PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	3D-manufactured Patient-Specific Models of Congenital Heart Defects for Communication in Clinical Practice: Feasibility and Acceptability
AUTHORS	Biglino, Giovanni; Capelli, Claudio; Wray, Jo; Schievano, Silvia; Leaver, Lindsay-Kay; Khambadkone, Sachin; Giardini, Alessandro; Derrick, Graham; Jones, Alexander; Taylor, Andrew

VERSION 1 - REVIEW

REVIEWER	ADJ ten Harkel Leiden University Medical Center, Leiden, The Netherlands
REVIEW RETURNED	07-Jan-2015

GENERAL COMMENTS	The authors of the present article present their experience with 3D models in communicating with parents the childs' heart disease. Although this is an interesting new topic, there are several shortcomings. Overall, the only conclusion that can be made is that both physicians and parents like the models, which is not really surprising. Other potential effects of the 3 D model as adjustment following treatment, psychological impact are not studied. Furthermore the us of the models did not improve the learning of the parents. The methods are incomplete. It is not clear why MRI was made in all these children, was it a routine MRI, medically necessary or otherwise. What was the timing between the MRI and the final consultation? Were both parents involved or just one? The power calculation on page 6 can be moved to the statistics section.

REVIEWER	John P. Cheatham Nationwide Children's Hospital Columbus, Ohio, USA
REVIEW RETURNED	23-Mar-2015

GENERAL COMMENTS	I commend you on a thoughtful manuscript. Undoubtedly, 3D printing has changed the way interventional cardiologists and perhaps cardiac surgeons plan and prepare for complex procedures. As specialists, we want our patients and their parents to be as informed and knowledge as possible. Assessing the utility of patient-

	specific 3D models to achieve this goal is important because as you point out - perhaps a complex 3D model may actually hinder the "non-expert" learning process and negatively impact how much they end up understanding. Some general and specific points: 1. Page 5. First 2 paragraphs. Overall, the introduction is too long and wordy. Several important points are made that set up the manuscript well but the first two paragraphs have several run-on sentences that should be condensed. Shortening the introduction will strengthen this manuscript further. 2. Page 22, Table 1. Can you provide a U.S. equivalent of 6th form so the US readers of this manuscript have a sense of this level of education. 3. Page 11, Line 47. This sentence does not need to be included in the results. If you wish to state something about how the study was powered this can be moved up to the data analysis section. 4. The range of diagnoses negatively impacted your ability to interpret the overall affect the 3D model had on the encounter. Controlling for a few diagnoses or perhaps limiting it to one diagnosis only would improve this study significantly. While perhaps difficult to do – one could repeat this study using only patients being considered for either catheter-based or surgically placed pulmonary valve replacements. 5. The manuscript needs to be further polished with a focus on eliminating run-on sentences and some redundant sentences.

VERSION 1 – AUTHOR RESPONSE

Reviewer Name ADJ ten Harkel
Institution and Country Leiden University Medical Center, Leiden, The Netherlands
Please state any competing interests or state 'None declared': None declared
Please leave your comments for the authors below

The authors of the present article present their experience with 3D models in communicating with parents the childs' heart disease. Although this is an interesting new topic, there are several shortcomings.
We thank the Reviewer for the constructive feedback.

Overall, the only conclusion that can be made is that both physicians and parents like the models, which is not really surprising.
This is a first attempt to study and quantify the usefulness (and liking) of this technology in the realm of cardiology visits and communication. While intuitively there can indeed be benefits of 3D models for communication (often advocated in the literature, but never systematically tested or quantified), there could be other intuitive aspects whereby models might not be necessarily liked by either users, for instance:
• Cardiologists: Clinicians might find that using models requires unnecessary explanations on, e.g., how images are processed, where the model comes from, who made it etc
• Parents: An element of shock has emerged from this study, when realising the anatomy is patient-specific, and this may have impinged more on parental liking
Preliminary data from this study can then inform future studies on long-term knowledge (for parents and patients, still not evaluated), repeated use of the models, additional psychological considerations

(e.g. deflecting anxiety), having established and quantified liking in a relatively large group of congenital heart disease cases.

Other potential effects of the 3D model as adjustment following treatment, psychological impact are not studied.
We agree with the Reviewer that adjustment following treatment would be an extremely interesting and important point to evaluate, requiring a longer study with follow up. Based on the encouraging preliminary results from this study, we are in fact designing a study involving the patients themselves.

Addition to the manuscript (page 15)
Personalised models could facilitate this learning process, with a potential long-term impact on lifestyle adjustments that would be important to observe and quantify in future studies. From a psychological standpoint, other factors (e.g. deflecting anxiety) could also be investigated in future studies.

