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ORIGINAL RESEARCH

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Diagnostic accuracy of barium enema versus full-thickness rectal biopsy in children with clinically suspected Hirschsprung's disease: A comparative cross-sectional study

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Abstract

Background and Aims: Hirschsprung's disease (HSD) remains a common cause of pediatric intestinal obstruction. Barium contrast enema (BE) is the primary imaging modality for the evaluation of clinically suspected cases. Here, we aimed to assess the diagnostic accuracy of BE in children with clinically suspected HSD when compared to a gold standard full-thickness rectal biopsy (FTRB).

Methods: We recruited and consecutively enrolled children with clinically suspected HSD at two tertiary teaching hospitals. Participants underwent BE imaging and two radiologists interpreted the findings independently. Participants further underwent FTRB by pediatric surgeons as the confirmatory test. Sensitivity, specificity, positive predictive value (PPV), negative predictive value (NPV), and receiver operating characteristics (ROC) with the area under the curve (AUC) were calculated on Stata version 14.2, taking FTRB as the standard.

Results: We enrolled 55 cases, of which 49 completed the evaluation and were included in the final analysis. The median age was 9.4 months (interquartile range: 2–24], with a male-to-female ratio of 4.4:1. The sensitivity, specificity, PPV, and NPV of BE were 0.95 (95% confidence interval [CI] [0.81–0.99]), 0.73 (95% CI [0.39–0.94]), 0.92 (95% CI [0.82–0.97]), and 0.80 (95% CI [0.50–0.94]), respectively. On AUC, the diagnostic accuracy of BE compared to the confirmatory FTRB was 0.84 (95% CI [0.69–0.98]). The diagnostic accuracy was higher in neonates (ROC: 1.00) when compared to infants (ROC: 0.83) or those above 1 year of age (ROC: 0.798). HSD-suggestive BE findings were associated with absence of ganglion cells on FTRB ($\chi^2 = 23.301$, $p < 0.001$). Inverted rectosigmoid ratio and transition zone were more sensitive in detecting HSD of 0.92 (95% CI [0.74–0.98]) and 0.81 (95% CI [0.63–0.92]), respectively.

Conclusion: BE is sufficiently accurate in the diagnosis of children with HSD, suggesting BE would likely be used to inform surgical management in settings where confirmatory biopsy is lacking. However, clinical judgment is warranted in interpreting negative BE findings.

KEYWORDS

aganglionosis, barium enema, diagnostic accuracy, Ethiopia, Hirschsprung's disease, rectal biopsy

1 | INTRODUCTION

Hirschsprung's disease (HSD), a common cause of functional intestinal obstruction in children, is caused by congenital absence of ganglion cells in the distal bowel wall.[1–4] HSD is most commonly diagnosed in newborns with delayed passage of meconium or other signs of intestinal obstruction.[1,5] However, patients often present later in infancy and beyond with advanced disease and associated complications, particularly in low and middle income countries (LMICs).[4–6] Although early diagnosis and treatment are essential to prevent complications such as enterocolitis and colonic rupture,[1,4] diagnosing or ruling out HSD presents significant challenges for physicians practicing in LMICs.[4]

HSD is more common in boys than girls, albeit in familial forms where fair sex distribution is expected.[2,3,6] The cause is thought to be multifactorial, with familial cases associated with a range of genetic mutations identified to date, in addition to sporadic cases.[2] The aganglionic segment in HSD usually begins at the rectum, extends cranially and is associated with the severity of the manifestations.[1] In about 80% of cases, there is short segment disease that is limited to the rectosigmoid region of the colon; while in 3%–10% of cases, long segment disease that can affect the entire colon is observed. In rare cases, both the small and large intestines can be affected.[6] A separate entity that remains controversial is ultrashort segment disease, which extends 3–4 cm cranial to the internal anal sphincter.

Patients with clinically suspected HSD undergo a diagnostic evaluation that includes barium contrast enema (BE), anorectal manometry (ARM), and histological examination of rectal biopsy specimen.[4,7] BE is relatively safe for the diagnosis of HSD, yet its sensitivity (Sn) and specificity (Sp) are debated in the literature.[6] Its diagnostic utility as a single agent in settings without access to pathology is also understudied. The presence of a transition zone (TZ) with proximally dilated bowel, abnormal rectosigmoid ratio (less than 1), irregular contractions of the colon, mucosal irregularities, and retention of contrast medium on the film after voiding are the major findings indicative of HSD.[4] These can have an Sn and Sp between 40% and 80%.[4,8,9] Some signs, such as the presence of a transition point, may not be obvious in early presentations. Thus, studies question their relevance in infants under the age of 6 months. ARM is often unavailable in LMICs.[4,6] Although rectal suction biopsy is the preferred method for confirming diagnosis, full-thickness rectal biopsy (FTRB) is performed on patients of all age groups in LMICs due to the unavailability of the former.[10]

