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Diagnosing developmental prosopagnosia: repeat assessment using the Cambridge Face Memory Test

Ebony Murray and Sarah Bate

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Revised submission: 6 August 2020
Final acceptance: 12 August 2020

Note: Reports are unedited and appear as submitted by the referee. The review history appears in chronological order.

Review History

RSOS-200884.R0 (Original submission)

Review form: Reviewer 1

Is the manuscript scientifically sound in its present form?
Yes

Are the interpretations and conclusions justified by the results?
Yes

Is the language acceptable?
Yes

Do you have any ethical concerns with this paper?
No

Have you any concerns about statistical analyses in this paper?
No

Recommendation?
Accept with minor revision (please list in comments)

Comments to the Author(s)
While the CFMT has been used extensively in Psychology to screen for Developmental Prosopagnosia, its test-retest reliability has not been examined. This is an important omission that is addressed by this paper. Performance on the CFMT is compared at different time points and also for different contexts (laboratory and online) and contrasted with several other appropriate – and commonly used – comparisons (CFPT, famous face recognition test, CCMT). The results show that test-retest reliability of the CFMT falls just short of psychometric standards at .68. Important individual cases are noted – for example, of the laboratory participants that were impaired on the CFMT at time 1, almost half (10/21) did not meet these criteria at time 2. The paper also presents comparison with the CFMT-Aus, investigates practice effects, and looks at shortening of the CFMT.

I read this work with great interest and, in my view, it is publishable in its present form. Test-retest reliability has been ignored for far too long in this domain, but is becoming more pertinent than ever. The importance of understanding test-retest reliability is laid out clearly in the introduction and motivated well. The methods are sound and the analyses are appropriate and carried out carefully. The results section is extensive and requires some careful focused reading. I would not cut it down but wonder whether the Discussion can be shortened to reduce some repetition here. And on a very minor note, I could not see any cases indexed by an Asterix in Table 5 in my copy of manuscript.

Review form: Reviewer 2 (Meike Ramon)

Is the manuscript scientifically sound in its present form?
Yes

Are the interpretations and conclusions justified by the results?
No

Is the language acceptable?
Yes

Do you have any ethical concerns with this paper?
No

Have you any concerns about statistical analyses in this paper?
No

Recommendation?
Accept with minor revision (please list in comments)

Comments to the Author(s)
See comments attached (Appendix A).

Decision letter (RSOS-200884.R0)

We hope you are keeping well at this difficult and unusual time. We continue to value your support of the journal in these challenging circumstances. If Royal Society Open Science can assist you at all, please don't hesitate to let us know at the email address below.

Dear Ms Murray

On behalf of the Editors, I am pleased to inform you that your Manuscript RSOS-200884 entitled "Diagnosing Developmental Prosopagnosia: Repeat Assessment using the Cambridge Face Memory Test" has been accepted for publication in Royal Society Open Science subject to minor revision in accordance with the referee suggestions. Please find the referees' comments at the end of this email.

The reviewers and handling editors have recommended publication, but also suggest some minor revisions to your manuscript. Therefore, I invite you to respond to the comments and revise your manuscript.

• Ethics statement
If your study uses humans or animals please include details of the ethical approval received, including the name of the committee that granted approval. For human studies please also detail whether informed consent was obtained. For field studies on animals please include details of all permissions, licences and/or approvals granted to carry out the fieldwork.

• Data accessibility
It is a condition of publication that all supporting data are made available either as supplementary information or preferably in a suitable permanent repository. The data accessibility section should state where the article's supporting data can be accessed. This section should also include details, where possible of where to access other relevant research materials such as statistical tools, protocols, software etc can be accessed. If the data has been deposited in an external repository this section should list the database, accession number and link to the DOI for all data from the article that has been made publicly available. Data sets that have been deposited in an external repository and have a DOI should also be appropriately cited in the manuscript and included in the reference list.

If you wish to submit your supporting data or code to Dryad (http://datadryad.org/), or modify your current submission to dryad, please use the following link:
http://datadryad.org/submit?journalID=RSOS&manu=RSOS-200884

• Competing interests
Please declare any financial or non-financial competing interests, or state that you have no competing interests.

