gastrointestinal disorders

MDPI

Systematic Review

The Diagnostic Accuracy of Abdominal X-ray in Childhood Constipation: A Systematic Review of the Literature

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Abstract: Background: Previous systematic reviews have found insufficient and conflicting evidence for an association between the clinical and radiographic diagnosis of functional constipation. Abdominal X-ray is frequently used for the diagnosis of functional constipation in clinical practice. The objective of this study was to evaluate the diagnostic accuracy of abdominal X-ray for the evaluation of functional constipation in children. Results: Three studies were included in the final qualitative analysis. They were heterogeneous in their study design, definition of constipation, and radiologic parameters used to evaluate the abdominal X-rays. Sensitivities ranged from 73–92%, specificities ranged from 26–92%, and diagnostic accuracies ranged from 78–90%. Methods: This study involved a systematic review of English literature published between 2012 and 2022 covering children 2–18 years of age with a diagnosis of functional constipation in whom abdominal X-ray was performed. The databases searched include Medline, Embase, and Scopus. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) and Quality Assessment of Diagnostic Accuracy Studies 2 (QUADAS-2) guidelines were followed. PROSPERO ID: CRD42022301833. Conclusions: There is insufficient evidence to support the use of abdominal X-ray as part of the diagnostic workup of functional constipation. More methodologically rigorous studies are needed to determine the utility of abdominal X-ray in the evaluation of functional constipation. The diagnosis of functional constipation should be based on history and clinical findings.

Keywords: functional constipation; constipation; abdominal X-ray; functional gastrointestinal disorders

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1. Introduction

Childhood constipation is a common condition in children of all ages, with a global pooled prevalence of 9.5% [1], high healthcare costs [2], and a significant impact on the quality of life of affected children and families [3]. The most common form of constipation in children is functional in nature [4], a clinical diagnosis defined by the Rome IV criteria [5,6]. Symptoms of constipation in children include hard and painful bowel movements, infrequent defecation, withholding behaviour, and abdominal pain, often accompanied by faecal incontinence [5,6].

The use of imaging and laboratory tests in functional constipation is of limited value. The main role of laboratory tests is to rule out organic causes of constipation (e.g., coeliac disease, hypothyroidism, and hypercalcaemia) when alarm signs are present. Imaging investigations, such as abdominal X-ray (AXR), which are commonly used to assess for faecal impaction, are also of limited value and not recommended by clinical guidelines [7,8].

While available data discourage the use of AXR for the evaluation of functional constipation in children due to its limited value, radiation exposure, as well as its possible misleading nature [7–14], a significant number of AXRs continue to be performed on

children with constipation, particularly in the emergency department [15–18]. This has led to quality improvement projects that aim to reduce the number of abdominal X-rays performed in such children, with them showing promising results [19–21].

Given that it has been more than 10 years since the last systematic review on this topic was published [10], an updated systematic review is now warranted. The aim of this systematic review was to evaluate the diagnostic accuracy of abdominal X-ray in children with functional constipation.

2. Results

The search identified a total of 1125 articles, of which 855 were screened after removing 270 duplicates. One relevant article was uncovered from the bibliographies of a screened article. After the initial title and abstract screening, 845 articles were excluded, leaving 11 articles for the full-text screen. Of these, only three were included in the final qualitative analysis. Figure 1 shows the PRISMA flowchart. The three included studies were heterogeneous in their study design, the definition of constipation, and the radiologic parameters used to evaluate the AXRs. The selection process was not clearly described in all three studies.