Many centers in LMICs lack the expertise and infrastructure needed for timely diagnosis of HSD patients.[5] The situation is further exacerbated by the insufficient knowledge of the patients' relatives and the limited availability of primary healthcare services. Consequently, late presentations with advanced and complicated cases are common.[5] As part of the developing world, Ethiopia is not exempt from this reality, despite recent expansions in pediatric health services.[11,12]

BE can play an important role in resource-constrained settings given the need for locally available diagnostic modalities close to the patient's place of residence to expedite diagnosis and initiation of treatment.[4,5,9] However, the lack of pediatric surgeons and pediatric anesthesia, which is limited to tertiary teaching hospitals, and the backlog of patients on the waiting list for a definitive surgical diagnosis or management represent major obstacles.[4] Pathology, especially fresh-frozen biopsy interpretation, is unavailable even in referral centers.[6,10] Thus, relying on relatively simple diagnostic tools with improved accuracy, mainly BE, remains a promising alternative. However, the existing body of literature focusing on the diagnostic precision of BE in HSD remains scarce. Therefore, this study aimed to evaluate the diagnostic accuracy of barium enema in HSD and its utility as the sole means of early diagnosis and treatment decision-making in resource-constrained settings.

2 | MATERIALS AND METHODS

2.1 | Study design and setting

Institution-based cross-sectional study was conducted at two tertiary teaching hospitals located in Addis Ababa, Ethiopia, namely, Tikur Anbessa Specialized Hospital and Menelik II Specialized Hospitals during the study period from September 2020 to October 2021. These are the largest centers affiliated with Addis Ababa University, Ethiopia. They have a high burden of pediatric surgical cases, with more than 2000 procedures being performed yearly.[10] All children with clinically suspected HSD in the specified time period were included in the study. Patients with complete evaluation with both BE and FTRB were appropriate and thus included in the final analysis.

2.2 | Inclusion and exclusion criteria

2.2.1 | Inclusion criteria

- Children under the age of 18.
- History of a delayed passage of meconium (beyond 48 h for a full-term neonate and beyond 72 h for a preterm neonate).
- Patients with defecation problems since birth with abdominal distension.
- Other evidence of obstruction such as severe constipation with no medical causes identified and no response to treatment.

2.2.2 | Exclusion criteria

- Abdominal radiograph showing multiple air fluid levels.
- Children with established alternative diagnosis.
- Patients suspected to have necrotizing enterocolitis.

The modified Bell staging criteria[13] with a composite of clinical signs and symptoms (e.g., abdominal distention, bloody stools, or hypotension), biochemical parameters (e.g., thrombocytopenia or neutropenia), and radiographic signs (e.g., pneumatosis or pneumoperitoneum) was used to exclude necrotizing enterocolitis.

2.3 | Data collection and procedures

After ruling out contraindications, patients with suspected HSD underwent a quality-controlled BE examination which was administered by senior radiology technologists in the respective facilities. The obtained images were interpreted by experienced consultant pediatric (S. S.) and general (Z. S.) radiologists with near perfect interrater agreement, Cohen's kappa coefficient of 0.83 (95% confidence interval [CI] [0.65–1.00]). The rectosigmoid index was calculated by dividing the widest diameter of the rectum below the third sacral vertebra by the largest diameter of the sigmoid colon; The measurements were taken along the transverse axis. A rectosigmoid index less than or equal to 1 was considered suggestive of HSD (a sigmoid colon wider than the rectum).[14] The subjective finding of a relatively narrowed aganglionic segment distal to a dilated normal colon on barium enema was considered a TZ.[14] Irregular contraction indicates denervation hyperspasticity of the distal segment with a sawtooth configuration. Mucosal irregularity was defined based on jejunization of the rectosigmoid colon. The presence of any of the radiological signs was considered a positive BE finding.

After obtaining informed consent, experienced pediatric surgeons performed an FTRB under general anesthesia. Rectal suction biopsies were not available at the health facilities. Samples were processed using hematoxylin and eosin to stain formalin-fixed, paraffin-embedded tissue sections at pathology departments of the centers with no histochemical staining done.

