Pediatr Gastroenterol Hepatol Nutr. 2022 Jul;25(4):300-311
https://doi.org/10.5223/pghn.2022.25.4.300
pISSN 2234-8646·eISSN 2234-8840

Pediatric Gastroenterology, Hepatology & Nutrition
pghn

Original Article

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Role of Esophageal High-Resolution Manometry in Pediatric Patients

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OPEN ACCESS

Received: May 16, 2021
1st Revised: Oct 2, 2021
2nd Revised: Jan 23, 2022
Accepted: May 2, 2022
Published online: Jul 6, 2022

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Conflict of Interest
The authors have no financial conflicts of interest.

ABSTRACT

Purpose: Dysphagia, vomiting and feeding difficulties are common symptoms, with which children present. Esophageal function testing with high resolution manometry can help in diagnosing and treating these patients. We aim to access the clinical utility of high-resolution manometry of esophagus in symptomatic pediatric patients.
Methods: A retrospective chart review was done on all symptomatic patients who underwent esophageal high-resolution manometry between 2010 and 2019 at Sydney Children's Hospital, Australia. Manometry results were categorized based on Chicago classification. Demographic data, indication of procedure, manometric findings, and details of treatment changes were obtained and analyzed.
Results: There were 62 patients with median age of 10 years (9 months–18 years). The main indication for the procedure was dysphagia (56%). Thirty-two percent of patients had a co-morbid condition, with esophageal atresia accounting for 16%. The majority (77%) of patients had abnormal manometry which included, ineffective esophageal motility in 45.2%. In esophageal atresia cohort, esophageal pressurization was seen in 50%, aperistalsis in 40% and 10% with prior fundoplication had esophago-gastric junction obstruction. Patients with esophago-gastric junction obstruction or achalasia were treated by either pneumatic dilation or Heller's myotomy. Patients with ineffective esophageal motility and rumination were treated with a trial of prokinetics/dietary texture modification and diaphragmatic breathing.
Conclusion: Esophageal high-resolution manometry has a role in the evaluation of symptomatic pediatric patients. The majority of our patients had abnormal results which led to change in treatments, with either medication, surgery and/or feeding modification with resultant improvement in symptoms.

Keywords: Child; Dysphagia; Esophageal atresia; Manometry

INTRODUCTION

Children often present to pediatricians, gastroenterologists and surgeons with symptoms of dysphagia, chest pain, vomiting and feeding difficulties that may be caused by esophageal dysmotility. Esophageal dysmotility is common in neurologically impaired children and almost universal in children with repaired esophageal atresia (EA) due to congenitally and/

or surgically impaired neural innervation and musculature. Diagnostic investigations like contrast studies aim to exclude anatomical abnormalities of the esophagus like strictures, achalasia and hiatal hernia. Upper endoscopy helps to exclude eosinophilic and reflux esophagitis. However, data on motility, peristalsis and bolus transport in the esophagus is best elucidated using high-resolution manometry (HRM), ideally combined with impedance topography (HRIM) [1-4]. Where HRM/HRIM is not available the diagnosis may be delayed, or misdiagnosed, or rarely, the patient may even be referred to psychiatrists [5]. Therefore, the aim of this study was to determine the clinical utility of HRM and HRIM of esophagus in symptomatic pediatric patients.

MATERIALS AND METHODS

Subjects

This study retrospectively enrolled all pediatric patients who had undergone either esophageal HRM or HRIM between 2010 to 2019 at Sydney Children's Hospital, Randwick, for evaluation of symptoms of dysphagia, vomiting, non-cardiac chest pain, choking/gagging at meal times and for workup prior to fundoplication. We included all pediatric patients aged 0–18 years referred for high resolution esophageal manometry to our institution. We did not exclude any patient based on their age or underlying co-morbidity. Informed consents were obtained from patients and their parent. The study was approved by the Sydney Children's Hospitals Network Human Research Ethics Committee (approval no. LNR17/SCHN/60) and it conformed to the provisions of the Declaration of Helsinki in 1995 (as revised in Edinburgh 2000).

HRM and HRIM

Manometry Protocol: All patients fasted for at least 6 hours prior to HRM/HRIM. HRM catheters were used between 2010–2015, whereas HRIM catheters were utilized between 2016 and 2019. Sixteen patients (26.7%) had manometry catheter placement under direct vision at the end of an upper endoscopy procedure. This was mainly done in patients (median age, 6.5 years; range, 2–14 years) who were believed to be unstable to tolerate the catheter placement awake either, because of age or behavioral issues. In the remaining patients, the catheters were introduced transnasally and sensors were placed from above the upper esophageal sphincter (UES) to the esophago-gastric junction (EGJ) with at least two manometric sensor segments positioned in the stomach.