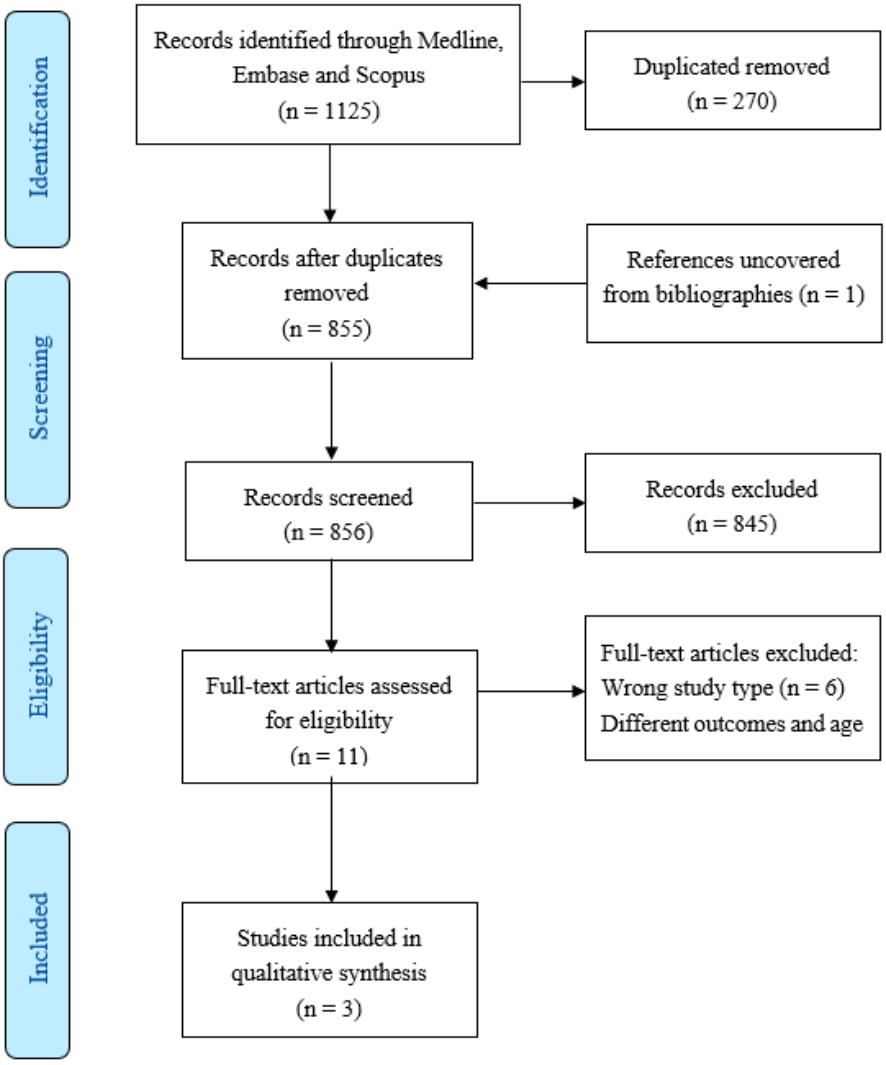

Figure 1. PRISMA flowchart.

Among the included studies, two studies [22,23] were conducted in the USA and one study was conducted in Iran, involving a total of 416 children with constipation. None

of the included studies recruited patients in a consecutive or random manner, or at least this was not clearly specified. While the two case-control studies [22,24] compared AXRs of children with and without a clinical diagnosis of constipation, the cohort study [23] retrospectively collected data on AXRs and calculated the sensitivity and specificity based on a later diagnosis of constipation. The diagnosis of constipation was made by the Rome III criteria in one study, and in two of the studies the definition of constipation was not specified. While in two of the studies the AXRs were interpreted by radiologists [25,26], in the third study both radiologists and clinicians interpreted the AXRs [22]. Table 1 shows the study characteristics.

Table 1. Study characteristics.

Reference	Year	Country	Study Type	Age Range (Years)	Number of Participants	Index Test	Reference Standard (Cases)	Controls
Marks et al. [22]	2013	USA	Retrospective, case–control study	4–12 (mean age: cases: 7.5; controls: 7.8)	80 (40 cases and 40 controls)	AXR criteria: caecal diameter, maximum length of contiguous stool, rectal diameter, total length of collapsed colon/air-filled colon, and length of stool in each colon segment. The total air-to-stool ratio was calculated from the length measurements	Children with FC	Children with constipation who were successfully treated for constipation before AXR
Rezazadeh et al. [24]	2016	Iran	Case–control study	2–14 (mean age [months]: cases: 68.39 ± 33.48; controls: 69.46 ± 32.60, $p = 0.82$)	204 (102 cases and 102 controls)	AXR criteria: Barr, Leech, and Blethyn scores	Children with abdominal pain and FC as per the Rome II criteria	Children with abdominal pain without constipation
Anwar ul Haq et al. [23]	2020	USA	Retrospective cohort study	2–18 (mean age: 10.3 ± 4.3)	132	AXR keywords: "constipation", "stool load", "fecal retention", and "fecal load"	Children diagnosed with constipation	N/A

USA: United States of America; AXR: abdominal X-ray; FC: functional constipation; N/A: not applicable.