2.4 | Statistical analysis

A data collection questionnaire was prepared and used to extract patient data. Data collection was performed by two authors (A. K. B. and T. H.). The data were entered into Microsoft Excel, cleaned, and transferred to Stata version 14.2 (StataCorp) for statistical analysis. The results of the statistical analysis were presented numerically and graphically. Descriptive statistics were generated for sociodemographic variables, key clinical data, as well as imaging and histologic examination findings. The accuracy of BE findings in detecting FTRB-confirmed HSD was evaluated using Sn, Sp, positive predictive value (PPV), negative predictive value (NPV), and receiver operating characteristic analysis with area under the curve (AUC). Point estimates with 95% CIs were reported. All analyses were two-tailed with statistical significance declared at a p-value < 0.05.

2.5 | Ethical approval

This study was approved by the standing committee on research ethics of the Department of Radiology, College of Health Sciences, Addis Ababa University. Written informed consent was obtained from the patient's parents. All patients received standard care according to national guidelines, and those diagnosed with HSD received definitive surgical treatment where applicable. All procedures performed in this study were in accordance with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

3 | RESULTS

3.1 | Clinical presentation and diagnostic imaging and pathology results

A total of 55 cases were evaluated during the study period, and 49 cases, 34 (69%) males, had complete CE and FTRB results and were therefore included in the final analysis. The median age was 1 year for all patients and 9.4 months (interquartile range: 2–24) for those with biopsy-proven HSD. The minimum and maximum ages were 6 days and 10 years, respectively. Among biopsy-confirmed cases, neonates accounted for six (16%) of patients (Table 1).

Constipation was the predominant clinical symptom present in 35 (71%) cases, followed by abdominal distension in 32 (65%) cases, a history of delayed meconium passage in 18 (37%), and vomiting in 4 (8%) of patients, respectively.

The BE study was suggestive of HSD in 39 of the cases, of whom 35 were aganglionated, 3 were ganglionated, and 1 was inconclusive due to an inadequate sample. This case was dropped from subsequent association and diagnostic accuracy analyses. The remaining 10 BE studies were not suggestive of HSD. However, two (20%) of those biopsy specimens were found to be

aganglionated upon histopathologic examination, thus leading to a diagnosis of HSD (Figure 1).

There was a statistically significant association between the presence of one or more HSD-suggestive radiologic findings on BE and biopsy-confirmed cases ($\chi^2 = 23.301$, $p < 0.001$). The sex ratios diagnosed with HSD showed a significant difference between men and women ($\chi^2 = 13.107$, $p < 0.001$). This finding implies that there are variances in predisposition to HSD between men and women.

TABLE 1 Sociodemographics, test results, and their association with biopsy confirmed cases at two health facilities in Ethiopia.

Sociodemographics and test findings	*N* (49) (%) All patients	*N* (37) (%) Biopsy confirmed cases	χ^2	*p*[a]
Health facility				
Menilik II Hospital	28 (57.14)	20 (54.05)	1.216	0.270
TASH	21 (42.86)	17 (45.95)	-----	-----
Age category				
<28 days	7 (14.29)	6 (16.22)	1.093	0.579
28 days–1 year	18 (36.73)	14 (37.84)	-----	-----
>1 year	24 (48.98)	17 (45.95)	-----	-----
Sex				
Male	34 (69.39)	31 (83.78)	13.107	0.000
Female	15 (30.61)	6 (16.22)	-----	-----
BE findings				
HSD suggestive findings	39 (79.59)	35 (94.59)	23.301	0.000
HSD nonsuggestive findings	10 (20.41)	2 (5.41)	-----	-----

Abbreviations: BE, barium contrast enema; HSD, Hirschsprung's disease; *N*, frequency.

[a]*p* Values are expressed from Fischer's exact tests.

3.2 | Accuracy of barium enema findings in predicting biopsy results

HSD suggestive BE findings had an overall good Sn of 0.95 (95% CI [0.81–0.99]), and marginal Sp with a wide CI of 0.73 (95% CI [0.39–0.94]). The PPV achieved a diagnostic accuracy of 0.92 (0.82–0.97), while the NPV was relatively low at 0.80 (0.50–0.94). The diagnostic accuracy of BE compared to a confirmatory FTRB yielded an AUC of 0.84 (95% CI [0.69–0.98]) (Figure 2).

The diagnostic accuracy of BE was higher in neonates, estimated at 1.00, and comparable in infants, 0.8333 (0.507–1.000), and children beyond infancy, 0.798 (95% CI [0.601–0.996]) (Figure 3).

