An Open Research approach to investigating how eye-tracking technology has been used as a tool to evaluate social cognition in intellectual disability

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BACKGROUND

THE SYSTEMATIC REVIEW

Social functioning is central to the conceptualisation of intellectual disability[1]. Yet, social-cognitive abilities (e.g., emotion discrimination, mental state reasoning) are not well understood. Floor effects are common when using traditional task batteries[2,3], as they are often demanding on language and domain-general cognition. **Reduced accessibility of measures limits 1) participation of individuals with mild-profound intellectual disability (inclusivity) and 2) the ability to capture meaningful differences in social-cognitive abilities (sensitivity).** Eye-tracking technology can be used to address these limitations, by measuring social-cognitive processes in a passive-viewing manner, without the need for explicit responses or verbal demands. **The systematic review (SR) aimed to characterise and evaluate the use of eye-tracking as a tool to measure social cognition among individuals with an intellectual disability.**

PRE-REGISTRATION

Quality of SRs relies on **comprehensive**, **systematic**, and **transparent** identification of all the relevant literature, followed by balanced synthesis and evaluation – **minimising biases** and **questionable reporting practices**[4,5]. However, popular guidelines for conducting and reporting SRs[6], have been designed for the synthesis of **intervention research**, meaning items are focused more on intervention and outcome, rather than detailed description of methodology. This is problematic for pre-registration of **non-intervention** SRs, as **not all items included in popular guidelines can be addressed**[7] - comprising both the transparency and robustness of the protocol.

An illustrative example of challenges encountered using PROSPERO[7]:

20. * Intervention(s), exposure(s).

Give full and clear descriptions or definitions of the nature of the interventions or the exposures to be reviewed. This is particularly important for reviews of complex interventions (interventions involving the interaction of several elements). If appropriate, an operational definition describing the content and delivery of the intervention should be given.

Ideally, an intervention should be reported in enough detail that others could reproduce it or assess its applicability to their own setting. The preferred format includes details of both inclusion and exclusion criteria.

21. * Comparator(s)/control.

Where relevant, give details of the alternatives against which the main subject/topic of the review will be compared (e.g. another intervention or a non-exposed control group). The preferred format includes details of both inclusion and exclusion criteria.

Control or comparison interventions should be described in as much detail as the intervention being reviewed. If the comparator is 'treatment as usual' or 'standard care', this should be described, with attention being paid to whether it is 'standard care' at the time that an eligible study was done, or at the time the review is done.

24. * Main outcome(s).

Give the pre-specified primary (most important) outcomes of the review, including details of how the outcome is defined and measured and when these measurement are made, if these are part of the review inclusion criteria.

- Intervention(s)/exposure(s) cannot be defined, as the review aims to extract and synthesise information about methodology.
- Comparisons and outcomes may vary across studies, depending on methodology used.
- Examples given to guide completion of fields related to intervention(s).
- Insufficient guidance to support data extraction and synthesis of different methodologies.
- Text box limited to 200 words

PUBLICATION BIAS

Data loss can be an issue in eye-tracking research, due to difficulties calibrating and maintaining participant's attention[8]. Individuals with neurodevelopmental conditions are harder to recruit, meaning sample size is often small, at times under-powered[9] and therefore, less tolerant to missing data. **Inclusion of only peer-reviewed studies would overlook the risk of publication bias, and consequently, over-estimate the effectiveness of eye-tracking technology** – leading to overoptimistic conclusions.

METHODS

NON-INTERVENTIONAL, REPRODUCIBLE, & OPEN SYSTEMATIC REVIEWS (NIRO-SR)

The Non-Interventional, Reproducible, and Open Systematic Reviews (NIRO-SR) guidelines and framework is a **68-item checklist** supporting **planning, pre-registering, and reporting of non-intervention research in SRs**. The preprint and open repository for NIRO-SR provided a comprehensive resource, which enabled us to pre-register:

- **Background, aims, research questions.**
- **Search strategy** (key components, search query for each database, free text & controlled vocab).
- **Screening** (applications/software used, manuals for co-reviewers, inclusion/exclusion criteria).
- **Data extraction** (variables of interest, forms/tables for co-reviewers).
- **Critical appraisal** (assessing risk of bias, methodological quality, publication bias).
- **Synthesis** (headings, tables, assessment of heterogeneity, weight of evidence).
- **Transparency** (scoping searches, conflicts of interest, updates since initial pre-registration).
- **Co-reviewers** (% reviewed at each stage, number of co-reviewers, how disagreements will be resolved).