Marks et al. [22] compared the AXRs of children with constipation and children whose constipation was successfully treated. The mean age of children with and without constipation was 7.5 and 7.8 years, respectively. The authors found a model with the most accurate predictive capability to identify constipation on AXR, which included a combination of caecal diameter, stool length in the rectum, total stool length, age, and gender, with an AUC of 0.87. Inter- or intra-observer agreement analyses were not investigated in this study. Table 2 shows the study findings.

Table 2. Reported sensitivities, specificities, PPV, NPV, and AUC/ROC.

Study	Sensitivity	Specificity	PPV	NPV	AUC/ROC
Marks et al. [22]	3.7 cm caecal diameter cutoff: 60% 5.9 cm stool length in the rectum cutoff: 63% 33.4 cm for total stool length in the colon cutoff: 70%	3.7 cm caecal diameter cutoff: 61% 5.9 cm stool length in the rectum cutoff: 63% 33.4 cm for total stool length in the colon cutoff: 68%	NA	NA	87% Model of a combination of caecal diameter, stool length in the rectum, total stool length, age, and gender
Rezazadeh et al. [24]	Barr score > 11: 83% Leech score > 9: 92% Blethyn score > 2: 79%	Barr score > 11: 79% Leech score > 9: 80% Blethyn score > 2: 92%	NA	NA	Barr: 87% Leech: 90% Blethyn: 78%
Anwar ul Haq et al. [23]	73.8%	26.8%	46.4%	54.3%	NA

AUC: area under the curve; ROC: receiver operating characteristic; NA: not applicable.

In the study by Rezazadeh et al. [24], the authors evaluated the Barr, Leech, and Blethyn scores in children with abdominal pain and constipation and children with abdominal pain without constipation (constipation was defined as per the Rome III criteria). The mean age was 68.39 ± 33.48 months and 69.46 ± 32.60 months for the cases and controls, respectively. All three scores were found to be significantly higher in children with constipation and the sensitivity and diagnostic accuracy of the Leech score was found to be better than that of the Barr and Blethyn scores, as shown in Table 2.

Anwar ul Haq et al. [23] retrospectively collected data from their hospital electronic medical records on AXRs that were performed in children during a three-year period for different indications in the emergency department and the inpatient setting. Charts of children whose AXR reports contained specific constipation keywords ("constipation", "stool load", "fecal retention", and "fecal load") were reviewed to determine if a diagnosis of clinical constipation was made based on history and physical examination within 45 days after the AXR was performed. The sensitivity, specificity, NPV, and PPV were calculated based on the difference between the initial radiologic diagnosis of constipation and the later diagnosis of clinical constipation.

Risk of Bias and Applicability

Based on the six QUADAS-2 items applied to each of the articles, the two case–control studies were judged to be at unclear-to-low risk of bias, while the cohort study was judged to be at unclear-to-high risk of bias. Some applicability concerns were also raised in the article by Anwar ul Haq et al. [23] around the patient selection and reference standard domains. Table 3 illustrates the assessment of the risk of bias and applicability concerns.

Table 3. Risk of bias assessment and applicability concerns.

Study	Risk of Bias				Applicability Concerns		
	Patient Selection	Index Test	Reference Standard	Flow and Timing	Patient Selection	Index Test	Reference Standard
Marks et al. [22]	?	☺	?	☺	☺	☺	☺
Rezazadeh et al. [24]	?	?	☺	☺	☺	☺	☺
Anwar ul Haq et al. [23]	☺	☹	?	☹	?	☺	?

? Unclear risk; ☺ Low risk; ☹ High risk.