The inverted sigmoid index and TZ signs, depicted in Figures 4 and 5, had an Sn of 0.92 and 0.81, respectively, with an Sp of 0.71 and 0.86, respectively (Table 2). Therefore, there remains a chance for patients with either or both of the above findings to have ganglion cells visualized on biopsy (Figure 6).

The presence of irregular contraction and mucosal irregularity (Figure 7) had an Sp of 1 (95% CI [0.56–1.00]) but an Sn of 0.46 and 0.40, respectively.

4 | DISCUSSION

In this study, the median age of presentation for HSD-confirmed cases was 9.4 months, which was relatively early in comparison to a study from Tanzania reporting 2 years,[6] and an Ethiopian study reporting 19 months.[15] Patients in the neonatal age group comprised a higher percentage of diagnosed patients in comparison to the 5.5% reported previously.[6] Our findings were comparable to a review of 10,112 cases, which reported a median age of 1 year.[16] It is known that neonates with suggestive symptoms or intestinal obstruction are more likely to be explored without BE, thus artificially increasing the median age at diagnosis in some studies.[15] However, patients in our series were diagnosed at an advanced age when compared to reports from developed countries.[4] A recent study found that late diagnosis of HSD, defined as diagnosis at or over 1 year of age, was associated with more complications.[17] In the current study, 17 (45%) patients

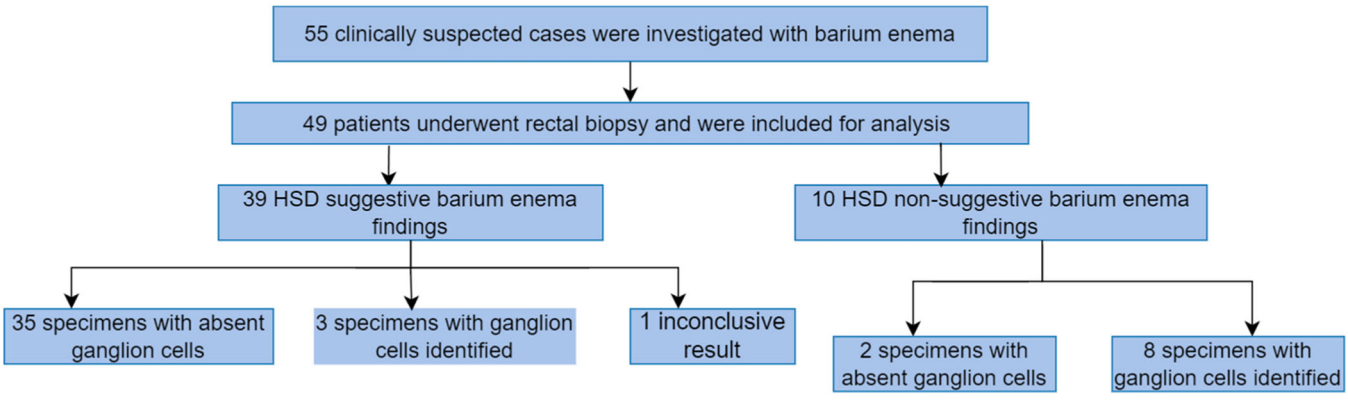

FIGURE 1 Flow chart for the diagnosis of Hirschsprung's disease (HSD) in children with clinically suspicious presentations.