• Authors' contributions
All submissions, other than those with a single author, must include an Authors' Contributions section which individually lists the specific contribution of each author. The list of Authors should meet all of the following criteria; 1) substantial contributions to conception and design, or acquisition of data, or analysis and interpretation of data; 2) drafting the article or revising it critically for important intellectual content; and 3) final approval of the version to be published.

All contributors who do not meet all of these criteria should be included in the acknowledgements.

We suggest the following format:
AB carried out the molecular lab work, participated in data analysis, carried out sequence alignments, participated in the design of the study and drafted the manuscript; CD carried out the statistical analyses; EF collected field data; GH conceived of the study, designed the study, coordinated the study and helped draft the manuscript. All authors gave final approval for publication.

• Acknowledgements
Please acknowledge anyone who contributed to the study but did not meet the authorship criteria.

• Funding statement
Please list the source of funding for each author.

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Because the schedule for publication is very tight, it is a condition of publication that you submit the revised version of your manuscript before 29-Jul-2020. Please note that the revision deadline will expire at 00.00am on this date. If you do not think you will be able to meet this date please let me know immediately.

To revise your manuscript, log into https://mc.manuscriptcentral.com/rsos and enter your Author Centre, where you will find your manuscript title listed under "Manuscripts with Decisions". Under "Actions," click on "Create a Revision." You will be unable to make your revisions on the originally submitted version of the manuscript. Instead, revise your manuscript and upload a new version through your Author Centre.

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Once again, thank you for submitting your manuscript to Royal Society Open Science and I look forward to receiving your revision. If you have any questions at all, please do not hesitate to get in touch.

Kind regards,
Anita Kristiansen
Editorial Coordinator

Royal Society Open Science
openscience@royalsociety.org

on behalf of Dr Bruno Rossion (Associate Editor) and Essi Viding (Subject Editor)
openscience@royalsociety.org

Reviewer comments to Author:
Reviewer: 1

Comments to the Author(s)
While the CFMT has been used extensively in Psychology to screen for Developmental Prosopagnosia, its test-retest reliability has not been examined. This is an important omission that is addressed by this paper. Performance on the CFMT is compared at different time points and also for different contexts (laboratory and online) and contrasted with several other appropriate – and commonly used – comparisons (CFPT, famous face recognition test, CCMT). The results show that test-retest reliability of the CFMT falls just short of psychometric standards at .68. Important individual cases are noted – for example, of the laboratory participants that were impaired on the CFMT at time 1, almost half (10/21) did not meet these criteria at time 2. The paper also presents comparison with the CFMT-Aus, investigates practice effects, and looks at shortening of the CFMT.

I read this work with great interest and, in my view, it is publishable in its present form. Test-retest reliability has been ignored for far too long in this domain, but is becoming more pertinent than ever. The importance of understanding test-retest reliability is laid out clearly in the introduction and motivated well. The methods are sound and the analyses are appropriate and carried out carefully. The results section is extensive and requires some careful focused reading. I would not cut it down but wonder whether the Discussion can be shortened to reduce some repetition here. And on a very minor note, I could not see any cases indexed by an Asterix in Table 5 in my copy of manuscript.

Reviewer: 2

Comments to the Author(s)
See comments attached

Author's Response to Decision Letter for (RSOS-200884.R0)

See Appendix B.

Decision letter (RSOS-200884.R1)

We hope you are keeping well at this difficult and unusual time. We continue to value your support of the journal in these challenging circumstances. If Royal Society Open Science can assist you at all, please don't hesitate to let us know at the email address below.

Dear Ms Murray,

It is a pleasure to accept your manuscript entitled "Diagnosing Developmental Prosopagnosia: Repeat Assessment using the Cambridge Face Memory Test" in its current form for publication in Royal Society Open Science.

You can expect to receive a proof of your article in the near future. Please contact the editorial office (openscience_proofs@royalsociety.org) and the production office (openscience@royalsociety.org) to let us know if you are likely to be away from e-mail contact -- if you are going to be away, please nominate a co-author (if available) to manage the proofing process, and ensure they are copied into your email to the journal.