3. Methods

The protocol for the present systematic review was registered (PROSPERO ID: CRD42022301833). We followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) [25] guidelines and the research question was formulated in accordance with the Quality Assessment of Diagnostic Accuracy Studies 2 (QUADAS-2) [26] approach as follows: what is the diagnostic accuracy of abdominal X-ray (index test) in children and adolescents aged 2 to 18 years (patients) with a clinical diagnosis of functional constipation (reference standard) in different settings, including outpatient and inpatient settings, as well as the emergency department?

3.1. Eligibility Criteria

Studies eligible included those that assessed the diagnostic accuracy of abdominal X-ray for the evaluation of functional constipation in children. The study population included children 2 to 18 years old with a clinical diagnosis of constipation in whom AXR was performed. The radiologic parameters that were used to evaluate the AXRs had to be specified by the authors. The exclusion criteria included children with organic causes of constipation and children younger than two years of age.

3.2. Information Sources, Search Strategy, and Selection Criteria

A reference librarian searched the Medline, Embase, and Scopus databases to identify all potentially relevant articles that fit the pre-established eligibility criteria from 2012 (the year of the last systematic review) to May 2022. The full search strategy is outlined in Supplementary file. The search was limited to English literature only. Covidence systematic review management software (Covidence systematic review software, Veritas Health Innovation, Melbourne, Australia. Available at www.covidence.org, accessed on 1 June 2022) was used for data screening and extraction purposes. In addition, reference lists from selected papers were screened for relevant articles.

3.3. Data Collection

Two authors (DAR and GD) independently screened the eligible articles. Disagreements between investigators were resolved by consensus with a third investigator. Extracted data was inputted into a structured collection sheet by the same authors (DAR and GD) working independently.

3.4. Risk of Bias Assessment and Applicability Concerns

To assess the risk of bias and applicability concerns, two investigators (DAR and GD) working independently applied six of the QUADAS-2 items to each of the included articles. The QUADAS-2 tool [26] aids in the assessment of potential risk of bias and applicability concerns by applying some or all the items (signaling questions) that fit the research question chosen by the authors.

4. Discussion

Two previous systematic reviews investigated the utility of AXR in functional constipation [10,11]. The first systematic review [11] found a limited amount of data and conflicting evidence for the association between the clinical and radiographic diagnosis of constipation. This systematic review included six studies, of which only two were judged to have good methodological quality. The second and more recent systematic review published in 2012 [10] also found insufficient evidence, with high study heterogeneity for the study design, definition of constipation, and radiologic parameters used to evaluate the AXRs.

The present systematic review identified three studies published in the last 10 years that assessed the diagnostic accuracy of AXR for the evaluation of constipation in children. A quantitative analysis was not conducted due to clinical and methodological heterogeneity. The three included studies used different definitions of constipation and study designs, as well as different radiologic parameters, to evaluate the AXRs. Unfortunately, none of the

included studies recruited participants in a consecutive or random manner, which is the preferred method of patient selection for diagnostic accuracy studies [26].

Two of the included studies, using different radiologic parameters, found a diagnostic accuracy of 78–90% for AXR to detect constipation, and the calculated sensitivities and specificities ranged from 73–92% and 26–92%, respectively. The two studies in which specific radiologic criteria were applied (i.e., Marks et al. [22] and Rezazadeh et al. [24]) rather than only commenting on the overall stool loading appearance of the AXR were possibly better in detecting constipation. However, this is rarely performed in clinical practice as radiologists tend to interpret the overall "fecal loading" appearance of the AXR.

Marks et al. [22] found a model with a diagnostic accuracy of 87% to detect constipation in children who were successfully treated for the condition. However, it is important to mention that the treatments for constipation were not standardized in time or regimen and the cutoff values used in this study may not be generalized to other age groups whose intestinal size and volume may differ depending on age [27–29]. A recent prospective study [30] used the same radiologic parameters to assess AXRs and predict the risk of urinary tract infection in children who underwent an initial scout X-ray as part of their voiding cystourethrogram, but unfortunately, the radiologic score was not validated in the study.