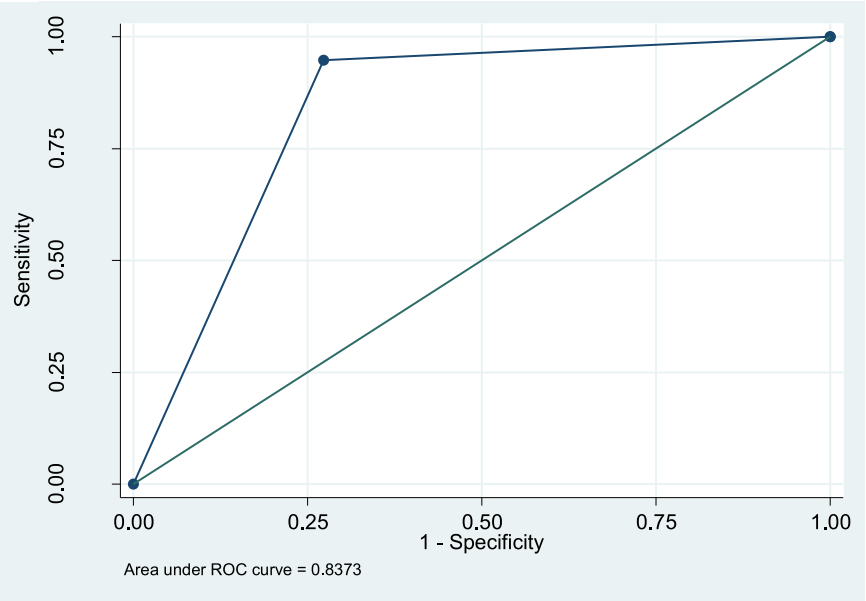

FIGURE 2 Receiver operating characteristics (ROC) curve analysis for test accuracy of suggestive barium contrast enema findings for the diagnosis of Hirschsprung's disease, taking full-thickness rectal biopsy as a gold standard The area under the ROC curve was 0.84 (95% confidence interval [0.69–0.98]).

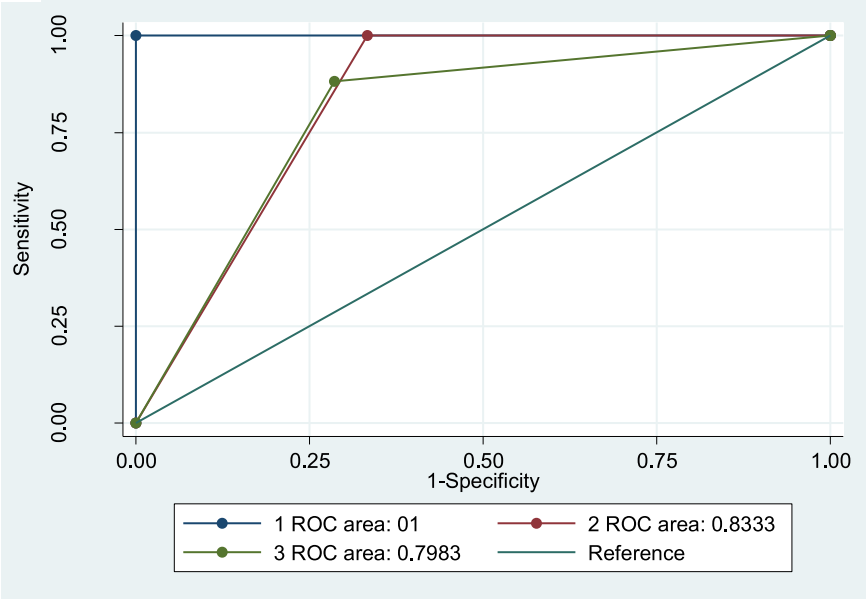

FIGURE 3 Receiver operating characteristic (ROC) curves of accuracy of barium contrast enema for a diagnosis of Hirschsprung's disease was determined for each age category: (1) up to 28 days, (2) 28 days–1 year, and (3) >1 year. The area under the ROC curve was higher in the first age group and comparable in the second and third age groups.

were diagnosed at more than a year of age, indicating the delayed presentation prevalent in our set-up relative to the high detection rate at a neonatal age in developed nations.[4] Male predominance was observed among diagnosed cases, which was in line with previous reports.[6,15,16]

The Sn, Sp, and accuracy of barium enema in the diagnosis of HSD in this study was 0.95 (0.81–0.99), 0.72 (0.39–0.94), and 0.84 (0.69–0.98), respectively, similar to reports from Indonesia and Taiwan.[9,18] The wider CIs can be attributed to the small sample size of our study. The diagnostic accuracy of BE was found to be high in neonatal age groups, approaching 100%. This is in sharp contrast to some studies that reported a lower Sn, Sp, and accuracy of BE, which was particularly lower in neonates.[8,15,19,20] One possible explanation for the observed difference could be that the previous studies did not include a proportional number of neonates, which may have biased the comparison. Another reason might be related to the common method of barium enema examination using 24-h delayed abdominal radiographs in studies reporting higher rates.[7,9] However, newborns were still underrepresented in our study, and we did not take a 24-h delayed BE film.