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Thank you for your fine contribution. On behalf of the Editors of Royal Society Open Science, we look forward to your continued contributions to the Journal.

Best regards,
Lianne Parkhouse
Editorial Coordinator
Royal Society Open Science
openscience@royalsociety.org

on behalf of Dr Bruno Rossion (Associate Editor) and Professor Essi Viding (Subject Editor)
openscience@royalsociety.org

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Appendix A

Review of RSOS-200884

The authors report an investigation into aspects concerning administration of the CFMT and other tools for the diagnosis of developmental prosopagnosia (DP). The authors determine the CFMT's (1) test-retest reliability (across time and administration modus), (2) relationship to procedurally identical, yet independent measures of face and object processing, (3) diagnostic value when 1/3 of trials are omitted. We appreciate the motivation for this study, which addresses important issues in DP research, and find the manuscript well written. We recommend publication after addressing the issues detailed below. We hope the authors will find our comments constructive and remain at their disposal for questions.

Sincerely,
Meike Ramon and Christopher Turner

Major

1. *Subject selection and identification as DPs*

The authors state that "All participants in this study self-referred to the research team reporting difficulties with face recognition." One would assume that over 8 years, more than 70 people will have contacted the lab, or accessed www.prosopagnosiaresearch.org. Please describe in detail the criteria applied to select individuals of the two subsets reported here, and if any individuals withdrew from the study. Provide a summary of individual scores across all tests, and refer to any previous publication of the cases and use consistent acronyms to allow cross-study comparison. This aligns with the authors emphasis of the importance of data sharing (p.18f.)

Considering the authors aimed to address "the diagnostic utility of administering" different tests, We would like to see the previous formal criteria used for DP diagnosis contrasted with those proposed based on their findings. Specifically, in our opinion, readers will be interested in seeing the differences in DP across both sets of criteria.

Note also that here norms of the original CFMT publication were used to identify atypical scores, which were collected in the lab. Based on previous findings, it is reasonable to assume that this original sample would have produced data suggesting a higher cut-off (cf. e.g. CFMT scores reported by Bobak et al., 2016, Frontiers in Psychology vs. Stacchi et al., 2020). Given that there is now ample data from the CFMT(+), please provide comparative data from a larger, more heterogeneous sample as well. This could yield substantially

different classification than provided on p.14, l.7ff. (21/30 of lab-tested and 16/40 of online-tested Ss below the cut-off).

2. *Data presentation / comprehensibility*

The relationships between individual test performance across various measures are very interesting, but we found it hard to integrate based on how the data are currently provided. Please represent the (individual) data in a more reader-friendly, visual way that allows a direct comparison of scores across different modes of testing and tests.

3. *Measures subject to analyses*

P.21, l.12ff.: in addition to considering multiple object categories, note that Geskin & Behrmann's findings resulted from parallel consideration of accuracy *and* RTs, which were not considered here. Please provide analyses related to this additional measure, which has proven to be highly informative when it comes to impaired populations (cf eg Delvenne et al. 2004; Michel & Rossion, 2018).

Minor

- For the less informed reader, please include a short terminological distinction between the different sub-processes involved in face processing in relation to each test used. This is not related to the content of this manuscript, but rather to address the conceptual confusion propagated by others, who you adopt a less stringent approach to definitions and concepts.
- The two groups did not differ according to age at their initial testing point […] nor at the second – would we expect them to?
- Include a visualization of the different tests, as well as a graphical summary / diagram of the information provided in the "Data overview" section.
- p.12, l.44ff.: "CFPT scores did not correlate with any stage of the CFMT, supporting the distinction between face perception and face memory and, consequently, existing models of face processing [63]." There are multiple tests of face perception that actually do correlate with the CFMT. We would like to see this acknowledged, as well as a discussion of the CFPT's utility.
- Provide demographic information for the subjects and include how it was recorded in the procedure. (Subject location and ethnicity is alluded to in the text but this information is not provided.)
- Clarify the Table 5 note "* denotes cases where there is a reversed pattern of impairment", as there is no asterisk ("*") in the table.

