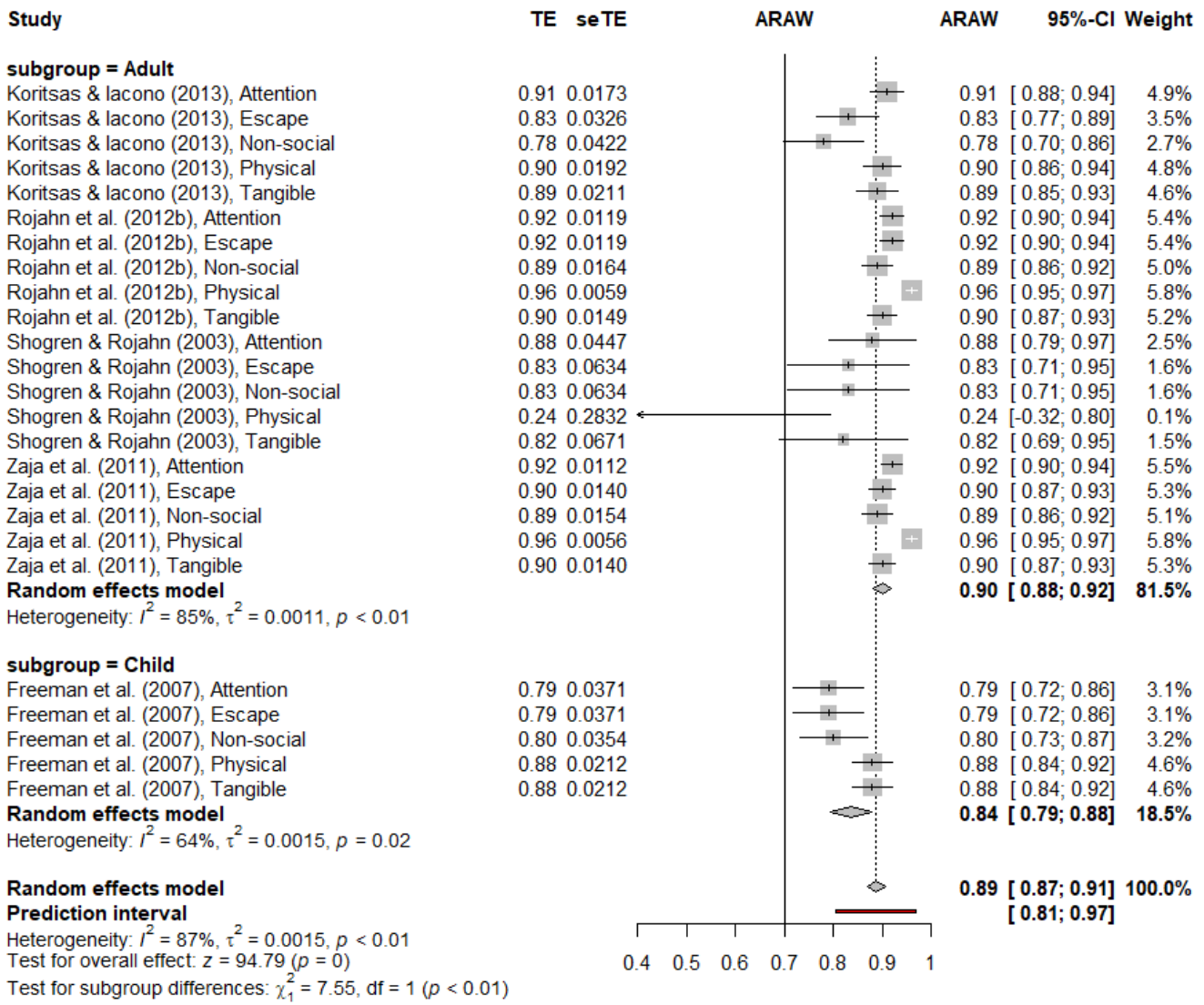
Questions About Behavioural Function (QABF): internal consistency



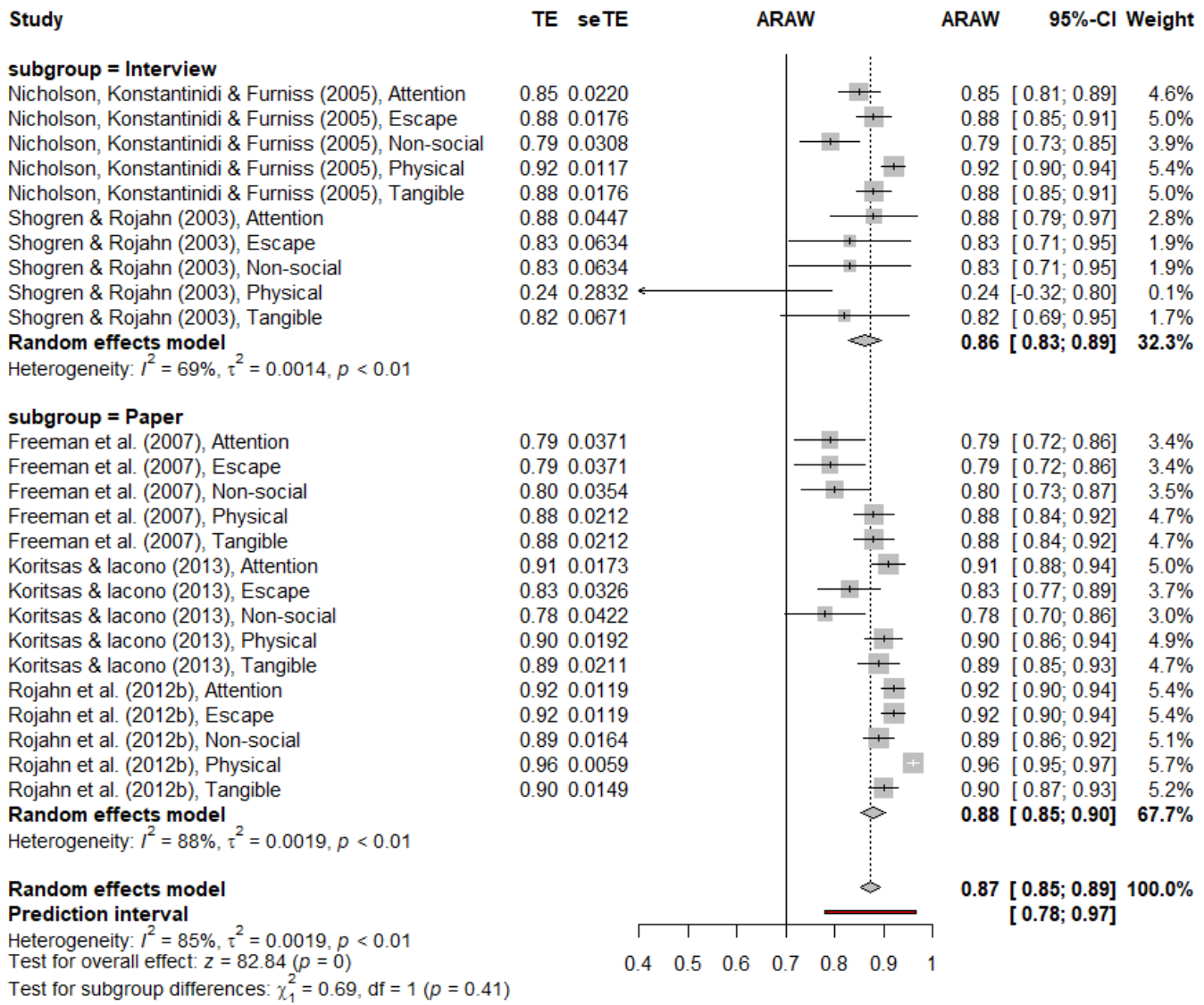
Appendices Figure 98. Subgroup forest plot assessing differences in the internal consistency of the QABF attributable to recruitment strategy.



Appendices Figure 99. Subgroup forest plot assessing differences in the internal consistency of the QABF attributable to informant completing the measure.



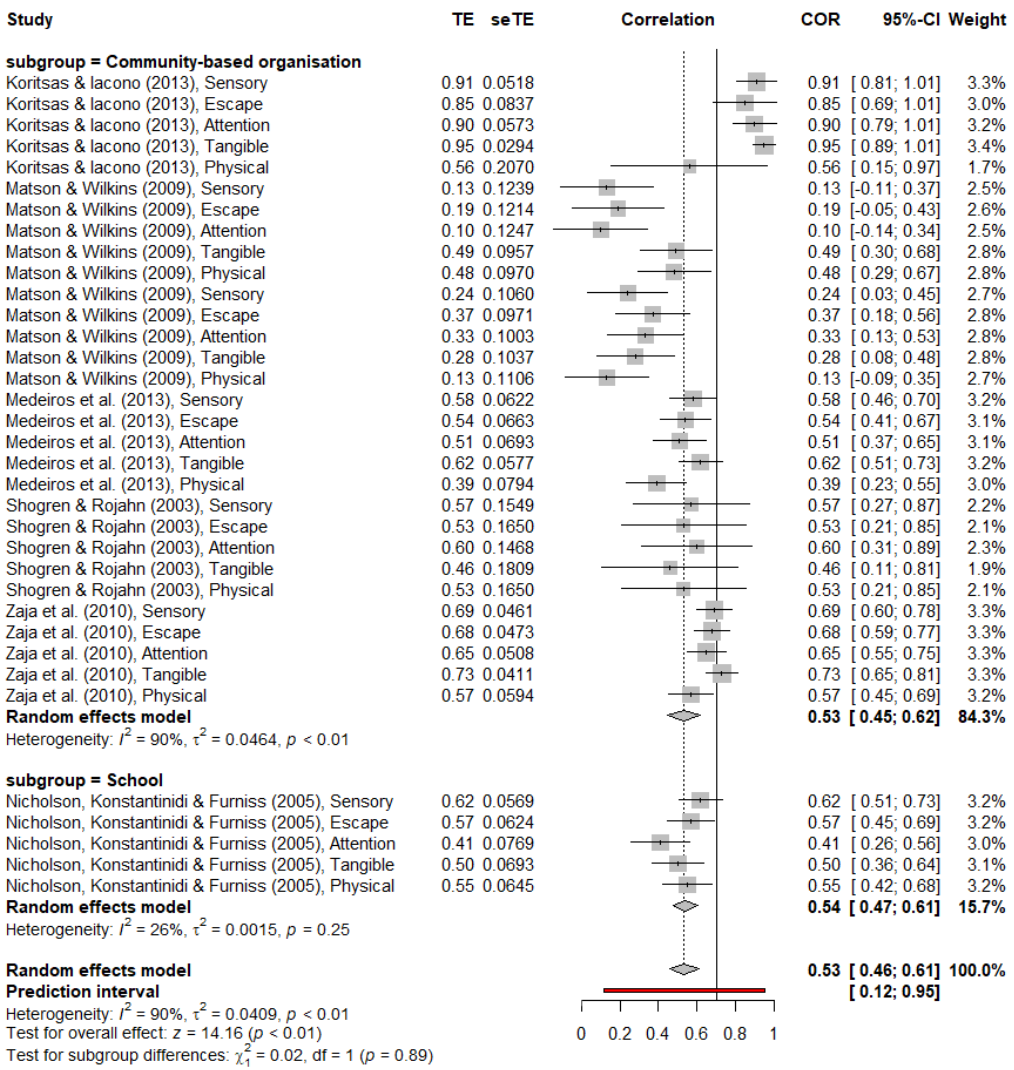
Appendices Figure 100. Subgroup forest plot assessing differences in the internal consistency of the QABF attributable to ratings for children or adults.



Appendices Figure 101. Subgroup forest plot assessing differences in the internal consistency of the QABF attributable to the method of administration.

Appendix Twenty

Questions About Behavioural Function (QABF): inter-rater reliability



Appendices Figure 102. Subgroup forest plot assessing differences in the inter-rater reliability of the QABF attributable to recruitment strategy.

Appendix 21: Chapter Three references for all studies included in the meta-analyses

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Appendix 22: Ethical approval for Chapters Four (interview study) and Five (remote assessment study)

Health Research Authority

London - Brighton & Sussex Research Ethics Committee
Health Research Authority
2 Redman Place
Stratford
London
E20 1JQ

Telephone: 0207 104 8241

Please note: This is an acknowledgement letter from the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

20 October 2021

Miss Lauren Shelley
Aston University
Aston Triangle
Birmingham
B4 7ET

Dear Miss Shelley

Study title:	**Behaviours that challenge in SATB2-associated syndrome**
REC reference:	**21/LO/0537**
Protocol number:	**008-2021-LS**
IRAS project ID:	**296378**

Thank you for your letter of 19 October 2021. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 30 September 2021

Documents received

The documents received were as follows:

Document	*Version*	*Date*
IRAS Checklist XML [Checklist_19102021]		19 October 2021
Other [Cover letter, summarising responses to favourable opinion with conditions]	-	14 October 2021
Participant consent form [Stage 2 declaration form for over 16's	3	14 October 2021

without capacity to consent]		
Participant consent form [Stage 2 consent form for over 16's able to consent parent/carer]	3	14 October 2021
Participant consent form [Stage 2 consent form for over 16's with capacity to consent]	3	14 October 2021
Participant consent form [Stage 1 declaration form for over 16's without capacity to consent]	3	14 October 2021
Participant consent form [Stage 1 consent form for under 16's with capacity to consent]	3	14 October 2021
Participant consent form [Stage 1 consent form for under 16's without capacity to consent]	3	14 October 2021
Participant consent form [Stage 1 consent form for consultees of over 16's without capacity to consent]	3	14 October 2021
Participant consent form [Stage 1 consent form for over 16's able to consent parent/carer]	3	14 October 2021
Participant consent form [Stage 1 consent form for over 16's with capacity to consent]	3	14 October 2021
Participant consent form [Stage 2 consent form for under 16's with capacity to consent]	3	14 October 2021
Participant consent form [Stage 2 consent form for under 16's without capacity to consent]	3	14 October 2021
Participant consent form [Stage 2 consent form for consultee's of over 16's without capacity to consent]	3	14 October 2021
Participant information sheet (PIS) [Stage 1 information sheet for parent/carer of child under 16]	3	14 October 2021
Participant information sheet (PIS) [Stage 1 information sheet for parent/carer of adult over 16]	3	14 October 2021
Participant information sheet (PIS) [Stage 2 information sheet for parent/carer of child under 16]	3	14 October 2021
Participant information sheet (PIS) [Stage 2 information sheet for parent/carer of adult over 16]	3	14 October 2021
Research protocol or project proposal [SATB2_protocol_14.10.2021_V3]	3	14 October 2021

Approved Documents

The final list of approved documentation for the study is therefore as follows:

Document	*Version*	*Date*
Copies of materials calling attention of potential participants to the research [Stage 1 expression of interest form]	1	12 February 2021
Copies of materials calling attention of potential participants to the research [Stage 2 expression of interest form]	1	12 February 2021
Copies of materials calling attention of potential participants to the research [Stage 1 study advert]	1	12 February 2021
Copies of materials calling attention of potential participants to the research [Stage 2 study advert]	1	12 February 2021
Covering letter on headed paper [Cover letter_IRAS 296378_22.06.2021]		22 June 2021
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Aston-PI]		01 August 2020

GP/consultant information sheets or letters [GP letter notifying them of involvement in study]	1	12 February 2021
GP/consultant information sheets or letters [Letter to GP notifying them of distress of person with SAS]	1	12 February 2021
Interview schedules or topic guides for participants [Semi-structured Interview Template Schedule]	1	12 February 2021
Interview schedules or topic guides for participants [Template Interview Coding]	1	12 February 2021
IRAS Checklist XML [Checklist_19102021]		19 October 2021
Letter from funder [Funding letter]		10 July 2020
Letter from sponsor [Approval in Principle]		15 June 2021
Letters of invitation to participant [Stage 1 invitation letter]	1	12 February 2021
Letters of invitation to participant [Stage 2 invitation letter]	1	12 February 2021
Non-validated questionnaire [Clinical Anxiety Screening Tool for Severe Intellectual Disability (CIASP-ID)]	1	12 February 2021
Non-validated questionnaire [Background questionnaire]	1	12 February 2021
Non-validated questionnaire [Template Sleep Diary]	1	12 February 2021
Other [Evidence of Sponsor insurance or indemnity (document 2)]	-	01 August 2020
Other [Capacity to consent protocol]	1	12 February 2021
Other [Stage 1 accessible picture/symbol guide]	1	12 February 2021
Other [Stage 2 accessible picture/symbol guide]	1	12 February 2021
Other [Stage 1 feedback report template]	1	12 February 2021
Other [Stage 2 feedback report template]	1	12 February 2021
Other [Stage 2 pre-visit risk assessment]	1	12 February 2021
Other [GCP certificate for Dr Joanne Tarver]	-	10 June 2021
Other [GCP certificate for Lauren Shelley]	-	06 November 2019
Other [GCP refresher certificate for Lauren Shelley]	-	08 February 2021
Other [GCP certificate for Dr Effie Pearson]	-	10 February 2020
Other [CV for Dr Effie Pearson]	-	15 June 2021
Other [CV for Dr Hayley Crawford]	-	22 June 2021
Other [GCP certificate for Dr Jane Waite]	-	22 June 2021
Other [CV for Dr Caroline Richards]	-	22 June 2021
Other [CV for Dr Stacey Bissell]	-	22 June 2021
Other [Face, Legs, Activity, Cry, Consolability (FLACC) Scale]	1	13 July 2021
Other [Cover letter 2, summarising responses to queries prior to ethical review]	-	14 July 2021
Other [Flowchart of consent process]	1	16 August 2021
Other [Table of responses to provisional opinion]	-	09 September 2021
Other [Cover letter, summarising responses to favourable opinion with conditions]	-	14 October 2021
Participant consent form [Image and Video Release Consent form]	1	12 February 2021
Participant consent form [Stage 1 declaration form for over 16's without capacity to consent]	3	14 October 2021
Participant consent form [Stage 1 consent form for under 16's with capacity to consent]	3	14 October 2021

(CSHQ)]		
Validated questionnaire [Behaviour Rating Inventory for Executive Function – Preschool Version]		
Validated questionnaire [Non-communicating Children's Pain Checklist - Revised]		
Validated questionnaire [Anxiety, Depression and Mood Scale]		

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

IRAS Project ID: 296378	**Please quote this number on all correspondence**

Yours sincerely

[Signature removed]

Charlotte Ferris
Approvals Officer

E-mail: brightonandsussex.rec@hra.nhs.uk

Copy to: Miss Lauren Shelley

Appendix 23: Chapter Four and Five Background Questionnaire

BACKGROUND INFORMATION

Please tick or write your response to these questions concerning background details:

Please answer the following about the person you care for:

1. **Today's date:** _____
2. **Gender:** Male ☐ Female ☐
3. **Date of Birth**: ___/___/____ **Age:**_____
4. **Is the person you care for verbal?** (i.e. more than 30 signs/words in their vocabulary)

   Yes/No (delete as appropriate)
5. **Is the person you care for able to walk unaided?**

   Yes/No (delete as appropriate)
6. **Has the person you care for been diagnosed with SATB2-associated syndrome?** Yes/No (delete as appropriate)
7. **Has the person you care for received any additional diagnoses? (E.g., Autism)?** If yes, please give details:

   _____
   _____
8. **What is the genetic mechanism causing the syndrome in the person you care for?**

Uni-parental disomy	☐	Sequence repetition	☐
Deletion	☐	Translocation	☐
Unknown	☐	Other _____	
9. **When was the person you care for diagnosed?** _____
10. **Who diagnosed the person you care for?**

Paediatrician	☐	Clinical Geneticist	☐
GP	☐	Other _____	
11. **Has the person you care for had any medical/health difficulties in the last six months? If yes, please give details:**

    _____

    _____

We may need to contact your child's/person you care for's GP in order to clarify any information regarding your child's health status (see consent form and information sheet for more information). If you are happy for us to do this, please complete the relevant details below:

12. Name of your child's/person you care for's GP_____

GP Address_____

GP Telephone number_____

Is your child's GP the same as your GP?

Yes/No (delete as appropriate)

If no, please give us your GP details:

13. GP_____

GP Address_____

GP Telephone number_____

The following questions ask for background information <u>about you and your family</u>. Please tick the appropriate boxes or write in the spaces provided.

1. Are you male or female? Male ☐ Female ☐

2. What was your age in years on your last birthday? _____years

3. Please tick the highest level of your educational qualifications.

No formal educational qualifications..... ☐
Fewer than 5 GCSE's or O Level's (grades A-C), NVQ 1, or BTEC First Diploma……. …. ☐
5 or more GCSE's or O Level's (grades A-C), NVQ 2, or equivalent……..…….. ☐
3 or more 'A' Levels, NVQ 3, BTEC National, or equivalent..... ☐
Polytechnic/University degree, NVQ 4, or equivalent..... ☐
Masters/Doctoral degree, NVQ 5, or equivalent……..... ☐

4. What is your relationship to your child with a genetic syndrome (e.g. mother, father, stepmother, grandmother, adoptive parent)? _____

5. In total how many people currently live in your home? _____ Adults _____ Children

6. Does your child with a genetic syndrome normally live with you?

Yes ☐ No ☐

If no, then where do they live? _____

7. What is your current marital status?

Married, and living with spouse..... ☐

Living with partner..... ☐

Divorced/Separated/Widowed/Single and NOT living with a partner..... ☐

If living with partner/spouse, please answer the following questions, if not, please go to question 12.