A recent study has found a comparatively high biopsy detection rate in patients under the age of 6 months when performed based only on clinical suspicion.[3] Thus, if BE can be done at a low clinical threshold and interpreted carefully, it can achieve a high diagnostic yield and increase the total number of cases diagnosed, obviating the need for a biopsy. This is important in LMICs with limited capacity for biopsy diagnosis. In addition, families may not accept a surgical consult for definitive diagnosis, as seen in six (11%) of cases in our

cohort.[5] On the other hand, in resource-rich settings, BE is an excellent initial investigation in HSD and can lead to a decrease in unnecessary rectal biopsies.[21]

The Sn of TZ of 0.81 was in agreement with previous studies.[5,22] Peyvasteh et al.[23] found relatively higher Sn and Sp rates of 0.9 and 0.8, respectively. A 24 h delayed film study from Pakistan found that TZ is the most sensitive radiological finding with an Sn rate of 0.91.[7] Esayias et al.,[15] however, have reported a very low Sn of TZ. There is conflicting literature over the diagnostic utility of TZ, with some reporting high Sn and Sp even in neonates.[23,24] Other authors have mentioned that the diagnosis of HSD becomes easier as the child gets older, since a TZ is more often visualized in the older child. However, a distal aganglionic segment passively dilated by impacted stool or a previously repeated cleansing enema may obscure the TZ in older children. The expertise of the radiologist reviewing the images, the specific imaging protocols used, and the extent of aganglionosis may be the most important factor in the discrepancy between studies.

FIGURE 4 A 3-month-old female infant with delayed passage of meconium. Barium contrast enema shows a dilated proximal colon and collapsed distal bowel segment with a clear transition point.

Regarding other BE radiologic signs, irregular contraction was found to have low Sn but higher Sp compared to previous studies, and this sign was absent in those without HSD in the current study.[7,23,24] Mucosal irregularity also had a low Sn and a higher Sp point estimate of 1.00. In contrast, this finding was in perfect agreement with the previous two studies.[23,24]

The Sn of the other radiologic signs of HSD, such as inverted rectosigmoid ratio, was high without compromised Sp. This radiologic sign was also found to be the most reliable sign in another study, having the highest Sn and Sp.[21] Inverted rectosigmoid ratio and TZ were the most sensitive radiologic signs in our study, which was in agreement with previous studies.[2,7,24]

Our study shows a high PPV of 0.92 (0.82–0.97) and a relatively lower NPV of 0.80 (0.50–0.94) for the totality of BE findings for the diagnosis of HSD. This finding suggests that a positive BE can be used to determine whether definitive surgical intervention is necessary. However, clinical judgment is warranted in negative studies, as 20% of patients with nonsuggestive BE findings were found to be aganglionated on biopsy. The reason for this finding can be considered multifactorial. First, some patients were well managed for constipation at presentation. Second, there were technical difficulties in conducting