Even though the Barr [31], Leech [32], and Blethyn [33] scores were found to have good diagnostic accuracy in one of the included studies [24], previous data show suboptimal sensitivities, specificities, and diagnostic accuracy for these scores [10,11,34]. A recent study of children who presented to the emergency department with abdominal pain and functional constipation (based on the Rome IV criteria) found perfect interobserver agreement (Kappa coefficient = 1.00) for the Leech score compared to a new scale devised by the authors ("faecal impaction grade by length percentage"), which had low interobserver agreement (Kappa coefficient = 0.133). An association between pain intensity and the radiographic grade of faecal impaction based on the Leech score was found to be statistically significant. It is important to mention, however, that the patients included in this study likely had severe constipation as indicated by the moderate-to-severe abdominal pain and therefore were more likely to have radiographic changes on the AXR [35]. In contrast, another recent study found poor interobserver agreement (Kappa coefficient = 36–48%) among radiologists, pediatric gastroenterologists, and pediatric emergency medicine physicians for the "subjective" interpretation of AXRs and poor (38.4%) and fair inter-observer agreement (60%) for the Blethyn and Barr scores, respectively. The Leech score was unfortunately not studied [36].

The study by Anwar ul Haq et al. [23] must be interpreted with caution given the unclear and high risk of bias assigned in three of the four QUADAS-2 domains, as well as the possible applicability concerns. The calculated sensitivity and specificity are likely inaccurate as it is unclear as to how many patients did or did not have a clinical diagnosis of constipation at the follow-up visit and therefore were not included in the final analysis. Also, patients might have received treatment for constipation in the interim between the radiologic diagnosis of constipation and the follow-up visit. Although not clearly stated in the methods, this study might have not excluded children with constipation of an organic nature; however, despite this, we decided to include this study.

Clinical practice guidelines discourage the use of AXR for the diagnostic workup of children with constipation due to its limited value, misleading nature, and radiation exposure [7–14]. AXR is an imperfect test for the assessment of constipation in children due to the absence of comparative radiologic normative data from children without constipation and the fact that symptoms may not correlate with the extent of faecal loading seen on the AXR or if other factors, such as air in the colon rather than the stool, may play a role in symptomatology. Children with abdominal pain who receive treatment for constipation in the emergency department are more likely to be admitted if an AXR is performed [37], and there is an increased risk of revisiting the emergency department with a clinically important alternate diagnosis (e.g., appendicitis, intestinal obstruction, or pneumonia) if

children diagnosed with constipation undergo an AXR [38]. The NASPGHAN [7], Rome IV criteria [6], and other literature [39] provide specific instances where AXR may be helpful. For example, children in whom faecal impaction is suspected but physical examination is not possible/unreliable [6]; those with poor or unreliable history; or when a rectal examination is considered traumatic [39].

There is evidence to suggest that other imaging modalities, such as abdominal ultrasound (including point-of-care ultrasound [POCUS]), may have a role in assessing for signs of constipation (e.g., rectal diameter and rectal anterior wall thickness). POCUS is increasingly being used in children by non-radiologists [40]; it is low-cost, non-invasive, and radiation-free [41]. Since the last systematic review [10], which found insufficient evidence for an association between clinical constipation and ultrasound findings, more research has been published in the last few years showing promising results [42–46]. These data need to be further evaluated to determine the role of abdominal ultrasound in functional constipation.

If an AXR is completed for the assessment of constipation due to any of the above reasons, rather than assessing the overall faecal loading appearance of the AXR, radiologists or clinicians should apply the Leech score, whenever possible, as this score appears to have better diagnostic accuracy based on very limited evidence.

5. Conclusions

There is insufficient evidence to support the use of AXR as part of the diagnostic workup of functional constipation. More methodologically rigorous studies are needed to determine the utility of AXR in the evaluation of functional constipation. The diagnosis of functional constipation should be based on history and clinical findings. AXR may have a role in specific clinical situations, such as when faecal impaction is suspected but physical examination is not possible/unreliable; in those with poor or unreliable history; or when a rectal examination is considered traumatic. If an AXR is completed, the Leech score may be more useful compared to other scores based on very limited evidence.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/gidisord6010006/s1.

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