Appendix B

We wish to thank both reviewers for their comments and feedback, and we are especially pleased to hear that Reviewer 1 believes the manuscript is publishable in its present form. We have made some adjustments to the manuscript to address the constructive comments provided by the reviewers, and we detail these below.

- **Please describe in detail the criteria applied to select individuals of the two subsets reported here, and if any individuals withdrew from the study. Provide a summary of individual scores across all tests, and refer to any previous publication of the cases and use consistent acronyms to allow cross-study comparison.**

This information has now been included in the Participants subsection, P.7. We have pointed readers to an existing publication, stated that no participants withdrew, and have detailed that all scores and details of the participants can be found in the Supplementary Information.

- **We would like to see the previous formal criteria used for DP diagnosis contrasted with those proposed based on their findings. Specifically, in our opinion, readers will be interested in seeing the differences in DP across both sets of criteria.**

Please note that existing DP diagnostic criteria is based on performance across multiple tests (including the CFPT and famous face recognition tests), and not just the CFMT. This paper only examines performance on the CFMT and how repeat testing is important to decipher whether or not a participant is impaired on this particular task. Because of this, and because not all participants took part in the CFPT and famous faces test that are required for a complete diagnostic profile, we cannot address the issue of wider patterns of performance here. We have made it clearer throughout the manuscript (see *Data Overview*, and P.12) that online participants did not complete the famous faces test, and only 19 of these participants completed the CFPT.

- **Given that there is now ample data from the CFMT(+), please provide comparative data from a larger, more heterogeneous sample as well. This could yield substantially different classification than provided on p.14.**

We thank you for this recommendation and have included information on P. 13 to address this. Data from a larger sample, such as that of Bowles et al. (2009), echoed that of the original publication and also suggested a cut-off of 42 and below. With this in mind, the present data do not change and no further edits are made.

- **Please represent the (individual) data in a more reader-friendly, visual way that allows a direct comparison of scores across different modes of testing and tests**

Thank you for this recommendation. We have now included a Figure which compares CFMT1 and CFMT2 scores, for lab-based participants and online participants.

- **In addition to considering multiple object categories, note that Geskin & Behrmann's findings resulted from parallel consideration of accuracy and RTs, which were not considered here. Please provide analyses related to this additional measure, which has proven to be highly informative when it comes to impaired populations**

Thank you for this comment. While we appreciate the importance of assessing RTs as well as accuracy, particularly for face perception tasks (e.g. Rossion & Michel, 2018), RTs are not assessed in the CFMT memory-based paradigm. Further, because we used the standard instructions of the task, participants were not informed that RTs would be analysed. Therefore, we have no reliable basis to do so here.

- **For the less informed reader, please include a short terminological distinction between the different sub-processes involved in face processing in relation to each test used.**

This information can already be found on P.3-4 in the Introduction.

- **The two groups did not differ according to age at their initial testing point [...] nor at the second –would we expect them to?**

Yes, this is possible as we did not impose a consistent time period between participants' first and second attempts. This information is included on P.11 where it is also explained that the time-lapse is a necessary covariate in the analyses.

- **Include a visualization of the different tests, as well as a graphical summary / diagram of the information provided in the "Data overview"**

  We have now included a visual for an example trial of the original CFMT (Figure 1). There is also a visualisation of the data overview section as a flowchart, now provided as Figure 2.

- **There are multiple tests of face perception that actually do correlate with the CFMT. We would like to see this acknowledged, as well as a discussion of the CFPT's utility.**

  We have now directed readers to a selection of papers which did find a correlation between tests of face perception and the CFMT on P.11. However, we have not included a discussion of the CFPT's utility as we believe this is out of the scope of the present paper. Furthermore, Reviewer 1 has suggested that the Discussion section is reduced in length and we therefore prioritise other information there.

- **Provide demographic information for the subjects and include how it was recorded in the procedure. (Subject location and ethnicity is alluded to in the text but this information is not provided.)**

  Thank you for bringing this to our attention. We have now included demographic information in the *Participants* subsection, and stated how it was collected in the *Procedure*.

- **Clarify the Table 5 note "* denotes cases where there is a reversed pattern of impairment", as there is no asterisk ("*") in the table.**

  We thank the reviewers for identifying this and we have now amended the Table.