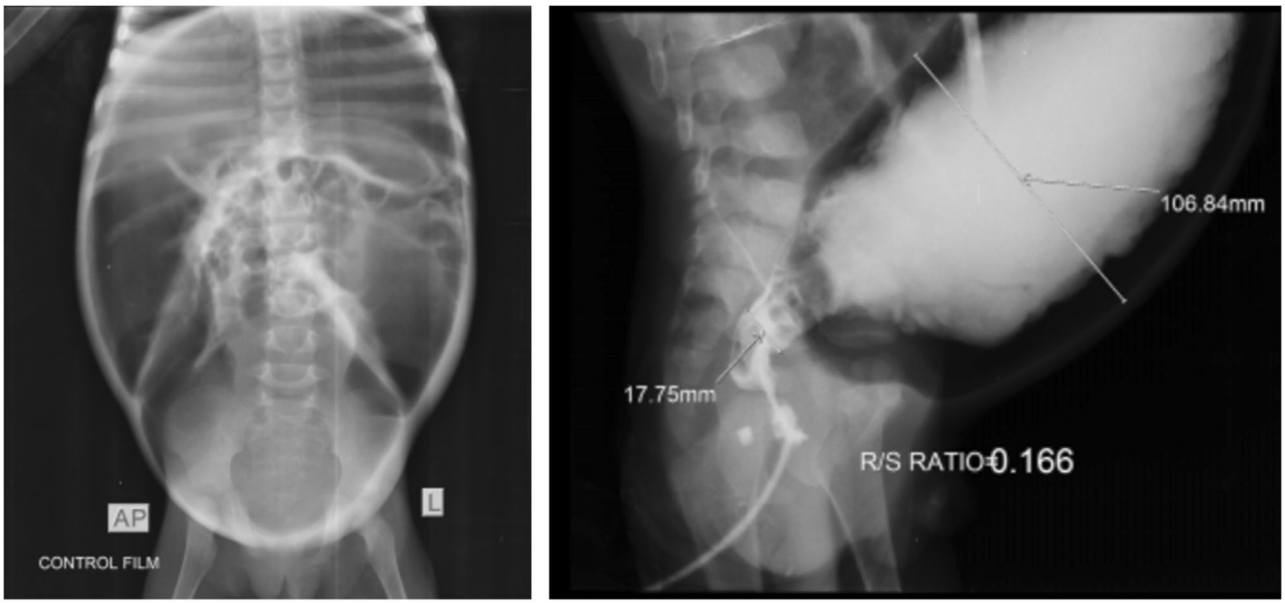

FIGURE 5 Biopsy confirmed Hirschsprung's disease in a 2-year-old male child presenting with suggestive clinical findings. Control film shows peripherally dilated bowel loops, and barium contrast enema revealed the presence of a transition point and a reversed rectosigmoid ratio.

TABLE 2 Sensitivity, specificity, and accuracy of BE diagnostic test, and individual imaging findings in reference to an FTRB diagnostic standard.

Barium enema findings[a]	Sn (%) (95% CI)	Sp (%) (95% CI)	PPV (%) (95% CI)	NPV (%) (95% CI)
Rectosigmoid index < 1 (37 of 48 cases)	91.66 (73.83–97.55)	71.43 (30.26–94.89)	93.55 (77.16–98.87)	62.50 (25.89–89.76)
Transition zone (33 of 48 cases)	81.25 (62.96–92.14)	85.71 (42.01–99.25)	96.30 (79.11–99.81)	50.00 (22.29–77.71)
Irregular contraction (22 of 48 cases)	45.83 (29.51–64.97)	100 (56.09–100.00)	100 (74.65–100.00)	29.17 (13.44–51.25)
Mucosal irregularity (19 of 48 cases)	39.58 (24.22–59.21)	100 (56.09–100.00)	100 (71.67–100.00)	26.92 (12.35–48.05)
Overall	94.59 (81.81–99.34)	72.73 (39.03–93.98)	92.11 (81.59–96.85)	80.00 (49.77–94.17)

Abbreviations: CI, confidence interval; FTRB, full-thickness rectal biopsy; NPV, negative predictive value; PPV, positive predictive value; Sn, sensitivity; Sp, specificity.

[a]The numbers represent total positive findings.

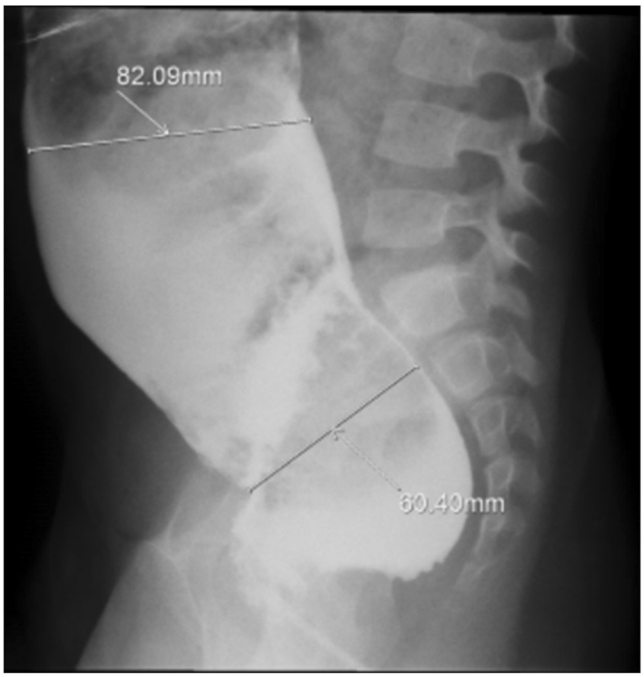

FIGURE 6 A 2-year- and 6-month-old child was investigated with barium contrast enema for suspicious symptoms. He had a reversed rectosigmoid ration but was ganglionated on biopsy.

