

## PEER REVIEW HISTORY

BMJ Paediatrics Open publishes all reviews undertaken for accepted manuscripts. Reviewers are asked to complete a checklist review form and are provided with free text boxes to elaborate on their assessment. These free text comments are reproduced below.

This paper was submitted to a another journal from ADC but declined for publication following peer review. The paper was subsequently accepted for publication at BMJ Paediatrics Open.

### ARTICLE DETAILS

<b>TITLE (PROVISIONAL)</b>	Newborn biliary atresia screening with the stool colour card: A questionnaire survey of parents
<b>AUTHORS</b>	Borgeat, Morgane; Korff, Simona; Wildhaber, Barbara

### VERSION 1 – REVIEW

<b>REVIEWER</b>	Reviewer name Claus Petersen Institution and Country Pediatric Surgery Hannover Medical School Germany Competing interests We introduced SCC in Lower Saxony, Germany, in 2017 and we´re going to extend the obligatory use for Germany in accordance with governmental provisions (GBA)
<b>REVIEW RETURNED</b>	24-Feb-2018

<b>GENERAL COMMENTS</b>	<p>Actually, there is no doubt about the fact that screening for biliary atresia (BA) is a precondition for improving the overall outcome of BA patients. Today, we know from several reliable and convincing studies that after introducing stool color cards (SCC) as a screening tool, diagnosis and treatment of BA began earlier and improved the outcome in terms of reduced need of liver transplantation and longer survival with native liver and normal bilirubin. The main hurdle in many places to introduce SCC as an obligatory screening tool is that skeptical people and categorical worriers raise individual, structural and other concerns without providing a feasible alternative. In this particular situation, any study is to be welcomed, which supports the acceptance of SCC at any position of this complex topic.</p> <p>The submitted study shows very clearly that there are - of course - some parents who are worried, when they become confronted with a disease, they never have been heard of and which is potentially life threatening for their newborn child. Taking this into consideration gives a clear signal that particularly those parents have to be informed better about the sense of SCC and the potential benefit for a minority of all newborns.</p> <p>From this point, I consider the submitted paper an extremely important contribution to the still ongoing discussion about introducing SCC as an obligatory screening tool in every country.</p>
-------------------------	--

<b>REVIEWER</b>	Reviewer name Peter Witters Institution and Country University Hospitals Leuven, Belgium Competing interests – None
<b>REVIEW RETURNED</b>	04-Mar-2018

<b>GENERAL COMMENTS</b>	<p>I've read with great interest "Newborn screening with the stool colour card: Does it evoke parental stress?" by Borgeat et al. It describes the results of a questionnaire given to 256 recent parents that received a stool color card.</p> <p>I'm all for the use of the SCC and value the authors' effort to take away any concerns that paediatricians may have in implementing this tool. However, one must admit that this is a very small study, with a very low response rate. Certain findings of the study are based only on 19 responses. This could easily be improved by a telephone consultation.</p> <p>Minor comments: the authors are not clear whether this asks additional time from the paediatrician or not. "most of the parents do not need further explanations..." vs that it should be discussed with the parents.</p> <p>The percentages in the abstract do not add up to 100%, please rephrase.</p> <p>Page 4 R31-34: please rephrase Page 4 R 53: jaundice clearance.</p>
-------------------------	---

<b>REVIEWER</b>	<p>Reviewer name Ming-Chih Lin Institution and Country Children's Medical Center, Taichung Veterans General Hospital, Taichung, Taiwan Competing interests None to declare</p>
<b>REVIEW RETURNED</b>	16-Mar-2018

<b>GENERAL COMMENTS</b>	<p>This is a questionnaire research. The main purpose is to investigate the anxiety caused by delivering a stool color card to parents for screening biliary atresia. This article is well written. I have several comments here.</p> <ol style="list-style-type: none"> <li>1. The return rate of questionnaire is lower than 50%. The authors should describe whether there is difference in the background between those who wrote the questionnaire and those who did not write the questionnaire.</li> <li>2. The authors concluded that the stool color card would not increase parents' anxiety. However, 18% of those parents expressed certain degree of anxiety. For a population-based screening program, the cost is not low. Furthermore, because biliary atresia is a rare disease, whether stool color card is cost-effective should be further discussed.</li> <li>3. The authors should describe the referring system for babies with suspected biliary atresia. Because efficient referring system is the key for the success of screening biliary atresia by stool color card.</li> </ol>
-------------------------	--

### VERSION 1 – AUTHOR RESPONSE

Reviewer: 1

- Actually, there is no doubt about the fact that screening for biliary atresia (BA) is a precondition for improving the overall outcome of BA patients.