8. Is your partner male or female? Male ☐ Female ☐

9. What was their age in years on their last birthday? _____ years

10. Please tick the highest level of your partner/spouse's educational qualifications.

No formal educational qualifications..... ☐

Fewer than 5 GCSE or O Level (grades A-C), NVQ 1, or BTEC First Diploma…… ☐

5 or more GCSE or O Level (grades A-C), NVQ 2, or equivalent……..… ☐

3 or more 'A' Levels, NVQ 3, BTEC National, or equivalent..... ☐

Polytechnic/University degree, NVQ 4, or equivalent..... ☐

Masters/Doctoral degree, NVQ 5, or equivalent……..... ☐

11. What is your partner/spouse's relationship to your child with a genetic syndrome (e.g., mother, father, stepmother, adoptive parent)? _____

12. Recent data from research with families of children with special needs has shown that a family's financial resources are important in understanding family member's views and experiences. With this in mind, we would be very grateful if you could answer the additional question below. We are not interested in exactly what your family income is, but we would like to be able to look at whether those with high versus lower levels of financial resources have different experiences.

What is your current total annual family income? Please include a rough estimate of total salaries and other income (including benefits) before tax and national insurance/pensions.

Please tick one box only:

Less than £15,000..... ☐

£15,001 to £25,000..... ☐

£25,001 to £35,000..... ☐

£35,001 to £45,000..... ☐

£45,001 to £55,000..... ☐

£55,001 to £65,000..... ☐

£65,001 or more..... ☐

Appendix 24: Challenging Behaviour Questionnaire – Expanded Version

THE CHALLENGING BEHAVIOUR QUESTIONNAIRE (CBQ)

Self-injurious behaviour

A) Has the person ever shown self-injurious behaviour (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing)?

Yes ☐ No ☐

1) Has the person shown self-injurious behaviour in the last month (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing)?

Yes ☐ No ☐

If the behaviour has not occurred, please go to question 2.

If the behaviour occurred in the past month, please answer questions 1a to 1d:

1a) Place a tick next to the item for any of the following list of behaviours which the person displays in a repetitive manner (repeats the same movement/ behaviour twice or more in succession):

Hits self with body part (e.g. slaps head or face) ……. ☐
Hits self against surface or object (e.g. bangs head on floor or table) ☐
Hits self with object…… ☐
Bites self (e.g. bites hand on wrist or arm) ……... ☐
Pulls (e.g. pulls hair or skin) ……... ☐
Rubs or scratches self (e.g. rub marks on arm or leg) ……. ☐
Inserts finger or objects (e.g. eye poking) …… ☐
Other form of self-injury, (please specify) _____

1b) In the last month, for how long did the **longest** episode or burst of this behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

1c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

1d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Aggression

B) Has the person ever shown aggression (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing)?

Yes ☐ No ☐

2) Has the person shown aggression in the last month (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing)?

Yes ☐ No ☐

If the behaviour has not occurred, please go to question 3.

If the behaviour occurred in the past month, please answer questions 2a to 2d:

2a) Place a tick next to the item for any of the following list of behaviours which the person displays:

Hits other with body part (e.g. slapping, punching, kicking, head-butting) …… ☐
Hits other with an object (e.g. throwing object or using a weapon) …… ☐
Bites other…….. ☐
Pulls or grabs other (e.g. hair-pulling, grabbing clothing) …… ☐
Rubs, pinches or scratches other ……... ☐
Spits at other …… ☐
Verbal aggression (e.g. threatening, swearing) …… ☐
Other form of physical aggression, (please specify) _____

2b) In the last month, for how long did the **longest** episode or burst of this behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

2c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

2d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Destruction of property

C) Has the person ever shown disruption and destruction of property or the environment (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal)?

Yes ☐ No ☐

3) Has the person shown disruption and destruction of property or the environment in the last month? (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal)?

Yes ☐ No ☐

If the behaviour has not occurred, please go to question 4.

If the behaviour occurred in the past month, please answer questions 3a to 3d:

3a) Place a tick next to the item for any of the following list of behaviours which the person displays:

Biting or chewing small objects (e.g. books, clothing) ……. ☐
Tearing or ripping small items (e.g. books, clothing) ……..…… ☐
Throwing, kicking or smashing small items (e.g. cups, mobile phones) …… ☐
Slamming, hitting, or kicking doors, walls or windows …….. ☐
Tipping, smashing or throwing large items (e.g. furniture, televisions) ……..… ☐
Pulling items from walls or shelves …….. ☐
Verbal aggression (e.g. threatening, swearing) …… ☐
Other form of disruption, (please specify) _____

3b) In the last month, for how long did the **longest** episode or burst of his behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

3c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

3d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Stereotypy

D) Has the person ever shown stereotyped behaviours? (e.g. rocking, twiddling objects, patting or tapping part of the body, constant hand movements, eye pressing)?

Yes ☐ No ☐

4) Has the person shown stereotyped behaviours in the last month? (e.g. rocking, twiddling objects, patting or tapping part of the body, constant hand movements, eye pressing)?

Yes ☐ No ☐

If the behaviour has not occurred, please go to question 5.

If the behaviour occurred in the past month, please answer questions 4a to 4d:

4a) Place a tick next to the item for any of the following list of behaviours which the person displays:

Full body movements (e.g. rocking, bouncing) ……. ☐
Movement of an object (e.g. twiddling or spinning object) ……..…… ☐
Movement of isolated body part (e.g. hand flapping, head shaking) ……..…… ☐
Eye-pressing or visual regard of movement (e.g. strobing, spinning object) …… ☐
Mouthing or sucking on body part or object ……..… ☐

Other form of stereotypy, (please specify) _____

4b) In the last month, for how long did the **longest** episode or burst of his behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

4c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

4d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Other challenging behaviour

E) Has the person ever shown any other form of challenging behaviour?

Yes ☐ No ☐

5) Has the person shown any other form of challenging behaviour in the last month?

Yes ☐ No ☐

If the behaviour occurred in the past month, please answer questions 5a to 5d:

5a) Place a tick next to the item for any of the following list of behaviours which the person displays:

Pica (e.g. eating paper, leaves, discarded food) …….…. ☐
Inappropriate vocalisations (e.g. screaming, shouting) ……..…… ☐
Removal of clothing (not for purpose of washing changing or toileting) …… ☐
Sexual behaviour (e.g. public masturbation, inappropriate sexual contact) ……. ☐
Anal poking…….…… ☐
Smearing of bodily substance (non-accidental) …… ☐
Stealing ……..……. ☐
Self-induced vomiting …… ☐

Other form of disruption, (please specify) _____

5b) In the last month, for how long did the **longest** episode or burst of his behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

5c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

5d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Appendix 25: Questions About Behavioural Function Scale

Questions About Behavioural Function Scale (QABF) removed due to copyright restrictions

Appendix 26: Vineland Adaptive Behavior Scales – Third Edition

Vineland Adaptive Behavior Scales – Third Edition (VABS-3) removed due to copyright restrictions

Appendix 27: Chapter Four interview schedule

Interview Schedule

Overview: This semi-structured interview has been developed to capture and characterise behaviours that challenge in children and adults with SATB2-associated syndrome (SAS). It aims to use open-ended questions to gather information regarding the triggers and functions of behaviours, potential maintaining factors, and the impact of behaviour on the individual and those around them. The interview has two sections: general introductory items (Section A) and items relating to the triggers, functions, and impact of behaviour (Section B). Section B is to be completed on one behaviour at a time and may be completed multiple times to gather information about more than one behaviour.

For the current study, we are particularly interested in aggression, as this category of behaviour has been identified as highly prevalent in SAS in previous research. We are also interested in a behaviour that parents/carers identify as being the most problematic or concerning, as this likely to be clinically informative. For each family, these two behaviours might be the same or different.

Format of the interview: If a parent/carer identifies aggression as being the behaviour of most concern, they will complete the interview questions once about this behaviour. If a parent/carer identifies that the behaviour of most concern is different to aggression, they will complete the interview questions once about aggression and once about the concerning behaviour that they identify. If a parent/carer reports that aggression does not occur in their child or person they care for, the parent/carer will complete the interview questions once about the behaviour that they identify to be of most concern.

The interview is fluid; therefore parents/carers are encouraged to talk freely, and the order and direction of the interview may vary between participants. During the interview, questions which are not deemed relevant may be omitted by the researcher and extra prompts may be added if the researcher considers that further information is necessary. All questions asked should be kept within the guidelines given although the interview will be guided by the participants and the order of questions may be changed to best suit the direction of the interview. Participants will not be subjected to leading questions.

Scoring: There is a separate coding sheet for this interview. Scoring for each question can be found in the right-hand column of the scoring sheet. These should be filled out based on the information given by the parent/carer, placing an 'X' in the relevant boxes, and giving descriptive answers where spaces have been provided. Further notes can be made in the 'notes' column. Prompts may be given to aid the researcher in scoring each item although the scoring categories will not be disclosed to the participant. Further clarification of the scoring categories and other useful information has been provided in the right-hand column of the interview. 'Other' categories have been included where relevant to ensure important information is not overlooked or missed.

Confidentiality statement: Before we start, I must let you know that everything you say is confidential to me and the members of the research team, however, if you do say something that makes me concerned about your safety, the safety of your child, or anyone else's safety, I will need to share my concerns beyond the team. If that is the case, I would speak to you before I did that. Is that all ok?

Section A: Introductory Items

Throughout this interview we are particularly interested in any behaviour that X shows which might be described as a behaviour that challenges. By this, we mean behaviours that may lead to injury to X or to those around them, or behaviours that may disrupt or restrict activities in X's day to day life. We are particularly interested in talking about aggressive type behaviours (such as hitting others or throwing objects, hair pulling, or grabbing others, pinching/scratching) as we have previously identified that these types of behaviours may be prevalent in SATB2-associated syndrome.

Question	Prompts	Information
1. Are there any behaviours X shows that you might describe as being aggressive? Can you describe what this behaviour looks like for me?	*Examples:* *-Hits others with body part (e.g., slapping, punching, kicking, head-butting)* *-Hits other with an object (e.g., throwing object or using it as a weapon)* *-Bites other* *-Pulls or grab other (e.g., hair-pulling, grabbing clothing)* *-Spits at other* *-Vocalised aggression (e.g., threatening, screaming, shouting)*	*Describe behaviour*

Within this interview we would also like to ask about behaviour X might show that you find is of most concern to you. This could be the same or different to the behaviour that you just described. It could be a behaviour that occurs less frequently but that is more intense and concerning when it does occur, or it could be something that might be less intense but is of concern because it occurs more frequently.

Question	Prompts	Information
2. Can you tell me about a behaviour X shows that is of most concern to you?	• Can you tell me a bit more about that? • What does that behaviour look like for X? *Example behaviours:*	*Describe behaviour*

	Self-injury, destruction of property, stereotyped behaviour, temper outbursts	
3. How does X communicate his/her needs or wants to you?	*E.g., seek people's attention? Show that they want something or to do something? Show that they do <u>not</u> want something or to do something?*	
4. Does (X) currently have any medical conditions?	• Are they currently receiving any treatment for this/these?	
5. Is (X) involved in any behavioural programmes or interventions that aim to improve his/her behaviour?	*(If yes)* • When did this start? • How do you think that this/these have impacted (X's) behaviour? • Have you seen any change to the behaviour? E.g., improved, got worse, stayed about the same?	
6. Does (X) take any medication to help improve his/her behaviour?		

SECTION B

We would now like to ask you about the behaviours you identified earlier in a bit more detail. You mentioned earlier that X displays [INSERT BEHAVIOUR] …

Useful prompt to be used when appropriate: 'can you tell me a bit more about that?'

Opening Question	**Sub Questions**	**Information/scoring**
1. Can you describe a recent example of when (X) showed this behaviour?	• What happened? • How frequently or often does this behaviour occur?	**Scoring**: Never (0) At least once a month (1) At least once a week (2) At least once a day (3) At least once an hour (4)
2. Is that a typical example for when this behaviour might occur?		*If yes, answer Q4. If no, answer Q3.*

Opening Question	**Sub Questions**	**Information/scoring**
3. Can you describe a typical example of when (X) shows this behaviour?	How frequently or often does this behaviour occur?	**Scoring**: Never (0) At least once a month (1) At least once a week (2) At least once a day (3) At least once an hour (4)
4. How long would you say the entire episode of the behaviour typically lasts? (minutes)	On average…?	**Scoring:** Less than a minute (1) Less than 5 minutes (2) Less than 15 minutes (3) Less than an hour (4) More than an hour (5)
5. Are there any other behaviours or is there a cluster of behaviours that occur alongside the behaviour?	• *If yes,* do these behaviours occur together in a predictable chain? • Do these behaviours always occur with the behaviour?	*E.g., repetitive requests, emotional vocalisations, self-injury, verbal/physical aggression towards others, non-compliance with requests, increased motor activity or vocalisations?*
6. Can you tell me about what things might trigger or cause the onset of this behaviour? *Please list all possible triggers.*	a. If they want to escape work/learning situations or when asked to do something that they do not want to do (e.g. get dressed, brush teeth, work, etc? *[demand escape]* b. If they want to escape from a social situation or do not want social attention or others to be around them (e.g., if somebody is interacting socially with him/her or if he/she wants to be left alone) *[social escape]* c. If they want your attention, to get a reaction or to draw attention to themselves? *[attention]* d. If they are bored or unoccupied, there is nothing else to do, think there is no one in the room/nearby, or they appear to enjoy the behaviour? *[self-stimulation]* e. If they are in pain, poorly, feeling unwell, physically uncomfortable, something is bothering them physically, or to indicate to you that they	*Internal and External.* I.e., setting events and common triggers.

Opening Question	Sub Questions	Information/scoring
	aren't feeling well? *[physical/pain-related]* f. If they want access to a preferred item (e.g. toy, food or drink), access to something you or another person has, or if something that they want is taken away from them? *[tangibles]* g. If there is change or disruption to their usual routine, something unexpected happens, they aren't able to complete a routine or ritual, or to try and re-establish a preferred routine or ritual? *[disruption to routine]* h. If they are worried/anxious about something or an event, or frustrated or disappointed? *[emotion-related]* i. If there is sensory input in the environment that they do not like, such as sounds or light, or they want to be removed from a noisy, crowded, or bright environment? *[sensory sensitivity]* j. If they did not sleep well the night before, had difficulty settling, or more night-waking's? [*biological- sleep*] k. If they are tired or hungry? [*biological- hunger/tiredness*] l. If it is a weekend or they are in a 'social' place? [*environmental*] • Are there any other patterns or high-risk situations that you think might lead to the behaviour occurring that I haven't asked about?	
7. What is it about this/these things that you think X finds difficult?		

Opening Question	Sub Questions	Information/scoring
8. In the minutes leading up to and after the behaviour, do you notice any changes or differences in (X) in terms of their body movements or facial expressions? (Prompt for changes before and after the behaviour). *List all possible changes.*	*For example.,* a. Changes in their vocalisations? *E.g., increase in crying, screaming, whining.* b. Changes in how they interact with you (social behaviours)? *E.g., withdrawing, seeking comfort.* c. Changes in their facial expressions or unusual facial expressions? *E.g., changes in eyes, mouth, brow* d. Changes in their activity levels? *E.g., moving more or less* e. Changes in how much they're moving around e.g. with their body, arms or legs? *E.g., increased/decreased tension, protecting body parts.* f. Changes physiologically? *E.g., changes in breathing, colour, sweating.* g. Do you ever notice how they've slept prior to a bad day of behaviour, or how they've slept after a bad day of behaviour? *E.g., increased/decrease in sleep, longer to get to sleep, more night waking's*. h. Changes to their eating habits? *E.g., increased/decreased interest*.	
9. *If medical/health conditions:* Have you noticed a relationship between the behaviour and medical, health or dental conditions?		
10. Is there a time of day or night when the behaviour more likely to happen?		

Opening Question	**Sub Questions**	**Information/scoring**
11. At what age did (X) begin to show this behaviour?	• Did the behaviour happen gradually overtime or suddenly?	
12. Were there any specific triggers or life events around the time that (X) first began to show the behaviour?		
13. Has the behaviour stayed at the same level overtime? *(same/improved/worsened)*	• Do you have any idea of why this might be?	
14. Is there anything that you do to prevent the behaviour from occurring?	• What if there is an upcoming situation or event where you know that the behaviour is likely to occur? • Are there any situations that you avoid because they lead to the behaviour occurring?	**List three most often used preventative strategies.**
15. Is there anything (X) does that helps him/her to stop the behaviour?	• Does he/she ever 'self-restrain?' • How often/frequently do they use these strategies? • Do they work? How well?	**List three most often used strategies.** *E.g., hugs self, sits on hands, use of clothing, covers face/head/ears, removes self from the environment.* **Frequency:** -Never -Sometimes: *several times a month* -Often: *several times a week* -Always: *several times a day*
16. When the behaviour does occur, is there anything that you can do that helps to stop or alleviate the behaviour?	• How often do you try using these techniques? • Do these strategies work? How often? • How does (X) respond to these strategies?	**List three most often used strategies.** *E.g., verbal discouragement, or reminders or distraction (social); ignore (non-social); rewards for good behaviours or offer alternatives in response to the behaviour (behavioural); removal from situation or environment, or removal of others from immediate environment (physical); give them the toy/activity/food/item they are requesting or want (tangible)* **Frequency**:

Opening Question	**Sub Questions**	**Information/scoring**
		-Never -Sometimes: *several times a month* -Often: *several times a week* -Always: *several times a day*
17. What happens if the strategies that help are not available?	• How long does this response typically go on for before the behaviour stopped? (*duration in minutes*)	*Example: if you do not do (insert strategy mentioned above).*
18. What usually happens after the behaviour has ended?		
19. How does the behaviour affect (X) on a day-to-day basis?	• What's the *biggest impact* it has? (e.g., Peer/family relationships, homelife, learning experiences, engagement in leisure activities) • Does the behaviour ever result in X physically injuring him/herself?	**N/A** (0) **Mild**: slight impact on relationships, home life, outside home life (1). **Moderate**: Clear interference, withdrawal from normal routine, conflicts with others (2) **Severe**: marked interferences, significantly affects relationships with others, totally or almost totally unable to maintain appropriate family relationships/function at home/outside of the home (3)
20. How does (X's) behaviour affect you and your family?	• What is the *biggest impact* it has on you and your family? • How severely would you say this affects you? • Has the impact changed over time (i.e., improved/worsened)? • Do you do anything to prevent/reduce the impact? • Does the behaviour ever result in you or others being physically injured?	*E.g., negative/positive influence on peer/family/partner relationship/s; decrease/increase in time spent with others/participating in activities; in the home/outside of the home; negative/positive influence on time spent with other siblings; financial implications as a result of the behaviour?* **N/A** (0)

Opening Question	Sub Questions	Information/scoring
		Mild: slight impact on relationships, home life, outside home life (1) **Moderate**: Clear interference, withdrawal from normal routine, conflicts with others (2) **Severe**: marked interferences, significantly affects relationships with others, totally or almost totally unable to maintain appropriate family relationships/function at home/outside of the home (3)
21. How do you feel about the behaviour?	• What emotions do you feel? Any positive or negative feelings towards parenting?	
22. Do you think that the emotion you feel at the time of onset of the behaviour affects how you respond?	• Are there any times when you are more or less likely to respond in a certain way to the behaviour?	*E.g., when happy, relaxed, stressed, worried, tired.*
23. How well do you feel you can manage X's behaviour?		0 = unable to manage 10 = can manage very well
24. Is there anything else you think is important to tell us?		

Appendix 28: Chapter Four invitation letter

Insert support group logo /CNDD logo as appropriate

Aston University

Dear [insert name of parent/caregiver],

A new research project is being carried out at Aston University. We wanted to let you know about this study so that you can decide whether you would like to take part. The research is an interview study that is looking at behaviours that challenge in children and adults with *SATB2*-associated syndrome. The aim of the research is to better understand factors that may influence behaviour in *SATB2*-associated syndrome.

We would like to speak with parents or caregivers of individuals with *SATB2*-associated syndrome about behaviour shown by their child or the person they care for in an in-depth interview. We would also like to ask parents or caregivers to complete a couple of questionnaires about their child or person they care for. During the interview, we will ask you about behaviour shown by your child or person you care for, such as asking about events or situations that lead to difficult or concerning behaviour, and what happens before and after these behaviours are shown. We would like to ask you about behaviour that is of most concern to you, as well as certain types of behaviour that your child or person you care for may or may not show, such as hair pulling, grabbing other's clothing, and biting. From previous work we have conducted, we know that some individuals with SATB2-associated syndrome show some of these forms of behaviour. However, our understanding of these behaviours in *SATB2*-associated syndrome is currently limited. The interview will take place over the phone or using video conferencing facilities (e.g., Microsoft Teams) at a time that is convenient for you.

If you are interested in finding out more about the study, you can do so in the following ways:

1) Complete an online expression of interest form that can be accessed at the web address below: [insert link to online expression of interest form].
   Your reference number: [INSERT ID; to be included for CNDD participant database invites]
2) Contact Lauren Shelley (Doctoral researcher) on shellel1@aston.ac.uk, or 0121 204 3203.

A member of the research team will then contact you to provide more information about the study.

If you are unclear about any aspect of the study or have any questions please contact Lauren Shelley, 0121 204 3203; shellel1@aston.ac.uk, or Dr Jane Waite, 0121 204 4307; j.waite@aston.ac.uk.

Thank you for your time and we look forward to hearing from you.

Yours sincerely,

Dr Jane Waite
Lecturer and Clinical Psychologist
Aston University

Lauren Shelley
Doctoral Researcher
Aston University

If you are on the mailing list of a SATB2-associated syndrome support group, you may have already received an invitation to participate in this research. If this is the case, we apologise for contacting you more than once. (to be included for recruitment from the participant database held at the CNDD)

Appendix 29: Chapter Four participant informant sheets

Aston University
BIRMINGHAM UK

Behaviours that challenge in SATB2-associated syndrome

Participant Information Sheet

Interview study: Parents/carers of children with SAS

Version 3 14.10.2021

Invitation

We would like to invite you to take part in a research study.

If you have questions or would like a verbal explanation of this study, contact Jane Waite (Lead Researcher) on 0121 204 4307 or j.waite@aston.ac.uk, or Lauren Shelley (Doctoral Researcher) on 0121 204 3203 or shellel1@aston.ac.uk.