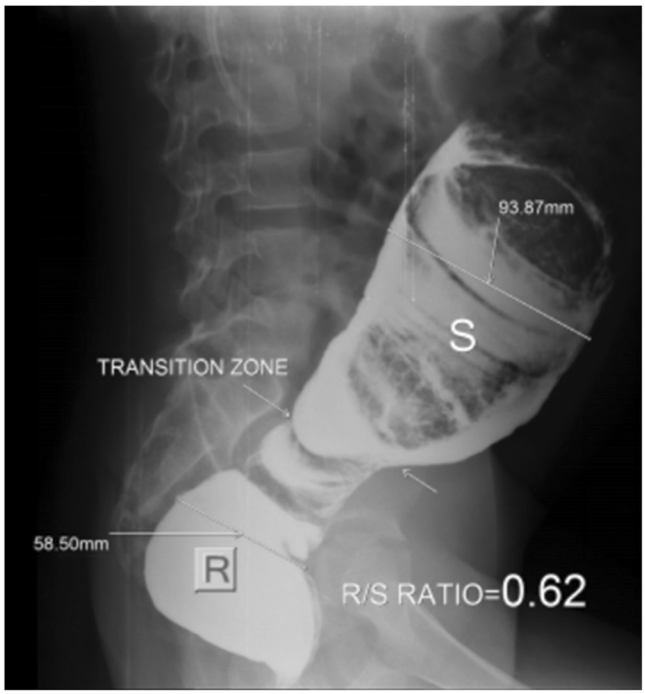

FIGURE 7 A biopsy confirmed Hirschsprung's disease in a 10-year-old female child. Barium contrast enema shows mucosal irregularity and contractions in addition to a reversed rectosigmoid ratio.

the test on irritable children. Finally, the expertise of the technicians and radiologists was also a factor, as barium enemas are not frequently performed in the centers. However, unlike previous studies, we did not encounter any missed long-segment disease.[15]

Considering the lack of pediatric surgical services in LMICs, the decision to provide treatment based on BE results may be prudent. However, diagnostic quality films should be acquired and interpreted by trained radiologists to reproduce the accuracy observed in our and other study contexts. We believe that teleradiology can play an important role in this situation and in the future; This could be an application of medical artificial intelligence in diagnostic imaging. The overall Sn of BE is high and Sp can be increased by careful exclusion of competing differential diagnoses. Cases in LMICs are typically delayed until after a failed medication trial for a number of possible causes and after overcoming many obstacles that prevent surgical consultation. Negative findings therefore require a careful approach. It is also noteworthy that expansion of pathology services is an essential part of quality care for HSD patients and should be planned.[4]

The study enrolled a limited cohort of patients and presented the experience of relatively affluent centers in terms of trained pediatric radiologists, pediatric surgeons and pathologists. This may be far from reality in many centers in LMICs, especially in rural areas. Twenty percent of patients with nonsuggestive BE findings were found to be aganglionated upon biopsy. This finding suggests the possibility of overlooking patients if we solely rely on imaging tests to establish a diagnosis. However, the study provided solid evidence that CE has good test accuracy in children and can be used to inform definitive

management when pathology services are unavailable. In addition, there are successful stories of surgical treatment of HSD without biopsy, such as endoanal pullthrough performed without a frozen section.[10]

5 | CONCLUSION

In sum, the accuracy of BE for the diagnosis of HSD is high and therefore it can be used as a primary diagnostic modality for HSD to inform definitive surgical management in areas where pathology services are not available. Reversed rectosigmoid ratio and TZ have high Sn whereas Irregular contraction and mucosal irregularity are the most specific findings. The overall Sn is high, and the Sp can be improved by careful exclusion of competing differentials. However, the NPV may not reach clinical significance.

AUTHOR CONTRIBUTIONS

Tesfahunegn Hailemariam: Conceptualization; data curation; formal analysis; investigation; methodology; resources; validation; writing—original draft. **Abenezer Kebede Bekele**: Conceptualization; data curation; formal analysis; investigation; methodology; resources; validation; writing—original draft. **Tsegahun Manyazewal**: Conceptualization; data curation; formal analysis; investigation; methodology; resources; validation; writing—original draft. **Daniel Zewdneh Solomon**: Methodology; validation; visualization; writing—review & editing. **Yocabel Gorfu**: Methodology; validation; visualization; writing—review & editing. **Zelalem Shiwarega**: Methodology; validation; visualization; writing—review & editing. **Tewodros Getinet**: Conceptualization; data curation; formal analysis; investigation; methodology; resources; validation; writing—original draft. **Meti Wole**: Conceptualization; data curation; formal analysis; investigation; resources; validation; writing—original draft. **Samrawit Solomon**: Conceptualization; data curation; formal analysis; investigation; methodology; resources; validation; writing—original draft. **Samuel Sisay Hailu**: Conceptualization; methodology; project administration; supervision; writing—review & editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data sets used and/or analyzed during the current study will be available from the corresponding author on reasonable request.

TRANSPARENCY STATEMENT

The lead author Samuel Sisay Hailu affirms that this manuscript is an honest, accurate, and transparent account of the study being reported; that no important aspects of the study have been omitted; and that any discrepancies from the study as planned (and, if relevant, registered) have been explained.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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