Scan to see **pre-registered protocol** & **search strategy** using NIRO-SR.

LITERATURE SEARCHES

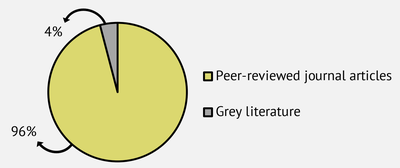
Key components of search strategy:

Intellectual disability	Eye-tracking/social cognition
• Idiopathic/syndromic. • Synonyms for each.	• Not unitary concepts. • Overlap (e.g., face-scan*).

Searches were conducted for peer-reviewed journal articles and grey literature (conference proceedings or posters, book chapters, dissertations, preprints, manuscripts under review):

- **Databases** *(PsycINFO, MEDLINE, Embase, Web of Science).*
- **Listservs** (*COGDEVSOC, ID-Research UK, Dev-Europe).*
- **Citation tracking** (*backwards & forwards).*

Types of literature included in review



CO-REVIEWER RELIABILITY

Agreement across co-reviewers (N; % studies co-reviewed)

Cohen's Kappa Coefficient (*κ*)

- .85 **Titles** (N = 3414; 50%).
- .89 **Abstracts** (N = 337; 100%).
- .92 **Quality rating** (N = 51; 100%).
- .90 **Data extraction** (N = 49; 30.6%).

SCREENING & DATA EXTRACTION

Detailed instructions on the screening and data extraction process were **piloted by a blind reviewer**, and facilitated the **standardisation** of review process across all co-reviewers.

Data extraction form for group characteristics

Intellectual disability (ID)	Comparison group
Sample size. Total sample included in analyses. **Chronological age.** Mean age and SD. **ID aetiology.** Idiopathic or genetic syndrome. **General ability.** Reported metric of IQ, such full-scale intelligence (e.g., Wechsler Intelligence Scale for Children; Wechsler, 2003) and/or adaptive behaviour composite score (e.g., Vineland Adaptive Behaviour Scales [VABS], Sparrow et al., 2005). Language ability as measured using either a specific scale (e.g., British Picture Vocabulary Scale [BPVS]; Dunn et al., 2009) or subscale (e.g., VABS communication subscale; Sparrow et al., 2005). Non-verbal ability, measured using either a specific measure (e.g., Raven's Coloured Progressive Matrices; Raven et al., 1998) or subscale (e.g., WISC processing speed; Weschler, 2003).	**Group.** A sample without ID specified, for example neurotypical or autism groups. Total sample (N) will be reported. In genetic syndromes where there is a known bimodal intelligence distribution (e.g., Klinefelter syndrome [Leggett et al., 2010], tuberous sclerosis complex [Tye et al., 2019] and CHARGE syndrome [Hsu et al., 2014]), either clinical diagnosis of ID or an appropriate metric indicating ID (e.g., IQ < 70) must be reported. **Matched**. If comparison group has been matched to the ID group, then criteria should be specified (e.g., BPVS).