Furthermore the use of the models did not improve the learning of the parents.
Indeed, we thought this was an interesting finding. It should be pointed out that short-term knowledge did not improve, and it was tested with a simple method (i.e. diagrams and keywords). Future work to evaluate any improvements in knowledge could involve in-depth interviews in the survey itself as well as evaluation of long-term knowledge, e.g. assess the same groups after one year, evaluating whether those who have kept the model have improved their understanding of the anatomy and/or have been prompted to gather additional information. This finding is important as it was systematically acquired in a large group, in contrast to advocated benefits in the literature.

The methods are incomplete. It is not clear why MRI was made in all these children, was it a routine MRI, medically necessary or otherwise. What was the timing between the MRI and the final consultation?
The CMR studies were performed based on medical indication and were retrieved retrospectively for the 3D reconstructions. The most recent available CMR was always used. Average timing between CMR and consultation was 1.7±1.6 years.

Addition to the manuscript (Page 6)
CMR studies were performed based on medical indication and were retrieved retrospectively for the 3D reconstructions. Timing between CMR and consultation was 1.7±1.6 years.

Were both parents involved or just one?
One parent was involved. In the large majority of cases, only one parent was accompanying the patient.

Addition to the manuscript (Page 7)
In cases in which both parents of a child were attending, only one was asked to participate.

The power calculation on page 6 can be moved to the statistics section.
We have moved this paragraph to the Methods section.

Addition to the manuscript (Page 8, Sample size)
Pilot data was not available to power the study. A retrospective power calculation based on clinicians' rating, as a more objective measure than perceived parental knowledge, showed that given the observed mean difference, standard deviations and the number of subjects included in each group, the study was sufficiently powered at 80% with $p = 0.05$.

Reviewer Name John P. Cheatham
Institution and Country Nationwide Children's Hospital, Columbus, Ohio, USA
Please state any competing interests or state ‘None declared’: None declared
Please leave your comments for the authors below

Dear authors:
I commend you on a thoughtful manuscript. Undoubtedly, 3D printing has changed the way interventional cardiologists and perhaps cardiac surgeons plan and prepare for complex procedures. As specialists, we want our patients and their parents to be as informed and knowledge as possible. Assessing the utility of patient-specific 3D models to achieve this goal is important because as you point out - perhaps a complex 3D model may actually hinder the “non-expert” learning process and negatively impact how much they end up understanding. Some general and specific points:
We thank the Reviewer for the positive feedback and for the constructive comments.

1. Page 5. First 2 paragraphs. Overall, the introduction is too long and wordy. Several important points are made that set up the manuscript well but the first two paragraphs have several run-on sentences that should be condensed. Shortening the introduction will strengthen this manuscript further.
The whole Introduction has been revised following the Reviewer’s recommendation.

2. Page 22, Table 1. Can you provide a U.S. equivalent of 6th form so the US readers of this manuscript have a sense of this level of education.
Thank for pointing this out, we have now clarified (12th grade US is equivalent to 6th form UK).

Addition to the manuscript (Page 21, Table 1)
Note: 6th form equivalent to 12th grade in the United States.

3. Page 11, Line 47. This sentence does not need to be included in the results. If you wish to state something about how the study was powered this can be moved up to the data analysis section.
We have moved this paragraph to the Methods section, as suggested (Please see page 8, “Sample size”).

4. The range of diagnoses negatively impacted your ability to interpret the overall affect the 3D model had on the encounter. Controlling for a few diagnoses or perhaps limiting it to one diagnosis only would improve this study significantly. While perhaps difficult to do – one could repeat this study using only patients being considered for either catheter-based or surgically placed pulmonary valve replacements.
This study was a first attempt to collect feedback and responses from a group of about 100 parents and, as no previous knowledge on the topic is available, we decided not to restrict the population to pre-selected diagnoses (e.g. testing only single ventricles) as we might impose a selection bias. On the other hand, we do agree with the Reviewer that, in the light of these results, it would be interesting to collect data for specific conditions (e.g. the valve replacement scenario the Reviewer suggests would be very interesting indeed) and refine our understanding this way. Numbers are unfortunately too small to perform sub-group analysis in this study. We have acknowledged this in the Discussion of the study.

Addition to the manuscript (Page 15)
While it would be interesting to assess the usefulness of 3D models for specific diagnoses or interventions (e.g. pulmonary valve replacement), the number of cases in this study is too small to allow for sub-group analysis.

5. The manuscript needs to be further polished with a focus on eliminating run-on sentences and some redundant sentences.
The whole manuscript has been re-read and revised.