Before you decide if you would like to participate, take time to read the following information carefully and, if you wish, discuss it with others such as your family, friends, or colleagues.

Please ask a member of the research team, whose contact details can be found at the end of this information sheet, if there is anything that is not clear or if you would like more information before you make your decision.

What is the purpose of the study?

We would like to invite parents or caregivers of a child with SATB2-associated syndrome, aged 15 years or under, to take part in an interview study about behaviours that are shown by their child. The aim is to understand more about behaviours that challenge in SATB2-associated syndrome, including the factors that are associated with these behaviours.

We would like to ask about certain types of behaviours that challenge that your child or person you care for may show, for example, aggressive behaviours such as pulling hair, grabbing other's clothing, hitting, and biting. From previous work we have conducted, we know that some individuals with SATB2-associated syndrome show some of these forms of behaviour, however our understanding of these behaviours in SATB2-associated syndrome is currently limited. We would also like to ask about behaviour shown by your child that is of most concern to you.

We hope that the study will help healthcare professionals working with individuals with SATB2-associated syndrome to understand more about the factors that influence behaviours that challenge.

The study is part of a larger study of behaviours that challenge in people with SATB2-associated syndrome that is being conducted as part of Lauren Shelley's PhD studies that are ongoing until 2023. The PhD research

is endorsed by the SATB2 Gene Foundation. You will be given the option for us to retain your details if you would like to be invited to take part in other aspects of the study. This is optional, if you take part in this interview study, then you do not have to take part in the next stages of the research.

Why have I been invited?

You are being invited to take part in this study because records held by the [SATB2 Gene Foundation/SATB2 Gene Trust UK/Cerebra Network for Neurodevelopmental Disorders (delete as appropriate)] indicate that you are a parent or carer of a person with SATB2-associated syndrome, aged 15 years or under, and that [you are happy to be contacted for research purposes/have consented to be contacted about future research (delete as appropriate)]. Alternatively, we may have contacted you because you responded to an advert about the study.

What will happen to me if I take part?

If you decide to take part in this study, you will be invited to complete an interview over the telephone or using online conferencing facilities (e.g., Microsoft Teams). The interview will take approximately 60 minutes (although it may take longer depending on how much you wish to tell us). This can be split into two shorter sessions if you would prefer. The interview will include questions that ask about situations that lead to behaviours occurring, functions of the behaviours, and the impact of the behaviours on your child and the people around them. We will ask you if you are happy for the interview to be audio recorded, so that we can analyse the information given by parents/carers after the interviews have finished.

If you would like to take part in this interview study, you will need to complete a consent form. If your child or person you care for can understand what is involved in the study, you will also need to support them to complete an assent/consent form. If your child or person you care for has the capacity to understand some or all the information about this study, you should explain the study to them in a way they understand. You can also request a copy of an accessible guide from the researcher to support your explanation (shellel1@aston.ac.uk).

If you are completing the consent forms online, the survey will direct you to the correct consent forms based on your responses. Once we have received your consent, we will ask you to complete some questionnaires to provide background information about your child or person you care for.

If you are completing a paper copy of the consent forms and you have a *child who can fully understand* what is involved in the study, you and your child should complete the yellow assent/consent form. If your child is *not able to understand* what is involved in the study, you should complete the blue consent form. Once we have received consent, we will ask you to complete some questionnaires to provide background information about your child and send these to us in the prepaid envelope provided.

Both online and paper questionnaires will take approximately 10 minutes to complete. Once we have received your consent and you have completed the questionnaires, we will contact you to arrange a convenient time and date for the interview.

We will also ask you if it is ok to ask you some additional questions about your child's adaptive ability (e.g., daily living skills) over the phone or using online conferencing facilities (e.g., Microsoft Teams). These questions can be completed within 6 weeks of completing the interview. If you would not like to do this, you can leave this question blank/select no on the consent form.

We will ask you to provide a copy of a genetic/clinical confirmation letter confirming your child's diagnosis of SATB2-associated syndrome, if possible, from a GP, paediatrician, geneticist or other professional. This will help us to confirm diagnosis of SATB2-associated syndrome. A copy of this letter can be uploaded onto an online form, emailed, or posted to the research team. This is entirely optional and there is no obligation to provide this document to participate.

We will be collecting information from participants between November 2021 and April 2022. After that we will spend some time understanding the data and writing reports.

Joining up research studies to better understand SATB2-associated syndrome:

If you have participated in a research study investigating the 'Behavioural phenotype of SATB2-associated syndrome', led by the Cerebra Network for Neurodevelopmental Disorders (CNDD), University of Birmingham, we will also ask whether you are happy for us to use your unique reference number to extract and link your responses from the CNDD study to the information you provide in this study. *This is optional*. The information from the ongoing CNDD study will help us to understand the results of the current study.

Do I have to take part?

No. It is up to you to decide whether or not you and/or your child/person you care for wish to take part.

If you do decide to participate, you will be asked to sign and date a consent form. After the date of the interview, you have 14 days to withdraw from the study, without giving a reason. After this period, we may have coded data from the interviews, and you will be unable to withdraw your data. If you decide to withdraw within the 14 days following the assessment day, we will not delete your data unless you tell us to.

Will my taking part in this study be kept confidential?

Yes. A code will be attached to all the data you provide to maintain confidentiality.

Your personal data (name and contact details) will only be used if the researchers need to contact you to arrange study visits or collect data by phone. Analysis of your data will be undertaken using coded data.

The data we collect will be stored in a secure document store (paper records) or electronically on a secure encrypted mobile device, password protected computer server or secure cloud storage device.

To ensure the quality of the research, Aston University may need to access your data to check that the data has been recorded accurately. If this is required, your personal data will be treated as confidential by the individuals accessing your data.

Appendix Twenty-Nine

How will the audio recordings and the information I provide be managed?

With your permission we will audio record the interview and take notes. The recording will be typed into a document (transcribed) by a transcriber approved by Aston University. During the transcription process any names that have been used will be replaced with a pseudonym.

Audio recordings will be destroyed as soon as the transcripts have been checked for accuracy.

We will ensure that anything you have told us that is included in the reporting of the study will be anonymous. You of course are free not to answer any questions that are asked without giving a reason.

Will my GP be informed of my involvement in the study?

If you live in the UK, with your consent, we will notify the GP of your child/person you care for that you are taking part in this research project about your child/person you care for.

If we become aware that your child may be experiencing an undiagnosed health difficulty, Dr Jane Waite will write to the GP of your child/person you care for to pass on this information. If you live outside of the UK, we will advise you to notify the people involved in your child or person you care for's usual care of your involvement in the study.

If it becomes apparent that you are showing signs of an undiagnosed health difficulty, we will advise you to contact your GP, specialist, or the relevant national contact of a SATB2-associated syndrome support group.

What happens if I tell you something that concerns you about my health or welfare or that of the person I care for?

In the unlikely event of this happening, we will discuss with you how this should be addressed. If necessary, to protect you and the person you care for, we will report your concern to the appropriate person or bodies. Any request for advice concerning your child/person you care for will be passed on to Dr Jane Waite (Clinical Psychologist), who will provide information about accessing local support.

If you experience emotional distress while speaking with a researcher, we will provide non-directive emotional support during that conversation and ask you whether you would like to take a break, or end the conversation. As you are taking part in research, we cannot offer health advice or further support with emotional distress, however, we will signpost you to relevant support services (e.g., GP, syndrome support group). We will only breach confidentiality if we suspect you or your child are at risk of harm (e.g., emergency/safeguarding concern). In an emergency, we would call the emergency services. Safeguarding concerns would be discussed with Aston University's safeguarding team who would contact relevant services (e.g., social care). We will notify you if we intend to breach confidentiality, unless doing so could increase risk to you or your child.

What are the possible benefits of taking part?

You will receive an individualised feedback report that will detail your responses on standardised measures completed as part of the study. This study will help us to better understand the factors that influence behaviour in SATB2-associated syndrome. It is hoped that the research will help us to find more about the needs of people with SATB2-associated syndrome who show behaviours that challenge.

What are the possible risks and burdens of taking part?

We will be asking you to think about times when your child or person you care for shows behaviours that challenge. Some parents or caregivers may find this distressing. You will be able to stop the interview at any time if you feel it is too upsetting.

Your decision to participate in this study will not impact your right or the right of your child/person you care for to access services.

The online consent forms and questionnaires are created through 'Qualtrics' and hosted on highly secure servers that comply with General Data Protection Regulations. However, as with all online activity, there is a risk that unauthorised individuals (hackers) may access data. If you are uncomfortable with this risk, or simply would prefer a paper copy of the consent forms, please contact the research team who can put one in the post to you.

What will happen to the results of the study?

The results of this study will be published in scientific journals and/or presented at conferences. If the results of the study are published, your identity will remain confidential. Anonymised quotes may be used in publications resulting from the study.

Research findings will also be published in newsletters of support groups and educational institutions.

A lay summary of the results of the study will be available for participants when the study has been completed and the researchers will ask if you would like to receive a copy. All participants will also receive an individualized feedback report that will detail their responses on standardised measures completed as part of the study.

The anonymized results may be shared with the company providing funding for this study. The results of the study will also be used in Lauren Shelley's PhD thesis.

Expenses and payments

Participants will not incur any expenses as a result of taking part in this study. However, if you complete the interview study, you will be entered into a prize draw to win one of two £50 online vouchers. We will notify participants by 31/07/2022 if they have been successful.

Who is funding the research?

The study is being funded by The Baily Thomas Charitable Fund.

Who is organising this study and acting as data controller for the study?

Aston University is organising this study and acting as data controller for the study. You can find out more about how we use your information in Appendix A.

Appendix Twenty-Nine

Who has reviewed the study?

This study was given a favorable ethical opinion by the London – Brighton and Sussex Research Ethics Committee.

What if I have a concern about my participation in the study?

If you have any concerns about your participation in this study, please speak to the research team and they will do their best to answer your questions. Contact details can be found at the end of this information sheet.

If the research team are unable to address your concerns or you wish to make a complaint about how the study is being conducted, you should contact the Aston University Research Integrity Office at research_governance@aston.ac.uk or telephone 0121 204 3000.

Research Team

This research is being led by:

Jane Waite (Lead Researcher)
Telephone: 0121 204 4307. Email: j.waite@aston.ac.uk

Lauren Shelley (Doctoral Researcher)
Telephone: 0121 204 3203. Email: shellel1@aston.ac.uk

Thank you for taking time to read this information sheet. If you have any questions regarding the study, please do not hesitate to ask a member of the research team.

Appendix A: Transparency Statement

Aston University takes its obligations under data and privacy law seriously and complies with the Data Protection Act 2018 ("DPA") and the General Data Protection Regulation (EU) 2016/679 as retained in UK law by the Data Protection, Privacy and Electronic Communications (Amendments etc) (EU Exit) Regulations 2019 ("the UK GDPR").

Aston University is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study. Aston University will process your personal data in order to register you as a participant and to manage your participation in the study. It will process your personal data on the grounds that it is necessary for the performance of a task carried out in the public interest (GDPR Article 6(1)(e). Aston University may process special categories of data about you which includes details about your health. Aston University will process this data on the grounds that it is necessary for statistical or research purposes (GDPR Article 9(2)(j)). Aston University will keep identifiable information about you for 6 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally identifiable information possible.

You can find out more about how we use your information at https://www.aston.ac.uk/about/statutes-ordinances-regulations/publication-scheme/policies-regulations/data-protection or by contacting our Data Protection Officer at dp_officer@aston.ac.uk.

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer who will investigate the matter. If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO).

Behaviours that challenge in SATB2-associated syndrome

Participant Information Sheet

Interview study: Parents/carers of individuals with SAS aged 16 years and over

Version 3 14.10.2021

Invitation

We would like to invite you to take part in a research study.

If you have questions or would like a verbal explanation of this study, contact Jane Waite (Lead Researcher) on 0121 204 4307 or j.waite@aston.ac.uk, or Lauren Shelley (Doctoral Researcher) on 0121 204 3203 or shellel1@aston.ac.uk.

Before you decide if you would like to participate, take time to read the following information carefully and, if you wish, discuss it with others such as your family, friends, or colleagues.

Please ask a member of the research team, whose contact details can be found at the end of this information sheet, if there is anything that is not clear or if you would like more information before you make your decision.

What is the purpose of the study?

We would like to invite parents or caregivers of an individual with SATB2-associated syndrome, aged 16 years or over, to take part in an interview study about behaviours that are shown by their child or person they care for. The aim is to understand more about behaviours that challenge in SATB2-associated syndrome, including the factors that are associated with these behaviours.

We would like to ask about certain types of behaviours that challenge that your child or person you care for may or may not show, for example, aggressive behaviours such as pulling hair, grabbing other's clothing, hitting, and biting. From previous work we have conducted, we know that some individuals with SATB2-associated syndrome show some of these forms of behaviour, however our understanding of these behaviours in SATB2-associated syndrome is currently limited. We would also like to ask about behaviour shown by your child or person you care for that is of most concern to you.

We hope that the study will help healthcare professionals working with individuals with SATB2-associated syndrome to understand more about the factors that influence behaviours that challenge.

The study is part of a larger study of behaviours that challenge in people with SATB2-associated syndrome that is being conducted as part of Lauren Shelley's PhD studies that are ongoing until 2023. The PhD research is endorsed by the SATB2 Gene Foundation. You will be given the option for us to retain your details if you would like to be invited to take part in other aspects of the study. This is optional, if you take part in this interview study, then you do not have to take part in the next stages of the research.

Why have I been invited?

You are being invited to take part in this study because records held by the [SATB2 Gene Foundation/SATB2 Gene Trust UK/Cerebra Network for Neurodevelopmental Disorders (delete as appropriate)] indicate that you are a parent or carer of a person with SATB2-associated syndrome, aged 16 years or over, and that [you are happy to be contacted for research purposes/have consented to be contacted about future research (delete as appropriate)]. Alternatively, we may have contacted you because you responded to an advert about the study.

What will happen to me if I take part?

If you decide to take part in this study, you will be invited to complete an interview over the telephone or using online conferencing facilities (e.g., Microsoft Teams). The interview will take approximately 60 minutes (although it may take longer depending on how much you wish to tell us). This can be split into two shorter sessions if you would prefer. The interview will include questions that ask about situations that lead to behaviours occurring, functions of the behaviours, and the impact of the behaviours on your child/person you care for and the people around them. We will ask you if you are happy for the interview to be audio recorded, so that we can analyse the information given by parents/carers after the interviews have finished.

If you would like to take part in this interview study, you will need to complete a consent form. If your child or person you care for can understand what is involved in the study, you will also need to support them to complete an assent/consent form. If your child or person you care for has the capacity to understand some or all the information about this study, you should explain the study to them in a way they understand. You can also request a copy of an accessible guide from the researcher to support your explanation (shellel1@aston.ac.uk).

If you are completing the consent forms online, the survey will direct you to the correct consent forms based on your responses. Once we have received your consent, we will ask you to complete some questionnaires to provide background information about your child or person you care for.

If you are completing a paper copy of the consent forms and *the person with SAS can fully understand* what is involved in the study, you and the person you care for should complete the yellow consent forms. If the *person with SAS is not able to understand* what is involved in the study, you should complete the blue consent and declaration forms. Once we have received consent, we will ask you to complete some questionnaires to provide background information about your child or person you care for and send these to the research team in the prepaid envelope provided.

Both online and paper questionnaires will take approximately 10 minutes to complete. Once we have received your consent and you have completed the questionnaires, we will contact you to arrange a convenient time and date for the interview.

We will also ask you if it is ok to ask you some additional questions about your child or person you care for's adaptive ability (e.g., daily living skills) over the phone or using online conferencing facilities (e.g., Microsoft Teams). These questions can be completed within 6 weeks of completing the interview. If you would not like to do this, you can leave this question blank/select no on the consent form.

We will ask you to provide a copy of a genetic/clinical confirmation letter confirming the person you care for's diagnosis of SATB2-associated syndrome, if possible, from a GP, paediatrician, geneticist or other professional. This will help us to confirm diagnosis of SATB2-associated syndrome. A copy of this letter can

be uploaded onto an online form, emailed, or posted to the research team. This is entirely optional and there is no obligation to provide this document to participate.

We will be collecting information from participants between November 2021 and April 2022. After that we will spend some time understanding the data and writing reports.

Joining up research studies to better understand SATB2-associated syndrome:

If you have participated in a research study investigating the 'Behavioural phenotype of SATB2-associated syndrome', led by the Cerebra Network for Neurodevelopmental Disorders (CNDD), University of Birmingham, we will also ask whether you are happy for us to use your unique reference number to extract and link your responses from the CNDD study to the information you provide in this study. *This is optional*. The information from the ongoing CNDD study will help us to understand the results of the current study.

Do I have to take part?

No. It is up to you to decide whether or not you and/or your child/person you care for wish to take part.

If you do decide to participate, you will be asked to sign and date a consent form. After the date of the interview, you have 14 days to withdraw from the study, without giving a reason. After this period, we may have coded data from the interviews, and you will be unable to withdraw your data. If you decide to withdraw within the 14 days following the assessment day, we will not delete your data unless you tell us to.

Will my taking part in this study be kept confidential?

Yes. A code will be attached to all the data you provide to maintain confidentiality.

Your personal data (name and contact details) will only be used if the researchers need to contact you to arrange study visits or collect data by phone. Analysis of your data will be undertaken using coded data.

The data we collect will be stored in a secure document store (paper records) or electronically on a secure encrypted mobile device, password protected computer server or secure cloud storage device.

To ensure the quality of the research, Aston University may need to access your data to check that the data has been recorded accurately. If this is required, your personal data will be treated as confidential by the individuals accessing your data.

How will the audio recordings and the information I provide be managed?

With your permission we will audio record the interview and take notes. The recording will be typed into a document (transcribed) by a transcriber approved by Aston University. During the transcription process any names that have been used will be replaced with a pseudonym.

Audio recordings will be destroyed as soon as the transcripts have been checked for accuracy.

We will ensure that anything you have told us that is included in the reporting of the study will be anonymous. You of course are free not to answer any questions that are asked without giving a reason.

Will my GP be informed of my involvement in the study?

If you live in the UK, with your consent, we will notify the GP of your child/person you care for that you are taking part in this research project about your child/person you care for. If we become aware that your child may be experiencing an undiagnosed health difficulty, Dr Jane Waite will write to the GP of your child/person you care for to pass on this information. If you live outside of the UK, we will advise you to notify the people involved in your child or person you care for's usual care of your involvement in the study.

If it becomes apparent that you are showing signs of an undiagnosed health difficulty, we will advise you to contact your GP, specialist, or the relevant national contact of a SATB2-associated syndrome support group.

What happens if I tell you something that concerns you about my health or welfare or that of the person I care for?

In the unlikely event of this happening, we will discuss with you how this should be addressed. If necessary, to protect you and the person you care for, we will report your concern to the appropriate person or bodies. Any request for advice concerning your child/person you care for will be passed on to Dr Jane Waite (Clinical Psychologist), who will provide information about accessing local support.

If you experience emotional distress while speaking with a researcher, we will provide non-directive emotional support during that conversation and ask you whether you would like to take a break, or end the conversation. As you are taking part in research, we cannot offer health advice or further support with emotional distress, however, we will signpost you to relevant support services (e.g., GP, syndrome support group). We will only breach confidentiality if we suspect you or your child are at risk of harm (e.g., emergency/safeguarding concern). In an emergency, we would call the emergency services. Safeguarding concerns would be discussed with Aston University's safeguarding team who would contact relevant services (e.g., social care). We will notify you if we intend to breach confidentiality, unless doing so could increase risk to you or your child.

What are the possible benefits of taking part?

You will receive an individualised feedback report that will detail your responses on standardised measures completed as part of the study. This study will help us to better understand the factors that influence behaviour in SATB2-associated syndrome. It is hoped that the research will help us to find more about the needs of people with SATB2-associated syndrome who show behaviours that challenge.

What are the possible risks and burdens of taking part?

We will be asking you to think about times when your child or person you care for shows behaviours that challenge. Some parents or caregivers may find this distressing. You will be able to stop the interview at any time if you feel it is too upsetting.

Your decision to participate in this study will not impact your right or the right of your child/person you care for to access services.

The online consent forms and questionnaires are created through 'Qualtrics' and hosted on highly secure servers that comply with General Data Protection Regulations. However, as with all online activity, there

is a risk that unauthorised individuals (hackers) may access data. If you are uncomfortable with this risk, or simply would prefer a paper copy of the consent forms, please contact the research team who can put one in the post to you.

What will happen to the results of the study?

The results of this study will be published in scientific journals and/or presented at conferences. If the results of the study are published, your identity will remain confidential. Anonymised quotes may be used in publications resulting from the study. Research findings will also be published in newsletters of support groups and educational institutions.

A lay summary of the results of the study will be available for participants when the study has been completed and the researchers will ask if you would like to receive a copy. All participants will also receive an individualized feedback report that will detail their responses on standardised measures completed as part of the study.

The anonymized results may be shared with the company providing funding for this study. The results of the study will also be used in Lauren Shelley's PhD thesis.

Expenses and payments

Participants will not incur any expenses as a result of taking part in this study. However, if you complete the interview study, you will be entered into a prize draw to win one of two £50 online vouchers. We will notify participants by 31/07/2022 if they have been successful.

Who is funding the research?

The study is being funded by The Baily Thomas Charitable Fund.

Who is organising this study and acting as data controller for the study?

Aston University is organising this study and acting as data controller for the study. You can find out more about how we use your information in Appendix A.

Who has reviewed the study?

This study was given a favorable ethical opinion by the London – Brighton and Sussex Research Ethics Committee.

What if I have a concern about my participation in the study?

If you have any concerns about your participation in this study, please speak to the research team and they will do their best to answer your questions. Contact details can be found at the end of this information sheet.

If the research team are unable to address your concerns or you wish to make a complaint about how the study is being conducted, you should contact the Aston University Research Integrity Office at research_governance@aston.ac.uk or telephone 0121 204 3000.