Synthesis of group characteristics in excel

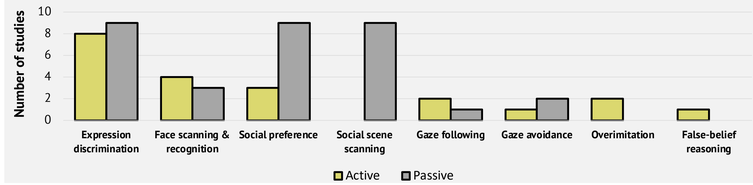
Author	Quality	ID sample/s	CA	General ability	Comparison	Matched?
Campbell et al. (2010)	10	22q11.2 deletion syndrome	17.2 ± 3.2	FSIQ 72.8 ± 13.2	22q11DS (17) TD (17)	CA, gender
Crawford et al. (2015) a	11	Fragile X syndrome	19.7 ± 9.07	VABSr 357.9 ± 95.6	FXS (13) ASD (15)	VABSr
Crawford et al. (2015) b	11	Cornelia de Lange syndrome Rubinstein Taybi syndrome	18.4 ± 9.8 17.3 ± 10.1	VABSc 59.9 ± 25.0 VABSc 58.5 ± 15.1	CdLS (15) RTS (17)	CA, gender, SCQ, VABSc
Dalton et al. (2008)	8	Fragile X syndrome	20.7 ± 2.8	FSIQ 66.1 ± 23.8	FXS (9) ASD (14) TD (15)	NA
Debladis et al. (2019)	12	Prader-Willi syndrome	28.0 ± 8.0	FSIQ 57.0 ± 10.0	PWS (39) TD (20)	CA, gender
Farzin et al. (2009)	9	Fragile X syndrome	17.0 ± 6.8	FSIQ 58.4 ± 9.8	FXS (16) TD (16)	CA, gender
Farzin et al. (2011)	10	Fragile X syndrome	18.8 ± 10.7	FSIQ 57.5 ± 14.5	FXS (15) TD (20)	CA
Franchini et al. (2016)	9	22q11.2 deletion syndrome	18.2 ± 5.9	FSIQ 69.5 ± 11.3	22q11DS (35) TD (31)	CA
Gomez et al. (2020)	11	Williams syndrome	12.4 ± 3.8	FSIQ 63.0 ± 12.7	WS (22) TD (21)	CA, gender
Hanley et al. (2013)	11	Williams syndrome	21.9 ± 9.3	BPVSr 87.6 ± 30.5	WS (15) TD-CA (15) TD-BPVS (14)	CA/BPVS
Hong et al. (2019)	8	Fragile X syndrome	16.6 ± 6.1	VABSc 61.2 ± 12.0	FXS (17) TD (17)	CA, gender
Kirk et al. (2013)	10	Williams syndrome	23.6 ± 6.9	BPVSr 132.0 ± 18.9	WS (13) TD-CA (13) TD-MA (13)	CA/MA
McCabe et al. (2011)	10	22q11.2 deletion syndrome	17.4 ± 3.1	FSIQ 73.8 ± 13.6	22q11DS (18) TD (17)	CA, gender
McCabe et al. (2013)	9	22q11.2 deletion syndrome	16.8 ± 3.7	FSIQ 72.1 ± 13.0	22q11DS (20) ASD (14) TD (31)	CA
Porter et al. (2010)	9	Williams syndrome	25.1 ± 11.7	FSIQ 61.0 ± 15.0	WS (16) TD (16)	MA, gender
Shaw & Porter (2013)	11	Fragile X syndrome	24.8 ± 12.9	FSIQ 64.0 ± 13.7	FXS (16) TD-CA (16) TD-MA (16)	CA/MA

RESULTS

KEY FINDINGS

A range of idiopathic (13%) and syndromic (Williams [28%]; Fragile X [24%]; 22q11.2 [10%]; Rett [5%]; Down [5%]; Phelan-McDermid [5%]; Cornelia de Lange [3%]; Rubinstein-Taybi [3%], Angelman [2%]; Prader-Willi [2%]) intellectual disability groups were studied. Eye movement data indicated **atypical social-cognitive processes**, particularly when compared to neurotypical samples. Some evidence of association between eye movement data and social behaviour.

Task demand across eye-tracking studies measuring social-cognitive processes



EVALUATION OF OPENESS & REPRODUCIBILITY

- **Variability** in eye-tracking protocol and **heterogeneity** of stimuli used, with the majority of studies using a novel paradigm.
- Studies were often **limited by sample size** and at times ran **exploratory analyses**, increasing the potential for sample dependent results and Type 1 error.
  - Only **one replication**[11] identified!

CONCLUSIONS

SYSTEMATIC REVIEW PRE-REGISTRATION

- ★ Detailed protocol written from outset (great if new to research area and/or SRs!).
- ★ Search query defining complex concepts made openly accessible.
- ★ Maximised the transparency and robustness of review processes.
- ★ Supported efficient and reliable screening, data extraction and synthesis across reviewers.
- ? Similar level of detail as a Registered Report…

INCLUSION OF GREY LITERATURE

- ★ Requesting literature on listservs opens an opportunity to discuss work and collaborate.
- ? Researchers must be motivated to share grey literature (e.g., upload preprint, reply to listerv request) in order to address publication bias.
- ? Grey literature is more likely to be excluded in SRs due to low methodological quality[12].

SUGGESTIONS FROM THE REVIEW

We recommend **presenting eye-tracking protocols transparently**, and **developing a bank of open-access, validated eye-tracking stimuli**, to **encourage replication** of findings within/between intellectual disability groups and **opportunities for data sharing**. Collaborative and open eye-tracking methods will strengthen theoretical and clinical implications regarding social cognition in intellectual disability.

References

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Preprint coming soon!