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https://doi.org/10.1016/j.jpeds.2021.01.062

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Considerations for future research on celiac disease in children with functional constipation

To the Editor:

Recognition of the symptomatology associated with celiac disease will allow for earlier diagnosis and ultimately better patient outcomes. We read the report by Fifi et al and appreciate the efforts they have made to attempt to improve the early diagnosis of celiac disease.[1]

However, we propose several questions regarding the methodology of the study. The authors fail to provide evidence why children under the age of 10 years had questionnaires answered by their parents, whereas those above age 11 years were able to self-report. Previously, research has validated self-reporting tools for children as young as 6 years old.[2,3] In addition, in self-reporting scales such as the Faces Pain Scale-Revised, only children under the age of 7 years had low congruent validity.[4] This evidence indicates that at least those between the ages of 7 and 10 years could have been given the ability to self-report.

The authors did well to recruit from multiple cities in the sample group. They also identify studies in different countries, such as the Netherlands and Turkey, which produced contrasting findings.[5,6] In these alternative studies, recruitment of the participants was from a single city, yet the authors do not comment on this as a potential confounder.

Based on this, we would encourage the authors, and future researchers, to use a consistent tool across their population when reviewing future data. Any further work should also aim to consider location as a confounder before drawing results from the data. In addition, future work would benefit from considering the economic implications of undertaking further work in this area. The quoted cost of diagnosing 1 child with functional constipation with celiac disease in America from a previous decade (over $67 000) will increase with the rest of healthcare costs.[7]

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https://doi.org/10.1016/j.jpeds.2021.01.009

The authors declare no conflicts of interest.

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Reply

To the Editor:

We want to address their disagreement with our determination to allow only children >10 years old to self-report. In their arguments, the authors ignore that our methods followed the Rome IV Committee guidelines for the use of the official questionnaire for the diagnosis of functional gastrointestinal disorders (Questionnaire on Pediatric Functional Gastrointestinal Disorders, QPGS-IV).[1] This document recommended using the self-report questionnaire in children >10 years of age (as opposed to parental report for children <10 years old). Thus, changing the self-reporting cutoffs as the authors suggested in their letter would contradict the instructions given by the Rome IV committee that issued the questionnaires. This would not only be inappropriate but would also be counterproductive as it would not allow comparing data with other studies that have also strictly followed the instructions on the use of the questionnaire.

Next, Al-Shamaa et al comment that we compared our results with other studies that were not as representative as ours. In our effort to put our data into context, we compared our results with the current literature, which

included many international papers from the US, Iran, Netherlands, and Turkey.[2-6] Unfortunately, these were all single city studies and there were no other multicity studies for comparison. We are being criticized for having a larger and probably more representative study than others, something that should be praised. Still, our study is not devoid of limitations in terms of external validity and we have alerted the readers in our limitations section: "Limitations of our study include the fact that it was conducted in Colombia and so our results might not be reproducible in other settings" and "Although we cannot rule out the possibility that the contradictory conclusions of the studies conducted in different countries are the result of the differences in prevalence of celiac disease and constipation in dissimilar regions, one of the studies with opposite conclusions came from Turkey."

Finally, we agree that the cost of diagnosing a patient with celiac disease must be considered when performing such studies. Actually, we explained the implications of our results in our discussion.[2] Our studies, together with others referenced in our report, found that testing every child with functional constipation would not result in a higher yield than testing all children indiscriminately. Based on these studies, indiscriminate testing of all children with functional constipation would not be cost-effective for the diagnosis of pediatric celiac disease. We hope that studies like ours would help inform the discussion on how to focus testing on those children more likely to have celiac disease reducing healthcare costs.

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https://doi.org/10.1016/j.jpeds.2021.01.010

The authors declare no conflicts of interest.

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