Research Team

This research is being led by:

Jane Waite (Lead Researcher)
Telephone: 0121 204 4307. Email: j.waite@aston.ac.uk

Lauren Shelley (Doctoral Researcher)
Telephone: 0121 204 3203. Email: shellel1@aston.ac.uk

Thank you for taking time to read this information sheet. If you have any questions regarding the study, please do not hesitate to ask a member of the research team.

Appendix A: Transparency Statement

Aston University takes its obligations under data and privacy law seriously and complies with the Data Protection Act 2018 ("DPA") and the General Data Protection Regulation (EU) 2016/679 as retained in UK law by the Data Protection, Privacy and Electronic Communications (Amendments etc) (EU Exit) Regulations 2019 ("the UK GDPR").

Aston University is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study. Aston University will process your personal data in order to register you as a participant and to manage your participation in the study. It will process your personal data on the grounds that it is necessary for the performance of a task carried out in the public interest (GDPR Article 6(1)(e). Aston University may process special categories of data about you which includes details about your health. Aston University will process this data on the grounds that it is necessary for statistical or research purposes (GDPR Article 9(2)(j)). Aston University will keep identifiable information about you for 6 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally identifiable information possible.

You can find out more about how we use your information at https://www.aston.ac.uk/about/statutes-ordinances-regulations/publication-scheme/policies-regulations/data-protection or by contacting our Data Protection Officer at dp_officer@aston.ac.uk.

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer who will investigate the matter. If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO).

Appendix 30: Chapter Four consent forms

Aston University
BIRMINGHAM UK

Behaviours that challenge in SATB2-associated syndrome

Interview Study

Chief Investigator: Dr Jane Waite

Consent Form A: For children with *SATB2*-associated syndrome who are able to provide assent to participate in the study

Section 1 (Assent): Please complete this section if you are a person with SATB2-associated syndrome. If needed, your parent/carer or the researcher can read this form to you and you can let them know your answers.

	Please circle
Has somebody else explained the project to you?	YES/NO
Have you asked all of the questions you want?	YES/NO
Have you had your questions answered in a way you understand?	YES/NO
Do you understand it is OK to stop taking part at any time?	YES/NO
We will tell your GP your parent/carer is taking part. Is that OK?	YES/NO
Are you happy for your parent/carer to take part?	YES/NO
The next question is optional:	
Are you happy for us to contact your parent/carer again in the future?	YES/NO

Please write your name here:_____

Please write the date here:_____

Name of researcher taking assent: _____

Aston University
BIRMINGHAM UK

Section 2 (Parent/carer consent): Please complete this section if you are a parent/carer/guardian of a person with SATB2-associated syndrome who has provided their assent/consent to participate in the study.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 3, 14.10.2021) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time, without giving any reason and without my or that of my child's/person I care for's legal rights being affected.	
3.	I agree to my personal data and that of my child/person I care for and data relating to me and that of my child/person I care for collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study, may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I agree to my child/person I care for's GP being informed of my participation in the study.	
6.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare or that of my child/person I care for they may need to breach my confidentiality.	
7.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
8.	I agree to the interview being audio recorded and to anonymised direct quotes being used in publications resulting from the study.	
9.	I agree to my anonymised data being used by research teams for future research.	
10.	I agree to take part in this study.	

Aston University
BIRMINGHAM UK

The following statements are optional:		
1.	I agree to complete a short interview about my child's/person I care for's ability.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of my child/person I care for that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome.	
3.	I agree to be contacted about the next stage of this research project to consider whether I would like to take part.	
4.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of Person receiving consent.	Date	Signature

Aston University
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Behaviours that challenge in *SATB2*-associated syndrome

Interview Study

Chief Investigator: Dr Jane Waite

Consent Form B: For parents/carers of a child under 16 years old who is not able to make an informed decision about parent/carer participation in the study.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 3, 14.10.2021) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I confirm that my child is not able to understand all the information needed to decide about participating in this study, but that I have shared as much information as possible with my child about the study.	
3.	I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time, without giving any reason and without my or that of my child's/person I care for's legal rights being affected.	
4.	I agree to my personal data and that of my child/person I care for and data relating to me and that of my child/person I care for collected during the study being processed as described in the Participant Information Sheet.	
5.	I understand that relevant sections of my data collected during the study may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
6.	I agree to my child/person I care for's GP being informed of my participation in the study.	
7.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare or that of my child/person I care for they may need to breach my confidentiality.	
8.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	

9.	I agree to the interview being audio recorded and to anonymised direct quotes being used in publications resulting from the study.	
10.	I agree to my anonymised data being used by research teams for future research.	
11.	I agree to take part in this study.	
The following statements are optional:		
5.	I agree to complete a short interview about my child's/person I care for's ability.	
6.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of my child/person I care for that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome.	
7.	I agree to be contacted about the next stage of this research project to consider whether I would like to take part.	
8.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of Person receiving consent.	Date	Signature

Aston University
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Behaviours that challenge in *SATB2*-associated syndrome

Interview study

Chief Investigator: Dr Jane Waite

Consent Form E: For adults with *SATB2*-associated syndrome who are able to provide consent to participate in the study.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 3, 14.10.2021) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason and without my legal rights being affected.	
3.	I agree to my personal data and data relating to me collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I agree to my GP being informed of my participation in the study.	
6.	I understand that if during the study I tell, or my parent/carer tells the research team something that causes them to have concerns in relation to my health and/or welfare they may need to breach my confidentiality.	
7.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
8.	I agree to the interview with my parent/carer being audio recorded and to anonymised direct quotes being used in publications resulting from the study.	
9.	I agree to my anonymised data being used by research teams for future research.	
10.	I agree to take part in this study.	

The following statements are optional:		
1.	I agree to my parent/carer completing a short interview about my ability.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of for the individual with SATB2-associated syndrome that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome	
3.	I agree to be contacted about the next stage of this research project to consider whether I would like to take part.	
4.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of Person receiving consent	Date	Signature

Aston University
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Behaviours that challenge in *SATB2*-associated syndrome

Interview study

Chief Investigator: Dr Jane Waite

Consent Form D: For parents/carers of individuals over the age of 16 and able to make an informed decision about participation in the study.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 3, 14.10.2021) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason and without my legal rights being affected.	
3.	I agree to my personal data and data relating to me collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I agree to the person with *SATB2*-associated syndrome's GP being informed of my participation in the study.	
6.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare or that of the individual with *SATB2*-associated syndrome they may need to breach my confidentiality.	
7.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
8.	I agree to the interview being audio recorded and to anonymised direct quotes being used in publications resulting from the study.	
9.	I agree to my anonymised data being used by research teams for future research.	
10.	I agree to take part in this study.	

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The following statements are optional:		
1.	I agree to completing a short telephone interview about the individual with *SATB2*-associated syndrome's ability.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of for the individual with *SATB2*-associated syndrome that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome	
3.	I agree to be contacted about future research projects to consider if I would like to take part.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of Person receiving consent	Date	Signature

Appendix Thirty

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Behaviours that challenge in *SATB2*-associated syndrome

Interview study

Chief Investigator: Dr Jane Waite

Consent Form C: For a personal/nominated consultee of a person with *SATB2*-associated syndrome who is over the age of 16 and not able to provide consent.

Before deciding whether to participate, please ensure you read the information on acting as a personal/nominated consultee in the attached document for the person you care for.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 3, 14.10.2021) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason and without my legal rights being affected.	
3.	I agree to my personal data and data relating to me collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare they may need to breach confidentiality.	
6.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
7.	I agree to the interview being audio recorded and to anonymised direct quotes being used in publications resulting from the study.	
8.	I agree to my anonymised data being used by research teams for future research.	
9.	I agree to take part in this study.	

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The following statements are optional:		
1.	I agree to complete a short interview about the ability of the person for whom I am acting as a consultee.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of the person for whom I am acting as consultee that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome	
3.	I agree to be contacted about the next stage of this research project.	
4.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of consultee	Date	Signature
_____	_____	_____
Name of Person receiving declaration.	Date	Signature

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Behaviours that challenge in *SATB2*-associated syndrome

Interview study

Chief Investigator: Dr Jane Waite

Declaration Form A: For a personal/nominated consultee of a person with *SATB2*-associated syndrome who is over the age of 16 and not able to provide consent.

Before deciding whether to participate, please ensure you read the information on acting as a personal/nominated consultee in the attached document for the person you care for.

Please initial boxes

1.	I have been consulted about (name of participant) _____'s participation in the above research project. I confirm that I have read and understand the Participant Information Sheet (Version 3, 14.10.2021) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I confirm that in my opinion he/she would have no objection to participating in the study.	
3.	I understand that the participation of the person for whom I am acting as a consultee is voluntary and that I am free to withdraw them at any time, without giving any reason and without their legal rights being affected.	
4.	I agree to the person for who I am acting as consultee's personal data and data relating to him/her collected during the study being processed as described in the Participant Information Sheet.	
5.	I understand that relevant sections of his/her data collected during the study may be looked at by responsible individuals from Aston University or from regulatory authorities, where it is relevant to their taking part in this research. In my opinion he/she would have no objection to this.	
6.	I agree to his/her GP being informed of their participation in the study.	
7.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to the health and/or welfare of the person for whom I am acting as consultee, they may need to breach confidentiality.	
8.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). In my opinion he/she would have no objection to this risk.	
9.	I confirm that in my opinion he/she would have no objection to the interview being audio recorded and to anonymised direct quotes being used in publications resulting from the study.	
10.	I confirm that in my opinion he/she would have no objection to their anonymised data being used by research teams for future research.	

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The following statements are optional:		
1.	I confirm that in my opinion he/she should have no objection to me completing a short interview about his/her ability.	
2.	I agree to be contacted about the next stage of this research project to consider whether the person for whom I am acting as consultee would like to take part.	
3.	I agree to my personal data being processed for the purposes of inviting me and the person for whom I am acting as consultee to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of consultee	Date	Signature
_____	_____	_____
Name of Person receiving declaration.	Date	Signature

Appendix 31: Chapters Four and Five consultee information sheet

Personal and Nominated Consultee Information Sheet

Please read this information sheet if you care for a person who you have judged *is not* able to make an 'informed' decision about whether they would like to take part in the study or *is not* able to communicate that decision to you.

If you are an unpaid carer (e.g. parent, legal guardian etc) we would like to invite you to act as a ***personal consultee*** for the person that you care for.

If you are a paid carer (e.g. paid carer, key worker, support worker etc) and there are no unpaid carers (e.g. parent, legal guardian etc) to act as a personal consultee for the person you care for then we would like to invite you to act as a ***nominated consultee*** (go to page 3).

Information for Personal Consultees

What is a Personal Consultee?

To understand illness and disability, and to improve treatment and care, research is essential. That research may focus on the people with the illness or disability or on children under the age of 16, and may invite those people to participate. Some people will have capacity to make their own decision whether to take part in the research.

Others, possibly the youngest children or those most affected by the illness or disability, may not have that capacity. They may not be able to understand enough of the research to be able to give 'informed consent'. They may not be able to communicate a decision.

The research provisions of the Mental Capacity Act are designed to allow such people to take part in research even though they cannot give valid consent of their own. Instead of asking the research participant for consent, the researcher asks a consultee for an opinion on whether the research participant would have wished to take part in the research.

Who can be a personal consultee?

Any person interested in the welfare of the proposed participant, for example:

- A family member, unpaid carer or friend
- A person acting under a Lasting Power of Attorney
- A court appointed deputy

Who cannot be a personal consultee?

- Paid carers and professionals (if you are a paid carer or professional please refer to page 3)
- People connected with the research (e.g. members of the research team)

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Why have I been asked?

You have been asked to act as a personal consultee by a researcher because the researcher thinks you might be willing and able to do this because of your close relation with the proposed research participant.

If I agree to be a personal consultee, what will I have to do?

You will need to think about what the proposed participant's wishes and feelings about the research would be if they had capacity to make an informed decision and decide whether in your view the person should be involved in the research or not. This means you need to

- Look at the study information sheet.
- Think about whether the person would want to be involved in the research project if he or she had the capacity to make that decision.

You should not put forward your personal views on participation in the specific project or research in general. You must consider only what the person's views and interests are or would likely be. You should think about:

- What the broad aims of the research and the practicalities of taking part will mean for the proposed participant.
- How the specific activities in the research might impact the participant. For example, if the study involves activities in the afternoon when the person is most tired they might find it a strain or the research might involve an activity that the person particularly enjoys and thus would give them more pleasure.
- Any view previously expressed by the person on the overall nature of the research.

If you advise that the proposed participant would not have wanted to be involved in the research, they cannot be included in the research.

If you advise that the proposed participant would want to be involved, they may be included in the research. If the research commences but the person shows any sign at any stage that they are not happy to be involved in the research you can change your advice at any time without giving a reason, whereby the researcher must withdraw the person from the research. If the person seems unhappy at any point or shows any signs of objection, then they will be withdrawn from the research.

This project has received a favourable opinion by London – Brighton and Sussex Research Ethics committee. If you wish to see proof of approval from this body, or you wish to discuss any concerns about acting as a

personal consultee for the person that you care for, please contact Jane Waite on 0121 204 4307 or by email at j.waite@aston.ac.uk or Lauren Shelley by email at shellel1@aston.ac.uk

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I don't want to be a personal consultee/ I am a paid carer and so cannot be a personal consultee- what do I do?

Please try to suggest an alternative person who might like to act as a personal consultee for the potential participant, please pass the project information pack on to that person.

Where can I get more information and guidance?

More information is available from:

Department for Constitutional Affairs (2007) *Mental Capacity Act 2005 Code of Practice*

http://www.dca.gov.uk/legal-policy/mental-capacity/mca-cp.pdf

Department of Health (2007) *Guidance on nominating a consultee for research involving adults who lack capacity to consent* (consultation)

http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_076207

Mental Capacity Implementation Programme (2007) *Making Decisions: a guide for family, friends and unpaid carers. Second edition*

http://www.dca.gov.uk/legal-policy/mental-capacity/mibooklets/booklet02.pdf

A printed copy of this booklet is available by telephoning 023 80878038.

I have decided that I want to be a personal consultee- what do I do?

Please go back to the Information Sheet enclosed with this form and continue reading.

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Information for Nominated Consultees

What is a Nominated Consultee?

In order to understand illness and disability, and to improve treatment and care, research is essential. That research may focus on the people with the illness or disability or on children under the age of 16, and may invite those people to participate. Some people will have capacity to make their own decision whether to take part in the research.

Others, possibly the youngest children or those most affected by the illness or disability, may not have that capacity. They may not be able to understand enough of the research to be able to give 'informed consent'. They may not be able to communicate a decision. The research provisions of the Mental Capacity Act are designed to allow such people to take part in research even though they cannot give valid consent of their own.

First, the research has to be approved by a Research Ethics Committee. Then, instead of asking the research participant for consent, the researcher must ask a consultee for an opinion whether the research participant would have wished to take part in the research.

Who can be a nominated consultee?

- Any person interested in the welfare of the proposed participant who works with the participant in a professional capacity.

Who cannot be a nominated consultee?

- People connected with the research (e.g. members of the research team)

Why have I been asked?

You have been asked to act as a nominated consultee by a researcher because the researcher thinks you might be willing and able to do this because of your professional relationship with the proposed research participant.

If I agree to be a nominated consultee, what will I have to do?

You will need to think about what the proposed participant's wishes and feelings about the research would be if they had capacity to make an informed decision and decide whether in your view the person should be involved in the research or not. This means you need to

- Look at the study information sheet.
- Think about whether or not the person would want to be involved in the research project if he or she had the capacity to make that decision.

- You may need to seek the advice of friends/ family/ other paid carers of the person you care for in order for you to best advise us on what the person's wishes and feelings would be.

You should not put forward your personal views on participation in the specific project or research in general, you must consider only what the person's views and interests are or would likely be. You should think about:

- What the broad aims of the research and the practicalities of taking part will mean for the proposed participant.
- How the specific activities in the research might impact the participant. For example, if the study involves activities in the afternoon when the person is most tired they might find it a strain or the research might involve an activity that the person particularly enjoys and thus would give them more pleasure.
- Any view previously expressed by the person on the overall nature of the research.

If you advise that the proposed participant would not have wanted to be involved in the research, they cannot be included in the research.

If you advise that the proposed participant would want to be involved, they may be included in the research. If the research commences but the person shows any sign at any stage that they are not happy to be involved in the research you can change your advice at any time without giving a reason, whereby the researcher must withdraw the person from the research. If the person seems unhappy at any point or shows any signs of objection, then they will be withdrawn from the research.

This project has received a favourable opinion by London - Brighton and Sussex Research Ethics Committee. If you wish to see proof of approval from this body, or you wish to discuss any concerns about acting as a personal consultee for the person that you care for, please contact Jane Waite on 0121 204 4307 or by email at j.waite@aston.ac.uk or Lauren Shelley by email at shellel1@aston.ac.uk

I don't want to be a nominated consultee - what do I do?

Please try to suggest an alternative person who might like to act as a nominated consultee for the potential participant, please pass the project information pack on to that person.

If no-one can be found who is willing and able to act as a consultee for the person you care for then the person will not be able to participate in the research study.

Where can I get more information and guidance?

More information is available from:

Department for Constitutional Affairs (2007) *Mental Capacity Act 2005 Code of Practice*

http://www.dca.gov.uk/legal-policy/mental-capacity/mca-cp.pdf

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Department of Health (2007) *Guidance on nominating a consultee for research involving adults who lack capacity to consent* (consultation)

http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_076207

Mental Capacity Implementation Programme (2007) *Making Decisions: a guide for family, friends and unpaid carers. Second edition*

http://www.dca.gov.uk/legal-policy/mental-capacity/mibooklets/booklet02.pdf

A printed copy of this booklet is available by telephoning 023 80878038.

I have decided that I want to be a nominated consultee- what do I do?

Please go back to the Information Sheet included with this study and continue reading.

Appendix 32: Chapter Four content analysis infrequent codes

Appendices Table 10

Content analysis: Infrequently reported (n≤3) caregiver-led strategies to manage behaviour

Strategy	n (%)	Description	Example codes	Example quotes
Caregiver-led strategies				
Preventative				
Avoid communication too far in advance	3 (9.1)	Caregiver avoids communicating things in advance/ahead of time.	Don't tell in advance, don't tell ahead of time.	*"I sometimes just try not to tell him things too far in advance because I know then he will get frustrated with waiting or um not understanding why things aren't happening straight away"* – (Participant 2) *"we never tell her anything ahead of time because […] it would take over her thoughts and she wouldn't be able to talk about anything else or concentrate on anything […] it's just not a good idea"* – (Participant 18)
Provide stability	2 (6.1)	Ensuring person is always a caregiver with the person to increase sense of stability.	Person with him, caregiver around him.	*"as long as he's got his one stable person that he wants there, as long as he feels in control where he's got that one person with him then he can, he can be ok"* – (Participant 32)
Calm environment	2 (6.1)	Creating a calm environment to help keep the person relaxed (e.g., play smooth music).	Keep calm in house, keep chill, calm around him.	*"I kind of try and play kind of like smooth jazz or smooth like magic music in the house when he's playing just to keep his… keep it quite calm […] which kind of relaxes him"* – (Participant 17)
Limit choice	1 (3.0)	Reduce choices to provide options while preventing distractions.	Not too many choices.	*"we make sure that there are not too many choices around […] she can choose, but we don't give her too much choices, because then she gets distracted and wants everything [and] not everything can happen"* – (Participant 4)

Appendices Table 10 Continued

Strategy	n (%)	Description	Example codes	Example quotes
Remove distractions	1 (3.0)	Removing distractions (e.g., toys) that may impact activities.	Clear toys away, nothing distracting.	*"try and keep everything away from sort of try and clear her toys away there's nothing distracting her"* – (Participant 3)
Active				
Provide food	2 (6.1)	Person offered food or snack.	Give food/snack.	*"I can give her food so the ways I can stop her tantrums typically are erm with a snack"* – (Participant 23)
PRN medication	1 (3.0)	Medication given as required to help alleviate behaviour.	Medicated.	*"[sometimes] she has to be medicated in order to kind of move on from being upset"* – (Participant 18)
Background				
Trial run activities	1 (3.0)	Trial running or test running high-risk activities that might cause upset.	Trial run activities	*"where we think there are sort of like high-risk activities for her getting very upset, we'll sort of like trial run them or test them in different ways to get more comfort where we wouldn't with a different child"* – (Participant 1)
Adaptations to home environment	2 (6.1)	Adaptation to the home environment e.g., soft toys in room instead of books, oven mitts on doors.	Oven mitts on doors, floor mats, soft toys.	*"I've got oven mitts tied around the doors, so it's around the handle so that he can't slam the door […] and I've got matts over - because we've got wooden floors in the hall so I've got lots of mats on the floor so that when he drops bags, zips don't clatter on the floor, it lands on the carpet, so we have to do all those things to try and […] stop X's triggers"* – (Participant 7) *"we've actually moved loads of stuff out of his bedroom, 'cause of what he throws, he had books and all sorts, but he was throwing it all down the stairs and denting the walls and all sorts so he's got nice like, lots of like soft toys in there now"* – (Participant 14)

Appendices Table 10 Continued

Strategy	n (%)	Description	Example codes	Example quotes
Changes to diet	2 (6.1)	Adapting individuals' diet and nutrition.	Diet change, modify nutrition.	*"I completely changed his diet […] and it completely changed the behaviour. So, he went from being like just kind of not aware of his physical space and pushing kids over and stuff like that, to much calmer, much more manageable, and so now most of those behaviours are pretty much gone"* – (Participant 10) *"I'm constantly trying to modify and try different things, within a safe you know realm with like nutrition and different things but I can tell you that it does affect him, at least"* – (Participant 28)

Note. PRN=Pro re nata (as required).

Appendix 33: Chapter Five invitation letter

Insert support group logo /CNDD logo as appropriate

Aston University

Dear [insert name of parent/carer],

A new research project is being carried out at Aston University. We wanted to let you know about this study so that you can decide whether you would like to take part.