Today, we know from several reliable and convincing studies that after introducing stool color cards (SCC) as a screening tool, diagnosis and treatment of BA began earlier and improved the outcome in terms of reduced need of liver transplantation and longer survival with native liver and normal bilirubin. The main hurdle in many places to introduce SCC as an obligatory screening tool is that skeptical people and categorical worriers raise individual, structural and other concerns without providing a feasible alternative. In this particular situation, any study is to be welcomed, which supports the acceptance of SCC at any position of this complex topic. The submitted study shows very clearly that there are - of course - some parents who are worried, when they become confronted with a disease, they never have been heard of and which is potentially life threatening for their newborn child. Taking this into consideration gives a clear signal that particularly those parents have to be informed better about the sense of SCC and the potential benefit for a minority of all newborns. From this point, I consider the submitted paper an extremely important contribution to the still ongoing discussion about introducing SCC as an obligatory screening tool in every country.

Resoponse: We thank Reviewer 1 for these comments.

Reviewer: 2

- I've read with great interest "Newborn screening with the stool colour card: Does it evoke parental stress?" by Borgeat et al. It describes the results of a questionnaire given to 256 recent parents that received a stool color card. I'm all for the use of the SCC and value the authors' effort to take away any concerns that paediatricians may have in implementing this tool. However, one must admit that this is a very small study, with a very low response rate. Certain findings of the study are based only on 19 responses. This could easily be improved by a telephone consultation.

We agree with Reviewer 2: indeed, the study is small, yet the first of such character. We added a paragraph at the end of the discussion, addressing the weakness of the study.

Minor comments:

- The authors are not clear whether this asks additional time from the paediatrician or not. "most of the parents do not need further explanations..." vs that it should be discussed with the parents.

Resoponse: Indeed, we were not clear. We thus added a sentence where we discuss the possible increase in expenditure of time.

- The percentages in the abstract do not add up to 100%, please rephrase.

In the abstract we did not summarize all possible answers, only the most quoted ones, this due to lack of abstract space (word count), and this explains why the percentages do not sum up to 100% for each question. We thus were not able to correct this issue, even though we also believe it would be more elegant and complete!

- Page 4 R31-34: please rephrase

Resoponse: Thank you; rephrasing done.

- Page 4 R 53: jaundice clearance.

Resoponse: Thank you; changed accordingly.

Reviewer: 3

This is a questionnaire research. The main purpose is to investigate the anxiety caused by delivering a stool color card to parents for screening biliary atresia. This article is well written. I have several comments here.

- The return rate of questionnaire is lower than 50%. The authors should describe whether there is difference in the background between those who wrote the questionnaire and those who did not write the questionnaire.

Indeed, there was a difference between the language regions from where the questionnaires were returned. The participants responding from Italian-speaking Switzerland returned 48/50 (96%) of questionnaires, from German-speaking Switzerland 20/100 (20%) were returned, and from the French-speaking 41/106 (39%). Since we omitted to ask for personal details of the responders, we are unable to say if this is due to the cultural background of the parents or due to the different introduction of the questionnaire by the local paediatrician, even more so since Switzerland is a very multi-cultural country in every language region.

Response: We added this information in the results and discussion part.

- The authors concluded that the stool color card would not increase parents' anxiety. However, 18% of those parents expressed certain degree of anxiety.

Response: We believe that we should look more in detail into this "conclusion": When receiving the SCC 5% of parents were worried. When using the SCC 6% of parents were worried. Only during the conversation later on with the paediatrician about pathologies linked to the stool colour 18% of parents expressed that they felt some degree of uneasiness during this conversation (4% "yes a lot", 14% "yes rather than no"). It seems to us, that this is a rather "normal" reaction and not clearly due to the SCC, since parents naturally often feel uncomfortable during the discussion about possible pathologies linked to their baby, mirrored by the 14%. Indeed, 4% were clearly anxious, which is in line with the numbers reported after receiving and during the use of the SCC. The 18% are also in agreement with the generally reported post-natal anxiety (as mentioned in the discussion, up to 18% of mothers experience some degree of anxiety in the first months after birth, and even more so if the newborn is a first-born, references 16-18).

Since this does not seem clear to the reader, we changed this part in the discussion.

- For a population-based screening program, the cost is not low. Furthermore, because biliary atresia is a rare disease, whether stool color card is cost-effective should be further discussed.

Response: We thank the Reviewer for addressing this issue. Yet, we consider that we can refrain from addressing this topic, as it is not in the scope of this paper. It has been addressed in several publications, and we thus concluded that we can add this fact to "What is known" (see also *J Pediatr Gastroenterol Nutr.* 2015 Jan;60(1):91-8: Cost-effective analysis of screening for biliary atresia with the stool color card, Mogul D et al., or *Medicine (Baltimore)*, 2016 Mar; 95(12): e3166: Infant Stool Color Card Screening Helps Reduce the Hospitalization Rate and Mortality of Biliary Atresia. A 14-Year Nationwide Cohort Study in Taiwan, Min Lee et al., or *Pediatrics.* 2009 May;123(5):1280-6: Impact of age at Kasai operation on its results in late childhood and adolescence: a rational basis for biliary atresia screening, Serinet MO et al.)

- The authors should describe the referring system for babies with suspected biliary atresia. Because efficient referring system is the key for the success of screening biliary atresia by stool color card.

Response: We added this information in the introduction.