The research project involves the research team you on how to complete some game-based assessments with the person you care for. There are also some questionnaires that we would like you to complete. The research is being carried out to understand the factors that may influence behaviour in individuals with *SATB2*-associated syndrome. The research can take place in your home, face-to-face or remotely via video call, depending on your preference and location, and it will take no longer than 4 hours to complete. For families in the UK there will be an option for the research team to administer the game-based tasks with your child/person you care for. When the research team have analysed the information, each family that participates will receive a personalised feedback report.

If you or your child/person you care for are interested in finding out more about the study and to decide whether you would like to take part, please complete one of the following:

1. Contact Lauren Shelley (Doctoral Researcher) on shellel1@aston.ac.uk or 0121 204 3203 to get further information about the research.
2. Access an online expression of interest form using the link below to register your interest in the research project. A member of the research team will then contact you to provide more information about the study.
   [insert link to expression of interest form]
   Your reference number: [INSERT ID; to be included for CNDD participant database invites]

If you are unclear about any aspect of the study or have any questions please contact Lauren Shelley, 0121 204 3203; shellel1@aston.ac.uk, or Dr Jane Waite at Aston University, 0121 204 4307; j.waite@aston.ac.uk.

Thank you for your time and we look forward to hearing from you.

Yours sincerely,

Dr Jane Waite
Lecturer and Clinical Psychologist
Aston University

Lauren Shelley
Doctoral Researcher
Aston University

If you are on the mailing list of a SATB2-associated syndrome support group, you may have already received an invitation to participate in this research. If this is the case, we apologise for contacting you more than once. (To be included for recruitment from the participant database held at the CNDD)

AppendixJ_Stage2_Invite_06.07.2022_V2
IRAS ID: 296378

Appendix 34: Chapter Five participant information sheets

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Behaviours that challenge in SATB2-associated syndrome

Participant Information Sheet

Direct Assessment study: Parents/carers of children with SAS

Version 4 06.07.2022

Invitation

We would like to invite you to take part in a research study.

If you have questions or would like a verbal explanation of this study, contact Jane Waite (Lead Researcher) on 0121 204 4307 or j.waite@aston.ac.uk, or Lauren Shelley (Doctoral Researcher) on 0121 204 3203 or shellel1@aston.ac.uk.

Before you decide if you would like to participate, take time to read the following information carefully and, if you wish, discuss it with others such as your family, friends, or colleagues.

Please ask a member of the research team, whose contact details can be found at the end of this information sheet, if there is anything that is not clear or if you would like more information before you make your decision.

What is the purpose of the study?

We would like to invite parents or carers of individuals with SATB2-associated syndrome, to take part in a research study about behaviours shown by the person they care for. This research study follows on from a recent questionnaire and interview study, however, the study is open to all families, even if you have not previously participated in our research. We are now also inviting individuals with SATB2-associated syndrome, aged between 4 and 15 years, to take part in a research assessment day. We hope that the study will help healthcare professionals working with individuals with SATB2-associated syndrome to understand the factors that influence behaviours that challenge.

The aim is to understand more about behaviour that challenges, such as behaviours directed towards others (e.g., hair pulling or hitting) or self-injurious behaviours, in SATB2-associated syndrome, and to examine which person and environmental characteristics are associated with these behaviours.

The study forms part of a larger study of behaviours that challenge in people with SATB2-associated syndrome that is being conducted as part of Lauren Shelley's PhD studies. This PhD research is endorsed by the SATB2 Gene Foundation.

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Why have I been invited?

[You are being invited to take part in this study because you recently took part in a questionnaire/interview study and indicated that you are happy for us to contact you for research purposes.] OR [You are being invited to take part in this study because records held by the [SATB2 Gene Foundation/SATB2 Gene Trust UK/Cerebra Network for Neurodevelopmental Disorders (delete as appropriate)] indicate that you are a parent or carer of a person with SATB2-associated syndrome, aged between 4 and 15 years, and that [you are happy to be contacted for research purposes/have consented to be contacted about future research (delete as appropriate)]].

Alternatively, we may have contacted you because you responded to an advert about the study.

What will happen to me if I take part?

If you would like your child to participate in the research, you will be asked to sign a consent form (see more information about this below). After you have consented to take part in the study, you will be asked to:

- Complete some online questionnaires about your child
- Complete an interview about your child's adaptive functioning over the phone or via online conferencing (e.g., Zoom or Microsoft Teams)
- Take part in a research assessment day including some fun play-based games and activities to be completed via online video conferencing with your child and the research team

The research assessment day will take place at your home. We will send a package containing all of the toys and equipment for you to use on the research assessment day. We will also send instructions for how to play the games and will guide you through each step using in-ear headphones. You can keep the toys as a gift for you and your child. For families located in the UK, it may be possible for the research team to come to your home for the assessment day.

Tasks on the assessment day will all be play-based games and activities, which will be engaging for your child. We will use picture cards, puzzles and role-playing activities to learn about your child's behaviour. We will take regular breaks in between tasks. The assessment day will be arranged at a time convenient for you and will last no longer than 4 hours overall. The assessment day can be split into shorter games sessions if preferred.

Completing the questionnaires and interview is expected to take approximately 2 hours of your time, at a time that is convenient for you. The play-based assessments with you and your child are estimated to take 1.5 hours in total. Some children may be able to complete all assessments in one session, but others will require frequent breaks between activities meaning that assessments could be spread over two sessions to suit your child.

With your permission, we will video record the tasks that you and your child complete during the assessment day. Your child and the behaviour of people in your child's immediate surroundings will be recorded using the webcam of your laptop/tablet device so that we can code some of the tasks after they have been completed.

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The videos will be captured via online conferencing facilities and stored on the University's secure online servers. The video recordings will only be seen by members of the research team.

We will ask you to provide a copy of a genetic/clinical confirmation letter confirming the person you care for's diagnosis of SATB2-associated syndrome, if possible, from a GP, paediatrician, geneticist or other professional. This will help us to confirm diagnosis on the direct assessment day. A copy of this letter can be uploaded onto an online form, posted to the research team, or given to the research team on the research assessment day. This is entirely optional and there is no obligation to provide this document to participate in the study.

We will be collecting information from participants between October 2022 and July 2023. After that we will spend some time understanding the data and writing reports. This means that the study will be finished in October 2023. You will be given the option for us to retain your details if you would like to be invited to take part in future research projects.

If you would like to take part, you will need to complete a consent form. If your child can understand what is involved in the study, you will also need to support them to complete an assent/consent form. If your child has the capacity to understand some or all the information about this study, you should explain the study to them in a way they understand. You can also request a copy of an accessible information sheet from the researcher (shellel1@aston.ac.uk).

If you are completing the consent forms online, the survey will direct you to the correct consent forms based on your responses.

If you are completing a paper copy of the consent forms and *your child can fully understand* what is involved in the study, you and your child should complete the yellow assent/consent forms. If *your child is not able to understand* what is involved in the study, you should complete the blue consent form. Once completed you can send these forms back to us in the prepaid envelope provided.

Once we have received your consent, we will contact you to arrange a convenient time for the interview about the person you care for's adaptive ability and a date for the research assessment day.

Joining up research studies to better understand SATB2-associated syndrome:

If you have participated in a research study investigating the behavioural phenotype of SATB2-associated syndrome, led by the Cerebra Network for Neurodevelopmental Disorders (CNDD), University of Birmingham, we will also ask whether you are happy for us to use your unique reference number to extract and link your responses from the CNDD study to the information you provide in this study. This is optional. The information from the ongoing CNDD study will help us to understand the results of the current study.

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Do I have to take part?

No. It is up to you to decide whether or not you wish to take part.

If you do decide to participate, you will be asked to sign and date a consent form. After the assessment day, you have 14 days to withdraw from the study, without giving a reason. After this period, we may have coded data from the video recordings, and you will be unable to withdraw your data. If you decide to withdraw within the 14 days following the assessment day, we will not delete your data unless you tell us to.

Will my taking part in this study be kept confidential?

Yes. A code will be attached to all the data you provide to maintain confidentiality.

Your personal data (name and contact details) will only be used if the researchers need to contact you to arrange study visits or collect data by phone. Analysis of your data will be undertaken using coded data.

The data we collect will be stored in a secure document store (paper records) or electronically on a secure encrypted mobile device, password protected computer server or secure cloud storage device.
To ensure the quality of the research, Aston University may need to access your data to check that the data has been recorded accurately. If this is required, your personal data will be treated as confidential by the individuals accessing your data.

How will the video recordings made during the study be managed?

The video recordings will be destroyed as soon as the research team have analysed the information in them to answer the research question.

We will ensure that anything from the analysis of the videos that is included in the reporting of the study will be anonymous.

Will my GP be informed of my involvement in the study?

With your consent, we will notify the GP of your child/person you care for with SAS that you are taking part in this research project about your child/person you care for. A copy of the feedback report for your child will be shared with their GP at the end of this study.

If we become aware that your child may be experiencing an undiagnosed health difficulty, Dr Jane Waite will write to the GP of your child/person you care for to pass on this information. If you live outside of the UK, we will advise you to notify the people involved in your child or person you care for's usual care of your involvement in the study.

If it becomes apparent that you are showing signs of distress or an undiagnosed health difficulty, we will advise you to contact your GP, specialist, or the relevant national contact of a SATB2-associated syndrome support group.

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What happens if I tell you something that concerns you about my health or welfare or that of the person I care for?

In the unlikely event of this happening, we will discuss with you how this should be addressed. If necessary, to protect you and the person you care for, we will report your concern to the appropriate person or bodies.

Any request for advice concerning your child/person you care for with SATB2-associated syndrome will be passed on to Dr Jane Waite (Clinical Psychologist), who will provide information about accessing local support.

What are the possible benefits of taking part?

Following your child's participation in the study, you will receive an individualised feedback report summarising the results of the assessments conducted. This report may be useful to you and the health and education professionals involved with your child to highlight strengths and difficulties that your child experiences and identify resources that may be useful for them. This study will help us to better understand the factors that influence behaviour in SATB2-associated syndrome. It is hoped that the research will help us to find more about the needs of people with SATB2-associated syndrome who show behaviours that challenge.

What are the possible risks and burdens of taking part?

The questionnaires and interview that you will be asked to complete include questions about your child's behaviour that you might find difficult to talk about. If you feel uncomfortable you can discuss this with the research team at any stage of participation.

Your decision to participate in this study will not impact your right or the right of your child or person you care for to access services.

The tasks we will do on the assessment day are tasks which are commonly used in individuals with neurodevelopmental conditions and intellectual disability. However, some individuals can find some of the tasks difficult. We can take regular breaks to make the tasks easier for the person you care for. Some children taking part in the study may display challenging behaviours (e.g. aggression, destruction of property) and self-injurious behaviour (e.g. head-banging, biting self, eye poking). These behaviours may occur in the presence of the researcher or when they are interacting with you during the study activities. If your child does display any challenging behaviours during the play session the research team will discuss with you how to proceed or if activities should cease. Before the play session, we will ask you about any challenging behaviours that your child shows and, if appropriate, create a risk-management plan to keep your child and those around them safe during the play session.

The online consent forms and questionnaires are created through 'Qualtrics' and hosted on highly secure servers that comply with General Data Protection Regulations. However, as with all online activity, there is a risk that unauthorised individuals (hackers) may access data. If you are uncomfortable with this risk, or simply would prefer a paper copy of the consent forms, please contact the research team who can put one in the post to you.

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What will happen to the results of the study?

The results of this study will be published in scientific journals and/or presented at conferences. If the results of the study are published, your identity will remain confidential. Research findings will also be published in newsletters of support groups and educational institutions.

A lay summary of the results of the study will be available for participants when the study has been completed and the researchers will ask if you would like to receive a copy.

The anonymized results may be shared with the company providing funding for this study. The results of the study will also be used in Lauren Shelley's PhD thesis.

Expenses and payments

At the end of the study, all participants will be entered into a prize draw to win a £50 online voucher. We will notify participants by October 2023 if they have been successful.

If you decide to come to Aston University for the research assessment day, your travel and accommodation expenses will be reimbursed.

Who is funding the research?

The study is being funded by The Baily Thomas Charitable Fund.

Who is organising this study and acting as data controller for the study?

Aston University is organising this study and acting as data controller for the study. You can find out more about how we use your information in Appendix A.

Who has reviewed the study?

This study was given a favorable ethical opinion by the London – Brighton and Sussex Research Ethics Committee.

What if I have a concern about my participation in the study?

If you have any concerns about your participation in this study, please speak to the research team and they will do their best to answer your questions. Contact details can be found at the end of this information sheet.

If the research team are unable to address your concerns or you wish to make a complaint about how the study is being conducted, you should contact the Aston University Research Integrity Office at research_governance@aston.ac.uk or telephone 0121 204 3000.

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Research Team

This research is being led by:

Jane Waite (Lead Researcher)
Telephone: 0121 204 4307. Email: j.waite@aston.ac.uk

Lauren Shelley (Doctoral Researcher)
Telephone: 0121 204 3203. Email: shellel1@aston.ac.uk

Thank you for taking time to read this information sheet. If you have any questions regarding the study, please do not hesitate to ask a member of the research team.

Appendix A: Transparency statement

Aston University takes its obligations under data and privacy law seriously and complies with the Data Protection Act 2018 ("DPA") and the General Data Protection Regulation (EU) 2016/679 as retained in UK law by the Data Protection, Privacy and Electronic Communications (Amendments etc) (EU Exit) Regulations 2019 ("the UK GDPR").

Aston University is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study. Aston University will process your personal data in order to register you as a participant and to manage your participation in the study. It will process your personal data on the grounds that it is necessary for the performance of a task carried out in the public interest (GDPR Article 6(1)(e). Aston University may process special categories of data about you which includes details about your health. Aston University will process this data on the grounds that it is necessary for statistical or research purposes (GDPR Article 9(2)(j)). Aston University will keep identifiable information about you for 6 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally identifiable information possible.

You can find out more about how we use your information at https://www.aston.ac.uk/about/statutes-ordinances-regulations/publication-scheme/policies-regulations/data-protection or by contacting our Data Protection Officer at dp_officer@aston.ac.uk.

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer who will investigate the matter. If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO).

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Behaviours that challenge in SATB2-associated syndrome

Participant Information Sheet

Direct Assessment study: Parents/carers of individuals with SAS aged 16 years and over

Version 4 06.07.2022

Invitation

We would like to invite you to take part in a research study.

If you have questions or would like a verbal explanation of this study, contact Jane Waite (Lead Researcher) on 0121 204 4307 or j.waite@aston.ac.uk, or Lauren Shelley (Doctoral Researcher) on 0121 204 3203 or shellel1@aston.ac.uk.

Before you decide if you would like to participate, take time to read the following information carefully and, if you wish, discuss it with others such as your family, friends, or colleagues.

Please ask a member of the research team, whose contact details can be found at the end of this information sheet, if there is anything that is not clear or if you would like more information before you make your decision.

What is the purpose of the study?

We would like to invite parents or carers of individuals with SATB2-associated syndrome, to take part in a research study about behaviours shown by the person they care for. This research study follows on from a recent questionnaire and interview study, however, the study is open to all families, even if you have not previously participated in our research. We are now also inviting individuals with SATB2-associated syndrome, aged 16 years and over, to take part in a research assessment day. We hope that the study will help healthcare professionals working with individuals with SATB2-associated syndrome to understand the factors that influence behaviours that challenge.

The aim is to understand more about behaviour that challenges, such as behaviours directed towards others (e.g., hair pulling or hitting) or self-injurious behaviours, in SATB2-associated syndrome, and to examine which person and environmental characteristics are associated with these behaviours.

The study forms part of a larger study of behaviours that challenge in people with SATB2-associated syndrome that is being conducted as part of Lauren Shelley's PhD studies. This PhD research is endorsed by the SATB2 Gene Foundation.

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Why have I been invited?

[You are being invited to take part in this study because you recently took part in a questionnaire/interview study and indicated that you are happy for us to contact you for research purposes.] OR [You are being invited to take part in this study because records held by the [SATB2 Gene Foundation/SATB2 Gene Trust UK/Cerebra Network for Neurodevelopmental Disorders (delete as appropriate)] indicate that you are a parent or carer of a person with SATB2-associated syndrome, aged 16 years or over, and that [you are happy to be contacted for research purposes/have consented to be contacted about future research (delete as appropriate)]].

Alternatively, we may have contacted you because you responded to an advert about the study.

What will happen to me if I take part?

If you would like the person you care for to participate in the research, you will be asked to sign a consent form (see more information about this below). After you have consented to take part in the study, you will be asked to:

- Complete some online questionnaires about the person you care for
- Complete an interview about the person you care for's adaptive functioning over the phone or via online conferencing (e.g., Zoom or Microsoft Teams)
- Take part in a research assessment day including some fun play-based games and activities to be completed via online video conferencing with the person you care for and the research team

The research assessment day will take place at your home. We will send a package containing all of the toys and equipment for you to use on the research assessment day. We will also send instructions for how to play the games and will guide you through each step using in-ear headphones. You can keep the toys as a gift for you and the person you care for. For families located in the UK, it may be possible for the research team to come to your home for the assessment day.

Tasks on the assessment day will all be play-based games and activities, which will be engaging for the person you care for. We will use picture cards, puzzles and role-playing activities to learn about the person you care for's behaviour. We will take regular breaks in between tasks. The assessment day will be arranged at a time convenient for you and will last no longer than 4 hours overall. The assessment day can be split into shorter sessions if preferred.

Completing the questionnaires and interview is expected to take approximately 2 hours of your time, at a time that is convenient for you. The play-based assessments with you and the person you care for are estimated to take 1.5 hours in total. Some individuals may be able to complete all assessments in one session, but others will require frequent breaks between activities meaning that assessments could be spread over two sessions to suit the person you care for.

With your permission, we will video record the tasks that you and the person you care for complete during the assessment day. The person you care for and the behaviour of people in the person you care for's immediate surroundings will be recorded using the webcam of your laptop/tablet device so that we can code some of the tasks after they have been completed. The videos will be captured via online conferencing

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facilities and stored on the University's secure online servers. The video recordings will only be seen by members of the research team.

We will ask you to provide a copy of a genetic/clinical confirmation letter confirming the person you care for's diagnosis of SATB2-associated syndrome, if possible, from a GP, paediatrician, geneticist or other professional. This will help us to confirm diagnosis on the direct assessment day. A copy of this letter can be uploaded onto an online form, posted to the research team, or given to the research team on the research assessment day. This is entirely optional and there is no obligation to provide this document to participate in the study.

We will be collecting information from participants between October 2022 and July 2023. After that we will spend some time understanding the data and writing reports. This means that the study will be finished in October 2023. You will be given the option for us to retain your details if you would like to be invited to take part in future research projects.

If you would like to take part, you will need to complete a consent form. If the person you care for can understand what is involved in the study, you will also need to support them to complete an assent/consent form. If the person you care for has the capacity to understand some or all the information about this study, you should explain the study to them in a way they understand. You can also request a copy of an accessible information sheet from the researcher (shellel1@aston.ac.uk).

If you are completing the consent forms online, the survey will direct you to the correct consent forms based on your responses.

If you are completing a paper copy of the consent forms and *the person with SAS can fully understand* what is involved in the study, you and the person you care for should complete the yellow consent forms. If *the person with SAS is not able to understand* what is involved in the study, you should complete the blue consent and declaration forms. Once completed you can send these forms back to us in the prepaid envelope provided.

Once we have received your consent, we will contact you to arrange a convenient time for the interview about the person you care for's adaptive ability and a date for the research assessment day.

Joining up research studies to better understand SATB2-associated syndrome:

If you have participated in a research study investigating the behavioural phenotype of SATB2-associated syndrome, led by the Cerebra Network for Neurodevelopmental Disorders (CNDD), University of Birmingham, we will also ask whether you are happy for us to use your unique reference number to extract and link your responses from the CNDD study to the information you provide in this study. This is optional. The information from the ongoing CNDD study will help us to understand the results of the current study.

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Do I have to take part?

No. It is up to you to decide whether or not you wish to take part.

If you do decide to participate, you will be asked to sign and date a consent form. After the assessment day, you have 14 days to withdraw from the study, without giving a reason. After this period, we may have coded data from the video recordings, and you will be unable to withdraw your data. If you decide to withdraw within the 14 days following the assessment day, we will not delete your data unless you tell us to.

Will my taking part in this study be kept confidential?

Yes. A code will be attached to all the data you provide to maintain confidentiality.

Your personal data (name and contact details) will only be used if the researchers need to contact you to arrange study visits or collect data by phone. Analysis of your data will be undertaken using coded data.

The data we collect will be stored in a secure document store (paper records) or electronically on a secure encrypted mobile device, password protected computer server or secure cloud storage device.

To ensure the quality of the research, Aston University may need to access your data to check that the data has been recorded accurately. If this is required, your personal data will be treated as confidential by the individuals accessing your data.

How will the video recordings made during the study be managed?

The video recordings will be destroyed as soon as the research team have analysed the information in them to answer the research question.

We will ensure that anything from the analysis of the videos that is included in the reporting of the study will be anonymous.

Will my GP be informed of my involvement in the study?

With your consent, we will notify the GP of <u>your child/person you care for</u> with SAS that you are taking part in this research project about your child/person you care for. A copy of the feedback report for your child will be shared with their GP at the end of this study.

If we become aware that your child may be experiencing an undiagnosed health difficulty, Dr Jane Waite will write to the GP of your child/person you care for to pass on this information. If you live outside of the UK, we will advise you to notify the people involved in your child or person you care for's usual care of your involvement in the study.

If it becomes apparent that you are showing signs of distress or an undiagnosed health difficulty, we will advise you to contact your GP, specialist, or the relevant national contact of a SATB2-associated syndrome support group.

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What happens if I tell you something that concerns you about my health or welfare or that of the person I care for?

In the unlikely event of this happening, we will discuss with you how this should be addressed. If necessary, to protect you and the person you care for, we will report your concern to the appropriate person or bodies.

Any request for advice concerning your child/person you care for with SATB2-associated syndrome will be passed on to Dr Jane Waite (Clinical Psychologist), who will provide information about accessing local support.

What are the possible benefits of taking part?

Following the person you care for's participation in the study, you will receive an individualised feedback report summarising the results of the assessments conducted. This report may be useful to you and the health and education professionals involved with the person you care for to highlight strengths and difficulties that the person you care for experiences and identify resources that may be useful for them. This study will help us to better understand the factors that influence behaviour in SATB2-associated syndrome. It is hoped that the research will help us to find more about the needs of people with SATB2-associated syndrome who show behaviours that challenge.

What are the possible risks and burdens of taking part?

The questionnaires and interview that you will be asked to complete include questions about the person you care for's behaviour that you might find difficult to talk about. If you feel uncomfortable you can discuss this with the research team at any stage of participation.

Your decision to participate in this study will not impact your right or the right of your child or person you care for to access services.

The tasks we will do on the assessment day are tasks which are commonly used in individuals with neurodevelopmental conditions and intellectual disability. However, some individuals can find some of the tasks difficult. We can take regular breaks to make the tasks easier for the person you care for. Some children taking part in the study may display challenging behaviours (e.g. aggression, destruction of property) and self-injurious behaviour (e.g. head-banging, biting self, eye poking). These behaviours may occur in the presence of the researcher or when they are interacting with you during the study activities. If your child does display any challenging behaviours during the play session the research team will discuss with you how to proceed or if activities should cease. Before the play session, we will ask you about any challenging behaviours that your child shows and, if appropriate, create a risk-management plan to keep your child and those around them safe during the play session.

The online consent forms and questionnaires are created through 'Qualtrics' and hosted on highly secure servers that comply with General Data Protection Regulations. However, as with all online activity, there is a risk that unauthorised individuals (hackers) may access data. If you are uncomfortable with this risk, or simply would prefer a paper copy of the consent forms, please contact the research team who can put one in the post to you.

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What will happen to the results of the study?

The results of this study will be published in scientific journals and/or presented at conferences. If the results of the study are published, your identity will remain confidential. Research findings will also be published in newsletters of support groups and educational institutions.

A lay summary of the results of the study will be available for participants when the study has been completed and the researchers will ask if you would like to receive a copy.

The anonymized results may be shared with the company providing funding for this study. The results of the study will also be used in Lauren Shelley's PhD thesis.

Expenses and payments

At the end of the study, all participants will be entered into a prize draw to win a £50 online voucher. We will notify participants by October 2023 if they have been successful.

If you decide to come to Aston University for the research assessment day, your travel and accommodation expenses will be reimbursed.

Who is funding the research?

The study is being funded by The Baily Thomas Charitable Fund.

Who is organising this study and acting as data controller for the study?

Aston University is organising this study and acting as data controller for the study. You can find out more about how we use your information in Appendix A.

Who has reviewed the study?

This study was given a favorable ethical opinion by the London – Brighton and Sussex Research Ethics Committee.

What if I have a concern about my participation in the study?

If you have any concerns about your participation in this study, please speak to the research team and they will do their best to answer your questions. Contact details can be found at the end of this information sheet.

If the research team are unable to address your concerns or you wish to make a complaint about how the study is being conducted, you should contact the Aston University Research Integrity Office at research_governance@aston.ac.uk or telephone 0121 204 3000.

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Research Team

This research is being led by:

Jane Waite (Lead Researcher)
Telephone: 0121 204 4307. Email: j.waite@aston.ac.uk

Lauren Shelley (Doctoral Researcher)
Telephone: 0121 204 3203. Email: shellel1@aston.ac.uk

Thank you for taking time to read this information sheet. If you have any questions regarding the study, please do not hesitate to ask a member of the research team.

Appendix A: Transparency statement

Aston University takes its obligations under data and privacy law seriously and complies with the Data Protection Act 2018 ("DPA") and the General Data Protection Regulation (EU) 2016/679 as retained in UK law by the Data Protection, Privacy and Electronic Communications (Amendments etc) (EU Exit) Regulations 2019 ("the UK GDPR").

Aston University is the sponsor for this study based in the United Kingdom. We will be using information from you in order to undertake this study. Aston University will process your personal data in order to register you as a participant and to manage your participation in the study. It will process your personal data on the grounds that it is necessary for the performance of a task carried out in the public interest (GDPR Article 6(1)(e). Aston University may process special categories of data about you which includes details about your health. Aston University will process this data on the grounds that it is necessary for statistical or research purposes (GDPR Article 9(2)(j)). Aston University will keep identifiable information about you for 6 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally identifiable information possible.

You can find out more about how we use your information at https://www.aston.ac.uk/about/statutes-ordinances-regulations/publication-scheme/policies-regulations/data-protection or by contacting our Data Protection Officer at dp_officer@aston.ac.uk.

If you wish to raise a complaint on how we have handled your personal data, you can contact our Data Protection Officer who will investigate the matter. If you are not satisfied with our response or believe we are processing your personal data in a way that is not lawful you can complain to the Information Commissioner's Office (ICO).

Appendix 35: Chapter Five consent forms

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Behaviours that challenge in *SATB2*-associated syndrome

Direct Assessment Study

Chief Investigator: Dr Jane Waite

Consent Form A: For children with *SATB2*-associated syndrome who are able to provide assent to participate in the study

Section 1 (Assent): Please complete this section if you are a person with SATB2-associated syndrome. If needed, your parent/carer or the researcher can read this form to you and you can let them know your answers.

	Please circle
Has somebody else explained the project to you?	YES/NO
Have you asked all of the questions you want?	YES/NO
Have you had your questions answered in a way you understand?	YES/NO
Do you understand it is OK to stop taking part at any time?	YES/NO
We will tell your GP you are taking part and show them your results. Is that OK?	YES/NO
Is it ok if we video record you?	YES/NO
Are you happy for your parent/carer to take part?	YES/NO
The next question is optional:	
Are you happy for us to contact your parent/carer again in the future?	YES/NO

Please write your name here:_____

Please write the date here:_____

Name of researcher taking assent: _____

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Section 2 (Parent/carer consent): Please complete this section if you are a parent/carer/guardian of a person with SATB2-associated syndrome who has provided their assent/consent to participate in the study.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 4, 06.07.2022) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time, without giving any reason and without my or that of my child's/person I care for's legal rights being affected.	
3.	I agree to my personal data and that of my child/person I care for and data relating to me and that of my child/person I care for collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I agree to my child/person I care for's GP being informed of their/my participation in the study.	
6.	I agree to the feedback report arising from this study being shared with my child/person I care for's GP.	
7.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare or that of my child/person I care for they may need to breach my confidentiality.	
8.	I agree to the study visits/research assessment day being video recorded.	
9.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
10.	I agree to my anonymised data being used by research teams for future research.	
11.	I agree to take part in this study.	

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The following statements are optional:		
1.	I agree to complete a short interview about my child's/person I care for's ability.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of my child/person I care for that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome	
3.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	
4.	I agree to be contacted about future research projects to consider whether I would like to take part.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of Person receiving consent.	Date	Signature

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Behaviours that challenge in *SATB2*-associated syndrome

Direct Assessment Study

Chief Investigator: Dr Jane Waite

Consent Form B: For parents/carers of a child under 16 years old who is not able to make an informed decision about parent/carer participation in the study.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 4, 06.07.2022) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time, without giving any reason and without my or that of my child's/person I care for's legal rights being affected.	
3.	I agree to my personal data and that of my child/person I care for and data relating to me and that of my child/person I care for collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study, may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I agree to my child/person I care for's GP being informed of their/my participation in the study.	
6.	I agree to the feedback report arising from this study being shared with my child/person I care for's GP.	
7.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare or that of my child/person I care for they may need to breach my confidentiality.	
8.	I agree to the study visits/research assessment day being video recorded.	
9.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
10.	I agree to my anonymised data being used by research teams for future research.	
11.	I agree to take part in this study.	

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The following statements are optional:		
1.	I agree to complete a short interview about my child's/person I care for's ability.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of my child/person I care for that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome	
3.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	
4.	I agree to be contacted about future research projects to consider whether I would like to take part.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of Person receiving consent.	Date	Signature

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Behaviours that challenge in *SATB2*-associated syndrome

Direct Assessment Study

Chief Investigator: Dr Jane Waite

Consent Form E: For adults with *SATB2*-associated syndrome who are able to provide consent to participate in the study.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 4, 06.07.2022) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason and without my legal rights being affected.	
3.	I agree to my personal data and data relating to me collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study, may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I agree to my GP being informed of my participation in the study.	
6.	I agree to the feedback report arising from this study being shared with my GP.	
7.	I understand that if during the study I tell, or my parent/carer tells the research team something that causes them to have concerns in relation to my health and/or welfare they may need to breach my confidentiality.	
8.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
9.	I agree to study visits/research assessment day being video recorded.	
10.	I agree to my anonymised data being used by research teams for future research.	
11.	I agree to take part in this study.	

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The following statements are optional:		
1.	I agree to my parent/carer completing a short interview about my ability.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of my child/person I care for that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome.	
3.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	
4.	I agree to be contacted about future research projects to consider whether I would like to take part.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of Person receiving consent	Date	Signature

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Behaviours that challenge in *SATB2*-associated syndrome

Direct Assessment Study

Chief Investigator: Dr Jane Waite

Consent Form D: For parents/carers of individuals over the age of 16 and able to make an informed decision about participation in the study.

		Please initial boxes
1.	I confirm that I have read and understand the Participant Information Sheet (Version 4, 06.07.2022) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason and without my legal rights being affected.	
3.	I agree to my personal data and data relating to me collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study, may be looked at by individuals from Aston University, or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I agree to the person with *SATB2*-associated syndrome's GP being informed of my participation in the study.	
6.	I agree to the feedback report arising from this study being shared with the person with *SATB2*-associated syndrome's GP.	
7.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare or that of the individual with *SATB2*-associated syndrome they may need to breach my confidentiality.	
8.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
9.	I agree to study visits/research assessment day being video recorded.	
10.	I agree to my anonymised data being used by research teams for future research.	
11.	I agree to take part in this study.	

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The following statements are optional:		
1.	I agree to completing a short telephone interview about the individual with *SATB2*-associated syndrome's ability.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of my child/person I care for that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome.	
3.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	
4.	I agree to be contacted about future research projects to consider whether I would like to take part.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of Person receiving consent	Date	Signature

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Behaviours that challenge in *SATB2*-associated syndrome

Direct Assessment Study

Chief Investigator: Dr Jane Waite

Consent Form C: For a personal/nominated consultee of a person with *SATB2*-associated syndrome who is over the age of 16 and not able to provide consent.

Before deciding whether to participate, please ensure you read the information on acting as a personal/nominated consultee in the attached document for the person you care for.

Please initial boxes

1.	I confirm that I have read and understand the Participant Information Sheet (Version 4, 06.07.2022) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason and without my legal rights being affected.	
3.	I agree to my personal data and data relating to me collected during the study being processed as described in the Participant Information Sheet.	
4.	I understand that relevant sections of my data collected during the study may be looked at by individuals from Aston University or from regulatory authorities, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.	
5.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare they may need to breach my confidentiality.	
6.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I am comfortable with this risk.	
7.	I agree to study visits/research assessment day being video recorded.	
8.	I agree to their/my anonymised data being used by research teams for future research.	
9.	I agree to take part in this study.	

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The following statements are optional:		
1.	I agree to complete a short interview about the ability of the person for whom I am acting as a consultee.	
2.	I agree to the research team using my unique reference number, if I provided one, to extract data relating to me and that of for the person for whom I am acting as consultee that may have been collected as part of an ongoing longitudinal research study, led by the Cerebra Network for Neurodevelopmental Disorders, University of Birmingham, that is investigating the behavioural phenotype of SATB2-associated syndrome.	
3.	I agree to my personal data being processed for the purposes of inviting me to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	
4.	I agree to be contacted about future research projects to consider whether I would like to take part.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of consultee	Date	Signature
_____	_____	_____
Name of Person receiving consent.	Date	Signature

Aston University
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Behaviours that challenge in *SATB2*-associated syndrome

Direct Assessment Study

Chief Investigator: Dr Jane Waite

Declaration Form A: For a personal/nominated consultee of a person with *SATB2*-associated syndrome who is over the age of 16 and not able to provide consent.

Before deciding whether to participate, please ensure you read the information on acting as a personal/nominated consultee in the attached document for the person you care for.

Please initial boxes

1.	I have been consulted about (name of participant) _____'s participation in the above research project. I confirm that I have read and understand the Participant Information Sheet (Version 4, 06.07.2022) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2.	I confirm that in my opinion he/she would have no objection to participating in the study.	
3.	I understand that the participation of the person for whom I am acting as a consultee is voluntary and that I am free to withdraw them at any time, without giving any reason and without their legal rights being affected.	
4.	I agree to the person for who I am acting as consultee's personal data and data relating to him/her collected during the study being processed as described in the Participant Information Sheet.	
5.	I understand that relevant sections of his/her data collected during the study may be looked at by responsible individuals from Aston University or from regulatory authorities, where it is relevant to their taking part in this research. In my opinion he/she would have no objection to this.	
6.	I agree to his/her GP being informed of their/my participation in the study.	
7.	I agree to the feedback report arising from this study being shared with his/her GP.	
8.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to the health and/or welfare of the person for whom I am acting as consultee, they may need to breach confidentiality.	
9.	I understand that data will be temporarily stored on highly secure servers at Aston University; however, as with all online activity there is a small risk of unauthorised access to my data (hackers). I confirm that in my opinion he/she would have no objection to this risk.	
10.	I confirm that in my opinion he/she would have no objection to study visits/research assessment day being video recorded.	
11.	I confirm that in my opinion he/she would have no objection to their anonymised data being used by research teams for future research.	

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The following statements are optional:		
1.	I confirm that in my opinion he/she would have no objection to me completing a short interview about his/her ability.	
2.	I agree to be contacted about future research projects to consider whether the person for whom I am acting as consultee would like to take part.	
3.	I agree to my personal data being processed for the purposes of inviting me and the person for whom I am acting as consultee to participate in future research projects. I understand that I may opt out of receiving these invitations at any time.	

_____	_____	_____
Name of participant	Date	Signature
_____	_____	_____
Name of consultee	Date	Signature
_____	_____	_____
Name of Person receiving consent.	Date	Signature

Appendix 36: Chapters Four and Five capacity protocol

Aston University

How do I know if my child or person I care for is able to provide their own consent to take part in this project?

The decision about whether to take part in this study must be 'informed'. This means that anyone making the decision must understand exactly what is involved in the study, what will be required from participants and why. You need to decide whether your child/the person you care for can understand enough about the study to make an 'informed' decision independently about whether or not they would like to participate and to communicate this decision to you. If you are not sure whether or not your child/person you care for is able to make this decision, then the following procedure may be helpful to you.

STEP ONE: Read the following statements to the person you care for:

1. The study is about behaviours that challenge in people with SATB2-associated syndrome.
2. The study will involve your parents/your carers answering questions about some of your behaviours and characteristics.
3. Your parents/carers will also be asked to answer some questions about your ability.
4. Your parent/carer will answer these questions over the phone, or by video call.
5. If you agree for your parents/your carers to be contacted again by the research team, you may be invited to take part in other research.
6. You do not have to agree to take part. If you agree to take part and change your mind at a later time that is OK. You just need to tell someone.
7. All information will be stored in a safe place and only the research team at Aston University will have access to your information.

STEP TWO: Ask the person you care for the following questions:

1. What is the study about?
2. What will you and your parents or carers need to do to take part in the study?
3. What will happen if you agree for me/your parents/your carers to be contacted again by the research team?
4. What do you need to do if you decide to take part and then change your mind?
5. Where will your information be kept?
6. Do you agree to take part in this study?

If your child/person you care for is able to answer each of the above questions correctly and without any support then it is likely that they have sufficient capacity to consent to take part in the research project. If this is the case, your child/person you care for can complete the relevant consent form online. If the person you care for has been unable to answer all of the questions above it is unlikely that they have sufficient capacity to consent to take part in the research project. If this is the case, a parent/carer will need to act as their nominated/personal consultee and complete Consent Form C.

Appendix 37: Chapter Five pre-visit risk assessment

Pre-Visit Risk Assessment

The following checklist should be used when arranging direct face to face research visits or assessments with participants. Prior to the research visit it is essential that the measure of adaptive functioning identified in the protocol (i.e., The Vineland Adaptive Behaviour Scales) has been administered to ensure that the researcher has an overview of the participant's developmental level. The following checklist should then be administered with the family over the telephone at the time of booking the research visit.

PART ONE

To be completed by the principal researcher conducting the assessment day:

DATE AND TIME OF THE VISIT:

VISIT LOCATION:

Q1) I have completed the Vineland Adaptive Behaviour Scale (VABS)
Y/N

Q2) I have shared these details with all researchers conducting the direct assessments
Y/N

Q3) I have gone through the task, room and device checks with the parent/caregiver
Y/N

Q4) I have worked out which Executive Function tasks to conduct according to the child's overall age equivalent on the VABS
Y/N

Q5) I have worked out where to start on the BPVS based on the child's receptive language age equivalent on the VABS
Y/N

Q6) I have shared the visit details with another researcher in the team who will contact me if I do not call within two hours of the end of the visit. That researcher will attempt to contact me, followed by the participant's family, my next of kin, and the police if necessary

Y/N

The name of the researcher who will contact me is: _____

Their contact numbers are: mob:_____other:_____

My contact numbers are: mob:_____other:_ _____

My next of kin is: _____ No:_____

The ID number of the participant is:_____ mob:_____

THIS FORM SHOULD BE PHOTOCOPIED AND SHARED WITH ALL RESEARCHERS AND CONTACT PERSON PRIOR TO THE VISIT

Appendix Thirty-Seven
PART TWO

To be completed with parent/carer/guardian:

Covid-19 specific questions for face-to-face visits.

Q1) Have you or anyone you have been in contact with tested positive for Covid-19 within the last week?

Q2) Have you or anyone you have been in personal contact with experienced any Covid-19 symptoms within the last week?

This includes: has confirmed or suspected coronavirus?

Is self-isolating?

Has a high temperature and/or a new, continuous cough?

Has experienced a sudden loss of smell and/or taste?

IF THE PERSON SAYS 'YES' TO ANY COVID-19 SYMPTOMS, DO NOT INVITE TO ATTEND A RESEARCH VISIT AT THIS TIME. CONSULT PUBLIC HEALTH ENGLAND TO CALCULATE THE TIME PERIOD THAT WOULD NEED TO PASS PRIOR TO RE-CONTACTING THAT FAMILY AND RE-ASSESSING THE SITUATION.

Q3) Are you or any member of your family or anyone that you are personally in contact with currently shielding?

Q4) Do you or any member of your family have a health condition that increases your/their risk of Covid-19? E.g., respiratory condition

Q5) Do you have any preference for Covid-19 risk prevention measures for the assessment day (e.g., wearing of masks). Notify parent/caregiver that all equipment is sanitised between uses and that the research team will take lateral flow tests prior to the assessment day.

Challenging behaviour.

Q6) Does your child/person you care for show any challenging behaviour? (e.g. self-injurious behaviour, aggressive behaviour, destruction of property)

Q6a) If yes, when is your child/person you care for most likely to show challenging behaviour?

Q6b) Are there any specific causes of behaviour that we need to be aware of because they may place the researcher or your child/person you care for at risk during the visit? (e.g. clothing, scents, phrases, actions).

Q6c) Note down functions of behaviour from parent/caregiver completion of the QABF.

Q7) How do you usually manage behaviour when it occurs? (ask parent what they do to stop behaviour e.g. distract child, change activity, use of restraint, protective clothing)

Q8) Do you have any household pets that we should be aware of (if visiting participants house only)?

Q9) Is there parking close to your house (if visiting participants house only)?

Q10) Is there anything we should be aware of to ensure the visit goes well, or that may place the researcher at risk?

Q11) Will you be available throughout the entire assessment day? (note: parent/guardian/carer needs to be available for the visit to take place).

Q12) To ensure we get off to a good start, what are your child/person you care for's main likes/dislikes?

Q13) Does your child/person you care for have any allergies?

Appendix 38: Chapter Five parent/caregiver laminated guide for remote testing session

Caregiver Guide removed due to copyright restrictions

Appendix 39: Chapter Five research manual and protocol for remote testing session

Researcher Manual and Protocol removed due to copyright restrictions

Appendix 40: Behavior Problems Inventory – Short Form

Behavior Problems Inventory – Short Form (BPI-S) removed due to copyright restrictions. Measure available at: https://bpi.haoliang.me/

Appendix 41: Behaviour Rating Inventory of Executive Function – Preschool Version

Behaviour Rating Inventory of Executive Function – Preschool Version (BRIEF-P) removed due to copyright restrictions

Appendix 42: Responses to Uncertainty and Low Environmental Structure Scale

AppendixQt_RULES_06.07.2022_V1
IRAS ID: 296378

Responses to Uncertainty and Low Environmental Structure (RULES)

For each of the following statements, please rate how well the statement describes your child by selecting among one of the five responses and circling the number corresponding to that response.

	1 Not at all	2	3 Somewhat	4	5 Very much
1. My child gets tense when unexpected events or transitions occur in his/her environment.	**0**	**1**	**2**	**3**	**4**
2. My child has a hard time coping with even minor changes.	**0**	**1**	**2**	**3**	**4**
3. My child says, "it is unfair" when he/she cannot know what will happen next.	**0**	**1**	**2**	**3**	**4**
4. My child always wants to know ahead of time what the plan is.	**0**	**1**	**2**	**3**	**4**
5. My child becomes upset if he/she has to enter a new situation.	**0**	**1**	**2**	**3**	**4**
6. My child seeks reassurance prior to entering an unfamiliar situation.	**0**	**1**	**2**	**3**	**4**
7. My child cries when he/she finds him/herself in an unfamiliar situation.	**0**	**1**	**2**	**3**	**4**
8. My child gets down on himself/herself if he/she doesn't know what will happen next.	**0**	**1**	**2**	**3**	**4**
9. My child performs best in highly structured environments.	**0**	**1**	**2**	**3**	**4**
10. My child tantrums when an unexpected event occurs.	**0**	**1**	**2**	**3**	**4**
11. My child avoids unstructured situations.	**0**	**1**	**2**	**3**	**4**

	1 Not at all	2	3 Somewhat	4	5 Very much
12. My child cannot relax if he/she does not know what will happen next.	**0**	**1**	**2**	**3**	**4**
13. My child cannot sleep if he/she anticipates an upcoming change.	**0**	**1**	**2**	**3**	**4**
14. My child becomes fidgety during transitions.	**0**	**1**	**2**	**3**	**4**
15. My child freezes up in the face of unexpected events.	**0**	**1**	**2**	**3**	**4**
16. Transitions are difficult for my child.	**0**	**1**	**2**	**3**	**4**
17. My child complains of physical symptoms (e.g., headaches, stomach aches) when he/she is about to enter a new situation.	**0**	**1**	**2**	**3**	**4**

Please check your answers and go on to the next questionnaire.

Appendix 43: Clinical Anxiety Scale for Persons with Intellectual Disability

AppendixQj_CLASP-ID_06.07.2022_V2
IRAS ID: 296378

Clinical Anxiety Scale for Persons with Intellectual Disability (ClASP-ID)

This questionnaire is going to ask you about behaviours you may or may not have seen in the person you care for, over the last MONTH.
For each item, you will be asked to rate how frequently the behaviours have occurred over the last month.
For some of the questions, we will also ask you to think about whether the behaviour has been occurring more or less than is usual for the person you care for, over the last month.
Please try to answer every question.
If you are unsure whether you have seen the behaviour, please select 'almost never' and move onto the next question.

<table>
<tr><td colspan="7">Has he/she…</td><td></td></tr>
<tr><td colspan="7">1. Seemed withdrawn or 'vacant'?</td><td>1b. Over the past one month, has this behaviour been…</td></tr>
<tr><td>Almost never</td><td>Less than a quarter of the time</td><td>Less than half the time</td><td>About half of the time</td><td>More than half the time</td><td>About three quarters of the time</td><td>All of the time</td><td>☐ Occurring a lot more than is typical of him/her</td></tr>
<tr><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐ Occurring a bit more than is typical of him/her</td></tr>
<tr><td colspan="7"></td><td>☐ Occurring at the same rate that is typical of him/her</td></tr>
<tr><td colspan="7"></td><td>☐ Occurring a bit less than is typical of him/her</td></tr>
<tr><td colspan="7"></td><td>☒ Occurring a lot less than is typical of him/her</td></tr>
<tr><td colspan="7">Does he/she…</td><td></td></tr>
<tr><td colspan="7">2. ever make negative or frustrated vocalisations? (e.g. whining, grumbling, growling, shouting, screaming)</td><td></td></tr>
<tr><td colspan="2">Almost never</td><td>Once a month</td><td>Less than once a week</td><td>Once or twice a week</td><td>At least 3-4 times per week</td><td>Every day</td><td>More than once a day</td></tr>
<tr><td colspan="2">☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td></tr>
<tr><td colspan="7">3. appear on edge OR on the look out for danger?</td><td></td></tr>
<tr><td colspan="2">Almost never</td><td>Once a month</td><td>Less than once a week</td><td>Once or twice a week</td><td>At least 3-4 times per week</td><td>Every day</td><td>More than once a day</td></tr>
<tr><td colspan="2">☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td></tr>
<tr><td colspan="7">4. ever seem protective of a particular part of his/her body? (e.g. holding it, guarding it, flinching)</td><td></td></tr>
<tr><td colspan="2">Almost never</td><td>Once a month</td><td>Less than once a week</td><td>Once or twice a week</td><td>At least 3-4 times per week</td><td>Every day</td><td>More than once a day</td></tr>
<tr><td colspan="2">☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td><td>☐</td></tr>
<tr><td colspan="7"></td><td></td></tr>
</table>

Does he/she…						
5. pace around the room?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
6. ever appear restless or agitated?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
7. ever run away or hide from certain objects or situations?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
8. ever cover himself/herself with a blanket or try to place a barrier between himself/herself and others or a situation?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
9. ever have watery eyes that is different from crying?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
10. ever freeze suddenly (stick to the spot) in response to specific situations?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
11. ever grind his/her teeth?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
12. avoid (or try to avoid) certain objects or places?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
13. ever take sharp intakes of breath or gasp?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐

14. ever look very worried or anxious?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
15. have an angry look on his/her face?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
16. startle easily, or easily alarmed?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
Over the past month, have you noticed…						
17. increased or different leg movements? (e.g. restlessness, tense, tremors, kicking, drawing legs up, jerking)?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
18. that he/she shakes or trembles?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
19. his/her face look tense?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
Over the past month, has he/she…						
20. been hitting, holding or touching a part of their body?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐
Over the past month, has his/her…						
21. movements ever become jerky?						
Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐

22. lips ever become tight, pout or quiver?

Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
☐	☐	☐	☐	☐	☐	☐

Has he/she…

23. lost interest in activities that he/she used to enjoy?

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

Does he/she…

24. lack energy?

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

25. get tired for no apparent reason?

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

Is he/she…

26. spending more time asleep than usual? (e.g. not waking in the morning, sleeping during the day)

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time`	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

26b. Over the past one month, has this behaviour been…

☐ Occurring a lot more than is typical of him/her

☐ Occurring a bit more than is typical of him/her

☐ Occurring at the same rate that is typical of him/her

☐ Occurring a bit less than is typical of him/her

☐ Occurring a lot less than is typical of him/her

27. quiet and spending time alone?							**27b. Over the past one month, has this behaviour been…**
Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time`	About three quarters of the time	All of the time	☐ Occurring a lot more than is typical of him/her
☐	☐	☐	☐	☐	☐	☐	☐ Occurring a bit more than is typical of him/her
							☐ Occurring at the same rate that is typical of him/her
							☐ Occurring a bit less than is typical of him/her
							☐ Occurring a lot less than is typical of him/her

How often are these statements true for your child/the person you support?

28. Preparing him/her before things happen helps to reduce his/her distress

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

29. Removing the person I care for from a situation, or removing an item/object, generally calms them down

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

30. When the person I care for is distressed, I am able to calm or comfort him/her

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

31. When in certain preferred environments (e.g. home, their bedroom) the person I care for generally appears calm and relaxed

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

32. We are unable to do 'typical' day to day activities because of the <u>emotional distress</u> that would cause him/her (e.g. holidays, visiting friends, going for meals, general days out).

Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

33. We are unable to do activities we used to do with the person I care for because of the emotional distress he/she would experience.						
Almost never	Less than a quarter of the time	Less than half the time	About half of the time	More than half the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐
34. Have you noticed any other changes in behaviour or mood not covered in this questionnaire? If yes, please give details						

Appendix 44: Social Communication Questionnaire

Social Communication Questionnaire (SCQ) removed due to copyright restrictions

Appendix 45: Chapter Five validity checklist

Validity checklist removed due to copyright restrictions

Appendix 46: Chapter Five variables associated with missingness

Appendices Table 11

Exploration of variables associated with missingness on task-based executive function assessments

		Missingness variable		
	Statistical test	Prohibition score	Working Memory variables [c]	Conflict and no conflict scales [d]
Chronological age [a]	Mann-Whitney U	69.500	109.000	80.000
Sex [a]	Fisher's Exact	- [e]	- [e]	- [e]
Speech [b]	Mann-Whitney U	45.000	69.000	79.500
Mobility [b]	Mann-Whitney U	27.500	59.000	102.500
Self-help Score [b]	Mann-Whitney U	51.500	75.000	78.500
Living arrangement [a]	Fisher's Exact	- [e]	- [e]	- [e]
Family income [a]	Fisher's Exact	- [e]	- [e]	- [e]
VABS overall DAE	Mann-Whitney U	2.000	65.000	58.000
VABS ABC	t-test	.979	2.207	.586
BRIEF-P Inhibition	t-test	1.721	-.809	-1.617
BRIEF-P Shift	t-test	.187	-.715	-.908
BRIEF-P Emotional Control	t-test	.134	-1.273	-.986
BRIEF-P Working Memory	t-test	1.947	-.087	-2.356
BRIEF-P Planning/ organisation	t-test	1.796	-.511	-2.023
BRIEF-P Global composite	t-test	1.408	-.751	-1.875
TAQ Impulsivity	Mann-Whitney U	32.500	95.000	143.000
TAQ Overactivity	t-test	1.414	-.079	-.471
RULES Total score	t-test	.147	.204	-.287
RBQ Stereotyped behaviour	Mann-Whitney U	19.000	67.500	98.500
RBQ Compulsive behaviour	Mann-Whitney U	42.500	62.000	92.500
RBQ restricted preferences	t-test	1.401	.631	.091
RBQ Insistence on sameness	Mann-Whitney U	36.500	56.500	120.000
RBQ repetitive language	Mann-Whitney U	27.000	59.000	118.000
SCQ RRSB	Mann-Whitney U	26.500	61.000	132.500
BPI-S AD Frequency	Mann-Whitney U	45.000	95.000	164.500
BPI-S AD Severity	Mann-Whitney U	55.000	86.000	156.000
ClASP-ID Anxiety	Mann-Whitney U	33.000	79.000	143.000

Note. Significant tests (p<.05) shaded in grey. VABS = Vineland Adaptive Behaviour Scales – 3rd Edition. DAE = Developmental age equivalent. ABC = Adaptive Behaviour Composite. BRIEF-P = Behaviour Rating Inventory of Executive Function – Preschool. TAQ = The Activity Questionnaire. RULES = Responses to Uncertainty and Low Environmental Structure. RBQ = Repetitive Behaviour Questionnaire. SCQ = Social Communication Questionnaire. RRSB = Restricted repetitive and stereotyped behaviour. BPIS-S = Behavior Problems Inventory. AD = Aggressive/destructive behaviour. CIASP-ID = Clinical Anxiety Screen for Persons with Intellectual Disability.

[a] Derived from background questionnaire.

[b] Derived from Wessex Questionnaire.

[c] Working Memory (WM) variables merged as the same results were found for WM Span, WM Efficiency and WM Perseveration.

[d] Conflict and no conflict scales merged as the same results were found for both scales.

[e] Fisher's Exact Test performed over Chi-square as there were less than five expected values in cells.

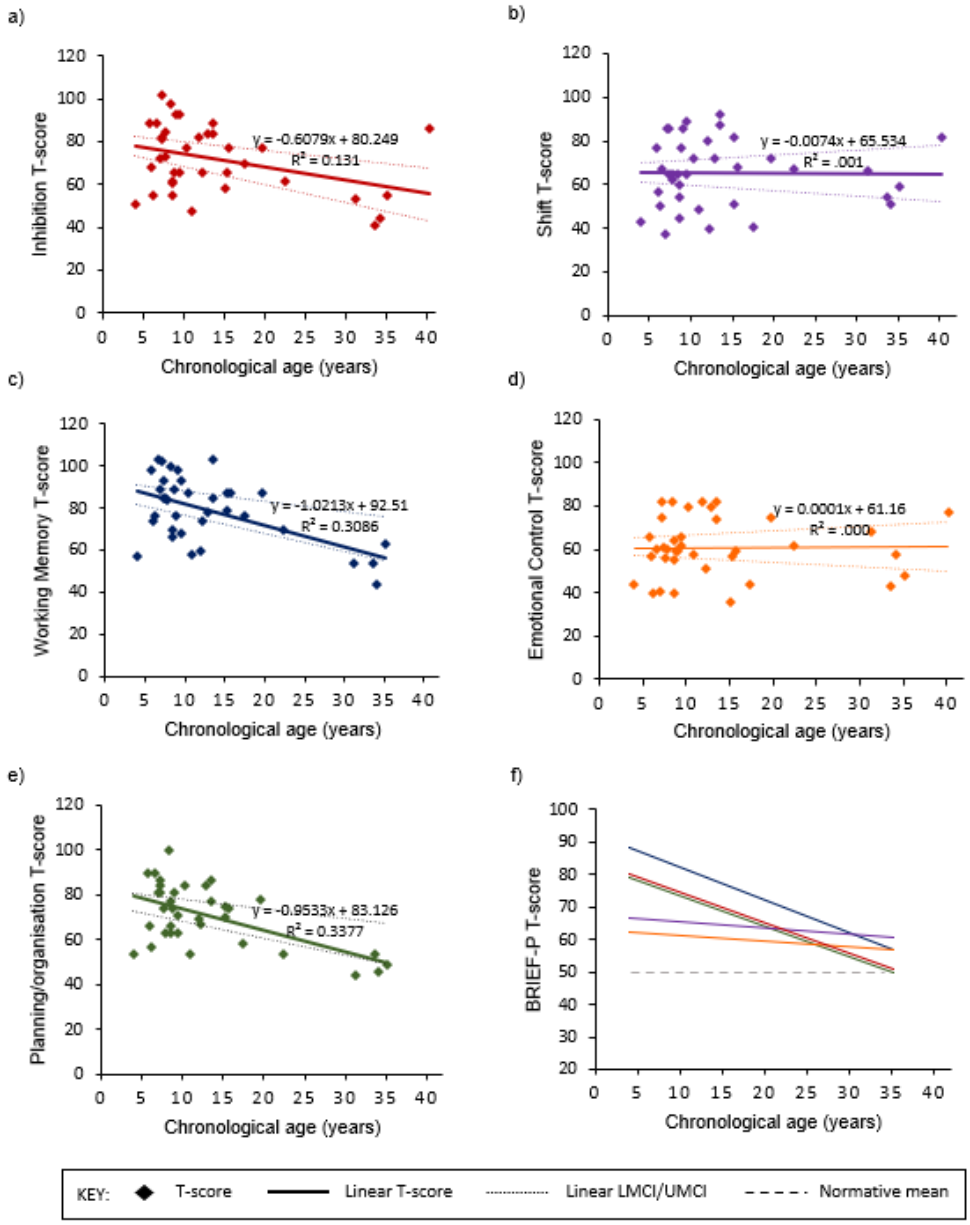
Appendix 47: Chapter Five additional developmental trajectory analyses

BRIEF-P T-score Developmental Trajectory Analysis Excluding the Outlier

Appendices Figure 103 depicts the developmental trajectories for the relationship between chronological age and T-scores on BRIEF-P subscales when the outlier is removed.

Appendices Figure 103

The relationship between chronological age and BRIEF-P T-scores for a) Inhibition, b) Shift, c) Working Memory, d) Emotional Control, and e) Planning/organisation when the outlier is removed; f) depicts the linear trajectories for the T-scores when the outlier is removed from all BRIEF-P subscales.



Note. The normative mean for BRIEF-P T-scores = 50. Greater T-scores denote greater levels of difficulty.

The straight line remained a reliable fit when the outlier was removed for *Working Memory* R^2 = .309, F(1,34)=15.181, p <.001, intercept = 92.511, gradient = -1.02; 95% CI: 80.29 to 100.73) and *Planning/organisation* (R^2 = .338, F(1,34)=17.337, p <.001, intercept = 83.13, gradient = -.95; 95% CI: 75.95 to 90.31). No outliers were identified for remaining BRIEF-P subscales, where a straight line was a reliable fit for Inhibition (R^2 = .131, F(1,35) = 5.29, p = .028, intercept (constant) = 80.25, gradient = -.61; 95% CI: 71.34 to 89.16) and a non-reliable fit for *Shift* (R^2 = .000, F(1,35) = .001, p = .979, intercept (constant) = 65.53, gradient = -.01; 95% CI: 56.26 to 74.81) and *Emotional Control* (R^2 = .000, F(1,35) = .000, p = .999, intercept (constant) = 61.16, gradient = .00; 95% CI: 52.97 to 69.35). As reported in Section 5.4.1.3, the rotation method revealed no systematic relationship for *Shift* and *Emotional Control.*

A repeated measures ANOVA revealed significant overall differences in the intercepts (means) of BRIEF-P subscale T-scores (F(1,35)= 8.042, p =.008, η^2 = .187). Descriptives are presented in Appendices Table 12. Post hoc analysis with a Bonferroni adjustment revealed *Working Memory* T-scores were significantly greater than T-scores for *Inhibition, Shift, Planning/Organisation,* and *Emotional Control*; and *Inhibition* and *Planning/Organisation* T-scores were significantly greater than T-scores for *Emotional Control* (see Appendices Table 13).

Appendices Table 12
Mean, standard deviation, and range of BRIEF-P subscale T-scores without the outlier

	Mean (*SD*)	Range
Inhibition	71.50 (*15.86*)	41.00-102.00
Working Memory	79.22 (*15.46*)	44.00-103.00
Planning/organisation	70.72 (*13.80*)	44.00-100.00
Shift	64.97 (*15.30*)	37.00-92.00
Emotional control	60.72 (*13.48*)	36.00-82.00

Note. Normative mean for T-scores is 50. Greater T-scores denote greater levels of difficulty; T-scores ≥65 indicate clinically significant ratings.

Appendices Table 13

Post hoc comparisons exploring differences in mean of BRIEF-P subscale T-scores without the outlier

Comparison			Mean difference	SE	95% CI	*p* value
Subscale		Subscale				
WM	-	PO	8.50	1.36	4.44-12.56	<.001
	-	IHN	7.72	1.53	3.15-12.30	<.001
	-	EC	18.50	2.84	10.00-27.00	<.001
	-	SHI	14.25	2.61	6.45-22.06	<.001
INH	-	PO	.78	1.51	-.376-5.31	1.00
	-	EC	10.78	2.15	4.35-17.21	<.001
	-	SHI	6.53	2.14	0.11-12.94	.044
PO	-	EC	10.00	2.20	3.42-16.58	<.001
	-	SHI	5.75	2.48	-1.67-13.17	.262
SHI	-	EC	4.25	1.87	-1.36-9.86	.268

Note. INH = Inhibition, WM = Working memory, PO = Planning/organisation, SHI =Shift, EC = Emotional control.

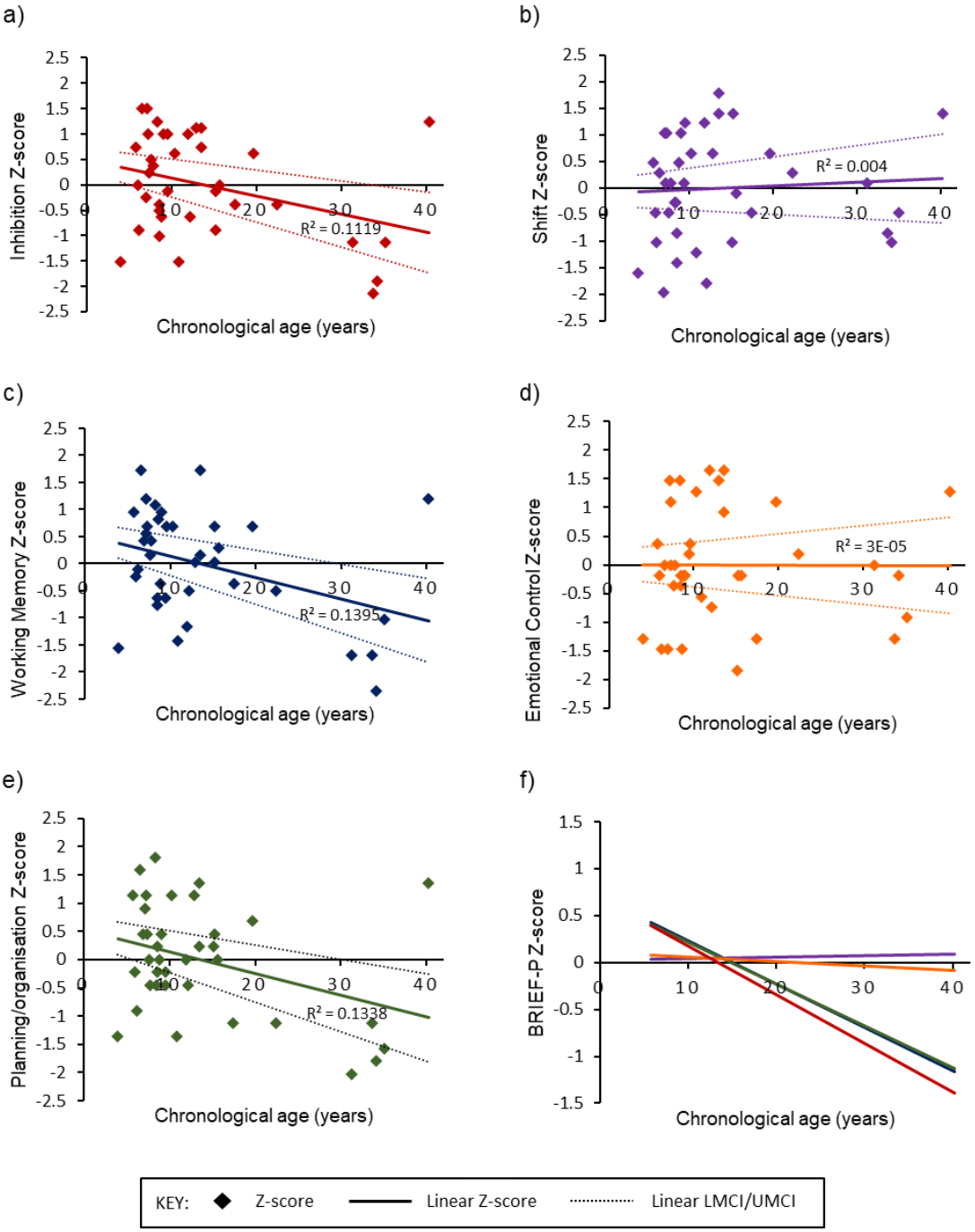
A repeated measures ANCOVA revealed a significant overall effect of chronological age on BRIEF-P subscale T-scores, indicating T-scores significantly improve with chronological age ($F(1,34)= 733.13$, $p <.001$, $\eta^2 = .956$). There was a non-significant overall interaction between chronological age and BRIEF-P subscale T-scores ($F(1,34)= 2.63$, $p = .114$, $\eta^2 = .072$). However, using a series of repeated measures ANCOVA's with two levels to explore differences in rates of improvement, interactions between age and T-scores revealed significantly slower rates of improvement in *Shift* compared to *Inhibition* ($F(1,34) = 10.75$, $p = .002$, $\eta^2 = .240$)*, Working Memory* ($F(1,34) = 8.47$, $p = .006$, $\eta^2 = .199$)*,* and *Planning/Organisation* ($F(1,34)= 7.76$, $p = .009$, $\eta^2 = .186$); and significantly slower rates of improvement in *Emotional Control* compared to *Inhibition* ($F(1,34)= 11.38$, $p = .002$, $\eta^2 = .251$)*, Working Memory* ($F(1,34)= 7.23$, $p = .011$, $\eta^2 = .175$)*,* and *Planning/Organisation* ($F(1,34)= 11.19$, $p = .002$, $\eta^2 = .248$)*.* There were no significant differences in rates of improvement between *Shift* and *Emotional Control* ($F(1,34)= .01$, $p = .938$, $\eta^2 = .000$), with a null trajectories indicated for both subscales. There were no significant differences in rates of improvement in *Inhibition* compared to *Working Memory* ($F(1,34)= .19$, $p = .666$, $\eta^2 = .006$) and *Planning/organisation* ($F(1,34)= .01$, $p = .944$, $\eta^2 = .000$), or in *Working Memory* compared to *Planning/organisation* ($F(1,34)= .17$, $p = .683$, $\eta^2 = .005$), suggesting similar trajectories of improvement with age between *Inhibition, Working Memory,* and *Planning/organisation*. This pattern of findings is consistent with those including the outlier (see Section 5.4.1.3).

BRIEF-P Z-score Developmental Trajectory Analysis Against Chronological Age

Appendices Figure 104 depicts the developmental trajectories for the relationship between chronological age and BRIEF-P subscale Z-scores.

Appendices Figure 104

The relationship between chronological age and BRIEF-P Z-scores for a) Inhibition, b) Shift, c) Working Memory, d) Emotional Control, and e) Planning/organisation; f) depicts the linear trajectories for the Z-scores for all BRIEF-P subscales.



Linearity was indicated for *Inhibition* (R^2 = .112, *F*(1,35) = 4.411, *p* = .043, intercept (constant) = .49, gradient = -.04, 95% CI: -.070 to -.001); *Working Memory* (R^2 = .140, *F*(1,35) = 5.675, *p* =.023, intercept (constant) = .55, gradient = -.040, 95% CI: -.073 to -.006); and *Planning/Organisation* (R^2 = .134, *F*(1,35) = 5.406, *p* =.026, intercept (constant) = .53, gradient = -.039, 95% CI: -.073 to -.005). Null trajectories were indicated for *Shift* (R^2 = .004, *F*(1,35) .141, *p* =.710, intercept (constant) = -.092, gradient = .007, 95% CI: -.030 to .043) and *Emotional Control* (R^2 = .000, *F*(1,35) = .001, *p* =.975, intercept (constant) = .008, gradient = -.001, 95% CI: -.037 to .036). The rotation method (Thomas et al., 2009; 2010) was applied to examine the observed null trajectories for *Shift* and *Emotional Control*. Minimal changes were observed in the R^2 values for these trajectories, indicating no systematic relationship between the Z-scores and chronological age.

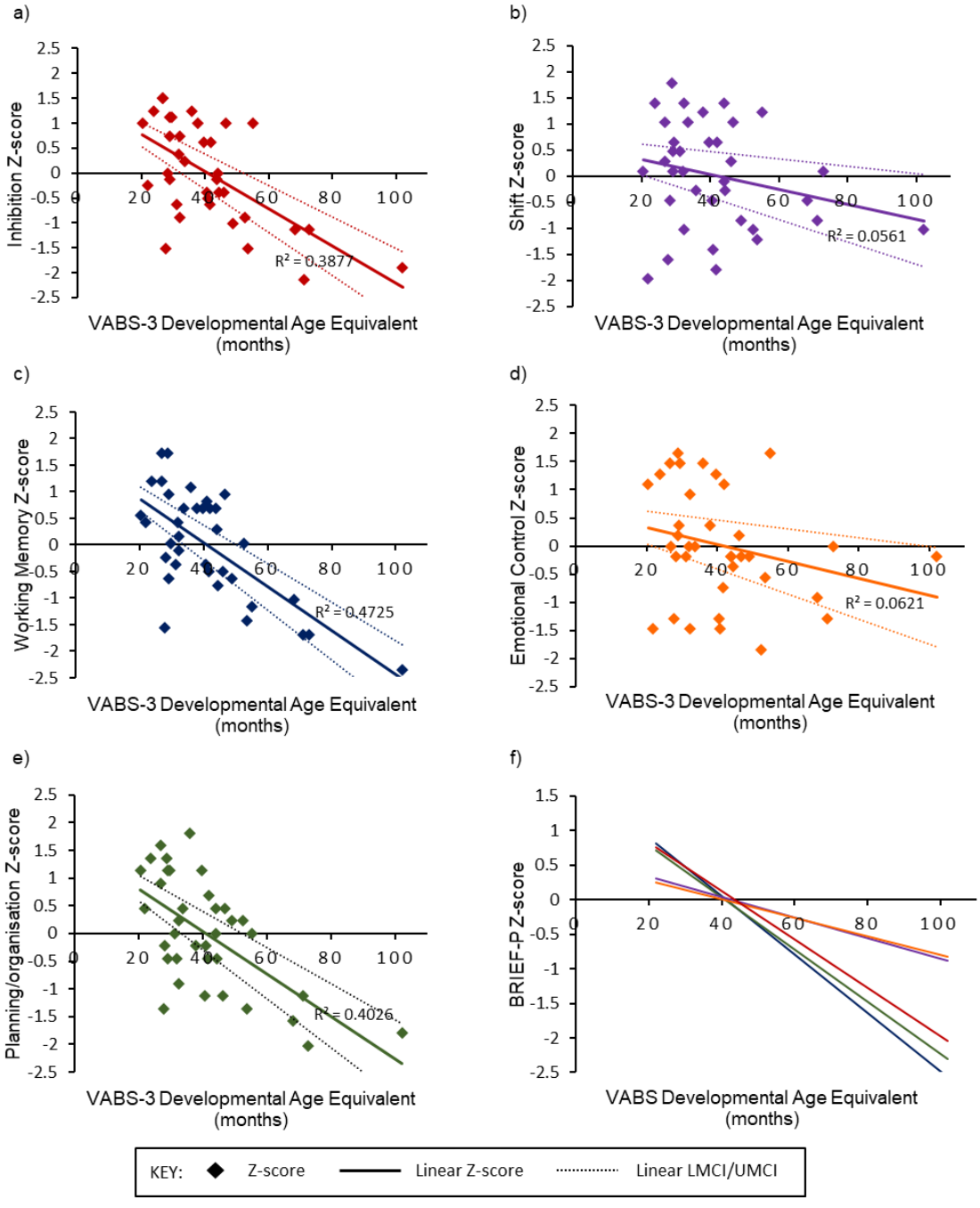
A repeated measures ANCOVA revealed a non-significant overall effect of chronological age on BRIEF-P subscale Z-scores, indicating that Z-scores did not significantly improve with chronological age (F(1,35) = 2.087, p = .157, η^2 = .056). Overall, a non-significant interaction between chronological age and BRIEF-P subscale Z-scores *F*(1,35) = 2.999, *p* = .104, η^2 = .074) was also observed, suggesting no overall differences in trajectory gradients. Trajectory gradients were further explored using a series of repeated measures ANCOVAS with two levels. Interactions between chronological age and Z-scores suggest significantly slower rates of improvement in *Shifting* compared in *Inhibition* (F(1,35) = 12.444, p = .001, η^2 = .262), *Working Memory* (F(1,35) = 8.215, p = .007, η^2 = .190), and *Planning/Organisation* (F(1,35) = 7.725, p = .009, η^2 = .181); and significantly slower rates of improvement in *Emotional Control* compared to *Inhibition* (F(1,35) = 9.322, p = .004, η^2 = .210), *Working Memory* (F(1,35) = 4.433, p = .043, η^2 = .112), and *Planning/Organisation* (F(1,35) = 6.712, p = .014, η^2 = .161). There were no significant differences in trajectory gradients between *Shift* and *Emotional Control* (F(1,35) = .343, p = .562, η^2 = .010), with no systematic relationship with chronological age indicated for both subscales. No significant differences were observed in trajectory gradients for *Inhibition* compared to *Working Memory* (F(1,35) = .145, p = .706, η^2 = .004) and *Planning/Organisation* (F(1,35) = .097, p = .757, η^2 = .003), or for *Working Memory* compared to *Planning/Organisation* (F(1,35) = .007, p = .934, η^2 = .000), suggesting similar rates of improvement between these EFs. This pattern of findings is consistent with those using BRIEF-P T-scores (see Volume I, Section 5.4.1.3).

BRIEF-P Z-score Developmental Trajectory Analysis Against Developmental Age

Appendices Figure 105 depicts the developmental trajectories for the relationship between VABS-3 overall developmental age equivalents and BRIEF-P subscale Z-scores.

Appendices Figure 105

The relationship between overall developmental age and BRIEF-P Z-scores for a) Inhibition, b) Shift, c) Working Memory, d) Emotional Control, and e) Planning/organisation; f) depicts the linear trajectories for the Z-scores for all BRIEF-P subscales.



Linearity was indicated for *Inhibition* (R^2 = .388, $F(1,34)$ = 21.535, p < .001, intercept (constant) = 1.53, gradient = -.038, 95% CI: -.054 to -.021); *Working Memory* (R^2 = .472, $F(1,34)$ = 30.454, p < .001, intercept (constant) = 1.71, gradient = -.042, 95% CI: -.057 to -.026); and *Planning/Organisation* (R^2 = .403, $F(1,34)$ = 22.910, p < .001, intercept (constant) = 1.58, gradient = -.038, 95% CI: -.055 to -.022). Null trajectories were indicated for *Shift* (R^2 = .056, $F(1,34)$ = 2.022, p = .164, intercept (constant) = .60, gradient = -.014, 95% CI: -.035 to .006) and *Emotional Control* (R^2 = .062, $F(1,34)$ = 2.253, p = .143, intercept (constant) = .63, gradient = -.015, 95% CI: -.035 to .005). The rotation method (Thomas et al., 2009; 2010) was applied to examine the observed null trajectories for *Shift* and *Emotional Control*. Minimal changes were observed in the R^2 values for these trajectories, indicating no systematic relationship between the Z-scores and developmental age.

A repeated measures ANCOVA revealed a significant overall effect of developmental age on BRIEF-P subscale Z-scores, indicating that Z-scores significantly improve with developmental age ($F(1,34)$ = 16.091, p < .001, η^2 = .321). However, there was a significant interaction between developmental age and BRIEF-P subscale Z-scores ($F(1,34)$ = 7.970, p = .008, η^2 = .190), suggesting differences in trajectory gradients (see Appendices Figure 105). These differences were explored using a series of repeated measures ANCOVAs with two levels. Interactions between developmental age and Z-scores suggest significantly slower rates of improvement in *Shift* compared in *Inhibition* ($F(1,34)$ = 11.708, p = .002, η^2 = .256), *Working Memory* ($F(1,34)$ = .8.760, p = .006, η^2 = .205), and *Planning/Organisation* ($F(1,34)$ = 6.323, p = .017, η^2 = .157); and significantly slower rates of improvement in *Emotional Control* compared to *Inhibition* ($F(1,34)$ = 13.202, p < .001, η^2 = .280), *Working Memory* ($F(1,34)$ = .6.490, p = .016, η^2 = .160), and *Planning/Organisation* ($F(1,34)$ = 7.722, p = .009, η^2 = .185). There were no significant differences in trajectory gradients between *Shift* and *Emotional Control* ($F(1,34)$ = .011, p = .916, η^2 = .000), with no systematic relationship with chronological age indicated for both subscales. No significant differences were observed in trajectory gradients for *Inhibition* compared to *Working Memory* ($F(1,34)$ = .419, p = .522, η^2 = .012) and *Planning/Organisation* ($F(1,34)$ = .019, p = 891, η^2 = .001), or for *Working Memory* compared to *Planning/Organisation* ($F(1,34)$ = .318, p = .577, η^2 = .009), suggesting similar rates of improvement between these EFs. This pattern of findings is consistent with those exploring the interaction between chronological age and BRIEF-P subscale T-scores (see Volume I, Section 5.4.1.3).

Appendix 48: Chapter Five correlations conducted on the original dataset

Appendices Table 14

Pairwise Spearman Correlations Between Questionnaire and Performance-based Tasks of Executive Function Based on the Original, Non-imputed Dataset

Task	Questionnaire measure					
	BRIEF-P Inhibition	BRIEF-P Working memory	BRIEF-P Shift	BRIEF-P GEC	TAQ Impulsivity	TAQ Overactivity
Prohibition score	**-.428***	-.330	-.276	-.421	-.285	-.177
Working memory efficiency	-.225	**-.465****	.026	-.259	-.278	-.261
Working memory perseveration	.032	.280	-.053	.087	.130	.045
Conflict score	-.415	-.400	-.347	-.416	-.417	-.383
No conflict score	-.293	-.274	-.239	-.282	-.223	-.275

Note. GEC = Global Executive Composite.
*= Deemed to approach significance (p=.011-.014). ***p*<.01.

Appendices Table 15

Correlations Between Caregiver-reported Behaviours that Challenge and Direct Assessment Measures using Direct EF Assessment Scores from the Original Non-imputed Dataset

	Original data [a, b]	
	BPI-S AD Frequency	BPI-S AD Severity
BPVS raw score	-.099	.077
Prohibition Score	-.257	-.114
Working memory efficiency	-.164	.016
Working memory perseveration	.063	.003
Conflict score	-.367	-.177
No conflict score	-.308	-.029
Anx-DOS spider fear composite	-.406	-.361
Anx-DOS global spider score	-.365	-.303
Anx-DOS jar fear composite	-.065	-.009
Anx-DOS global jar score	-.047	-.045
Anx-DOS separation distress score	-.133	-.172
Anx-DOS separation proximity seeking score	.108	.046
Anx-DOS global separation distress score	.128	.064

[a] *n*=23-34 across correlations.
[b] Spearman correlations were conducted due to non-normal distribution of data.

Appendix 49: Chapter Five intercorrelations between all questionnaire measure variables

Appendices Table 16

Inter-correlations Between Questionnaire Measure Variables (n=37)

	Chronological age	VABS DAE	BRIEF-P inhibition	BRIEF-P Shift	BRIEF-P Emotional control	BRIEF-P Working Memory	BRIEF-P Planning/ organisation	BRIEF-P Global composite	TAQ Impulsivity	TAQ Overactivity	RULES Total score	RBQ Stereotyped behaviour	RBQ Compulsive behaviour	RBQ restricted preferences	RBQ Insistence on sameness	RBQ repetitive language	SCQ Restricted, repetitive and stereotyped behaviour
VABS DAE	.594***	-															
BRIEF-P Inhibition	-.224	-.565***	-														
BRIEF-P Shift	.178	-.204	.713***	-													
BRIEF-P Emotional Control	.063	-.297	.768***	.790***	-												
BRIEF-P Working Memory	-.273	-.558***	.794***	.520***	.431*	-											
BRIEF-P Planning/ organisation	-.252	-.547***	.817***	.541***	.586***	.833***	-										
BRIEF-P Global composite	-.174	-.557***	.963***	.778***	.809***	.837***	.881***	-									
TAQ Impulsivity	-.124	-.508**	.859***	.636***	.756***	.639***	.704***	.855***	-								
TAQ Overactivity	-.299	-.613***	*.751***	*.452**	*.476***	*.748***	*.760***	*.754***	.728***	-							
RULES Total score	.296	.097	*.523***	*.777***	*.598***	*.350*	*.290*	*.584***	.401`	*.243*	-						
RBQ Stereotyped behaviour	-.153	-.449*	.570***	.661***	.511**	.540***	.561***	.650***	.492**	.602***	.474**	-					

Appendices Table 16 Continued

	Chronological age	VABS DAE	BRIEF-P inhibition	BRIEF-P Shift	BRIEF-P Emotional control	BRIEF-P Working Memory	BRIEF-P Planning/ organisation	BRIEF-P Global composite	TAQ Impulsivity	TAQ Overactivity	RULES Total score	RBQ Stereotyped behaviour	RBQ Compulsive behaviour	RBQ restricted preferences	RBQ Insistence on sameness	RBQ repetitive language	SCQ Restricted, repetitive and stereotyped behaviour
RBQ Compulsive behaviour	-.010	-.179	.366	.529***	.411`	.141	.180	.377	.446*	.373	.390	.552***	-				
RBQ restricted preferences	.109	.014	*.425**	*.405`*	*.361*	*.278*	*.273*	*.408`*	.435*	*.348*	*.394*	.377	.511**	-			
RBQ Insistence on sameness	.159	.021	.437*	.725***	.560***	.207	.222	.467**	.432*	.273	.788***	.571***	.684***	.542***	-		
RBQ repetitive language	.349	.320	.217	.319	.159	.317	.551	.197	.148	-.030	.548***	.074	.083	.559***	.404`	-	
SCQ RRSB	-.133	-.185	.623***	.623***	.510***	.525***	.622***	.661***	.536***	.528***	.557***	.683***	.475**	.554***	.557***	.211	-
ClASP-ID Anxiety	.126	-.199	.642***	.844***	.727***	.454**	.520***	.683***	.618***	.434*	.696***	.528***	.467**	.314	.673***	.166	.661***

Note. RRSB = Restricted, Repetitive and Stereotyped Behaviour. Spearman correlations were conducted due to non-normal distribution of data, except italicised correlation coefficients, where Pearson correlations were conducted due to normal distribution of data.
* p<.01, **p<.005, ***p<.001. ` = Deemed to approach significance (p=.011-.014).

Appendix 50: Chapter Five intercorrelations between all remote assessment variables

Appendices Table 17

Inter-correlations Between Assessments Completed with Individuals with SAS Based on the Original Non-imputed Data

	BPVS raw score	Prohibition score	Working memory efficiency	Working memory perseveration	Conflict score	No conflict score	Anx-DOS spider fear composite	Anx-DOS spider global score	Anx-DOS jar fear composite	Anx-DOS jar global score	Anx-DOS separation distress score	Anx-DOS separation proximity seeking score	Anx-DOS global separation
Prohibition Score	.398	-											
Working memory efficiency	**.582****	**.497***	-										
Working memory perseveration	-.371	**-.476***	**-.791*****	-									
Conflict score	**.587****	.351	**.609****	-.392	-								
No conflict score	**.600****	.461	**.643*****	-.346	**.847*****	-							
Anx-DOS spider fear composite	.375	.407	**.498***	-.330	.142	.299	-						
Anx-DOS global spider score	.376	.392	**.551****	-.409	.183	.332	**.983*****	-					
Anx-DOS jar fear composite	.110	.122	.089	-.007	.252	.434	.120	.096	-				
Anx-DOS global jar score	.108	.134	.136	-.118	.315	.455	.125	.103	**.963*****	-			
Anx-DOS separation distress score	.268	.043	-.208	.328	-.102	.030	.088	.014	.088	.124	-		
Anx-DOS separation proximity seeking score	-.316	-.133	**-.487`**	**.524***	**-.548***	-.393	-.161	-.226	-.139	-.133	.185	-	
Anx-DOS global separation distress score	-.199	.109	-.162	.272	-.431	-.285	.079	.030	-.014	.035	.336	**.880*****	-

Note. Spearman correlations were conducted due to non-normal distribution of data. Due to missing data in the original dataset, pairwise comparisons were conducted. The sample size ranges from 26-34 across correlations.
* *p*<.01, ***p*<.005, ****p*<.001. `= Deemed to approach significance (p=.011-.014).

Appendices Table 18

Inter-correlations Between Assessments Completed with Individuals with SAS Based on the Imputed Data

	BPVS raw score	Prohibition score	Working memory efficiency	Working memory perseveration	Conflict score	No conflict score	Anx-DOS spider fear composite	Anx-DOS spider global score	Anx-DOS jar fear composite	Anx-DOS jar global score	Anx-DOS separation distress score	Anx-DOS separation proximity seeking score	Anx-DOS global separation
Prohibition Score	.347	-											
Working memory efficiency	.394	**.469***	-										
Working memory perseveration	-.163	-.437	**-.736*****	-									
Conflict score	.493	.278	.364	-.234	-								
No conflict score	.517	.395	.417	-.262	**.665****	-							
Anx-DOS spider fear composite	.375	.397	**.520****	-.365	.121	.275	-						
Anx-DOS global spider score	.376	.383	**.557*****	-.416	.142	.291	**.983*****	-					
Anx-DOS jar fear composite	.110	.129	.141	-.108	.207	.343	.120	.096	-				
Anx-DOS global jar score	.108	.139	.179	-.195	.249	.355	.125	.103	**.963*****	-			
Anx-DOS separation distress score	.268	.049	-.284	.359	-.100	.012	.088	.014	.088	.124	-		
Anx-DOS separation proximity seeking score	-.316	-.162	**-.507***	**.535****	-.429	-.317	-.161	-.226	-.139	-.133	.185	-	
Anx-DOS global separation distress score	-.199	.069	-.155	.245	-.307	-.186	.079	.030	-.014	.035	.336	**.880*****	-

Note. Spearman correlations were conducted due to non-normal distribution of data. Pairwise comparisons conducted; n=34 for correlations including variables pertaining to the Anx-DOS and FLACC, n=35 for all other correlations.
* $p<.01$, ** $p<.005$, *** $p<.001$.