Stationary motility recordings in patients were performed according to the same protocol and with either a HRM or an HRIM, 8Fr solid-state catheter, which in the case of the HRIM catheter also incorporated impedance sensors in addition to the pressure sensors which the HRM catheter has. Topical anesthesia (2% lignocaine spray or gel) was used, and patients were studied sitting in a semi-reclined posture (patients were lying supine with their upper body raised 15–30° from horizontal on the examination bed). A stationary pull-through protocol was used as the standard to determine esophageal length, by determining swallow onset reliably by visualization of the UES high pressure zone (HPZ), the length of the EGJ HPZ and localization of the respiratory inversion point, defined as the location at which inspiratory pressure deflections changed from positive (abdomen) to negative (chest). Raw data was acquired at 20 Hz (Solar GI acquisition system; MMS, Enschede, The Netherlands). The bolus test protocol optimally included repeat administration (at >30-seconds intervals) of 10×5 mL liquid and 10×1 cm^2 solid (white bread over which butter was spread) swallows. If the parent mentioned that certain foods were more likely to result in symptoms, they

https://doi.org/10.5223/pghn.2022.25.4.300

were also trialed as part of the solid swallows. For the HRM studies water was used for the wet swallows. To ensure standardized bolus conductivity for HRIM studies, a commercially available bolus medium product conforming to the International Dysphagia Diet Standardization Initiative (IDDSI, Levels 1–4, Level 1=thin consistency) was used (SBMkit; Trisco Foods Pty Ltd, Brisbane, Australia). Use of a standardized bolus medium enables HRIM with pressure-impedance analysis and objective calculation of bolus transit time under stable bolus conductivity conditions. As part of protocol, provocative multiple rapid swallow (MRS) testing (rapid swallows of 5 mL×10) was also utilized. For MRS testing water was used for HRM and SBMkit for HRIM studies. Patients with a clinical suspicion of rumination spectrum disorder were also given a light sandwich meal and then underwent a period of extended monitoring for up to 1 hour after commencement of study. While the protocol is usually well tolerated the number of repeats may be titrated down (case by case). The minimum pediatric protocol for a diagnostic outcome was completion of 5×5 mL wet/liquid swallows. The duration of procedure usually less than an hour depended on cooperation of the patients. All the motility procedures were done by the same gastroenterologist and an experienced motility nurse practitioner. A child life therapist was always present during the study to play with the child which helped obtain patient cooperation and thereby increased the success rate for these procedures.

Tracings were analyzed with MMS automated analysis software version 9.5 (Laborie Medical Technologies Corporation, Portsmouth, NH, USA) and manometric diagnosis was made based on ten wet swallows, using the Chicago classification of esophageal Motility Disorders version 3.0 [6]. Patient with no abnormalities detected in the manometric study were classified as normal.

The following patient demographic data were obtained: age, sex, co-morbidity, route of feeding, current medications, indication for procedure. If patient was on acid suppressive therapy, this was continued but prokinetics (in 8 patients) were ceased at least a month prior to the procedure as per usual protocol in our hospital. Data was also collected on the results of other investigations that the patient had including contrast study, endoscopy and biopsy, pH-impedance testing and gastric emptying study were performed. Both reflux and eosinophilic esophagitis (EoE) was defined based on histopathological assessment of the biopsy specimens. If there was >15 eosinophils/HPF in the esophageal biopsy the patient was defined as having EoE [7].

Details of post procedure changes to treatment were obtained from patient medical records.

24 hours pH impedance

Combined multichannel intraluminal impedance and pH testing was considered to be abnormal based on criteria published by the German Pediatric Impedance Group (GPIG) [8].

Statistical analysis

A descriptive analysis of the population was carried out: quantitative values were presented as mean±standard deviation, median (25th and 75th percentiles) and qualitative values as numbers and percentages. Comparative analyses of the groups (patients with normal manometry vs. patients with abnormal manometry) were carried out using the Fisher exact tests for qualitative data and Student *t*-test for quantitative data. All test were univariate analysis and statistical significance was defined as $p<0.05$. The statistical analyses were performed with the statistical package SPSS Statistics 21 (IBM Co., Armonk, NY, USA).

https://doi.org/10.5223/pghn.2022.25.4.300

RESULTS

Demographic characteristics

A total of 62 patients had either HRM or HRIM at our institution during the study period. There were 38 female patients (61.3%). The median age of the study was 10 years, ranging from 9 months to 18 years. Nine patients were under 5 years of age. Eighteen patients had co-morbidities and EA was the most common co-morbidity in 10 (16.1%) of patients. The patient demographic and medication details are shown in **Table 1**. Dysphagia was main indication in 35 patients (56.4%). Indications for manometry are shown in **Fig. 1**.

Manometry results

Fourteen (22.6%) patients had normal findings and forty-eight patients (77.4%) had an abnormal result. The most common abnormality detected was ineffective esophageal motility (IEM) 45.2% followed by EGJ obstruction, aperistalsis and achalasia (**Table 2**). IEM was seen in 100% of patients with neurological impairment (NI). Abnormal manometry results were also seen in 100% of the EA patients. Thirty-three of the forty-eight patients (69%) with abnormal manometry results had treatment changes based on results of manometry (**Supplementary Table 1**). Three patients who had achalasia, two patients with Type 1 and Type 3 achalasia sub-types had Hellers myotomy done while the patient with Type 1 sub-

Table 1. Demographic characteristics

Characteristic	Study children (n=62)
Sex (Female/Male)	38 (61.3)/24 (38.7)
Age (yr)	10 (9 mo–18 yr)
Co-morbidities	18 (29.0)
Esophageal atresia	10 (16.1)
Neurological impairment	3 (4.8)
Celiac disease	2 (3.2)
Gastroparesis	2 (3.2)
Hirschsprung's disease	1 (1.6)
Prior fundoplication	17 (27.4)
Feeding route	
Oral	49 (79.0)
NG/NJ/PEG	13 (21.0)
Medication	
Nil	30 (48.4)
Proton pump inhibitor	20 (32.3)
Prokinetic	1 (1.6)
Proton pump inhibitor and Prokinetic	11 (17.7)

Data are presented as number (%) or median (interquartile ranges).
NG: nasogastric tube, NJ: nasojejunal tube, PEG: percutaneous endoscopic gastrostomy

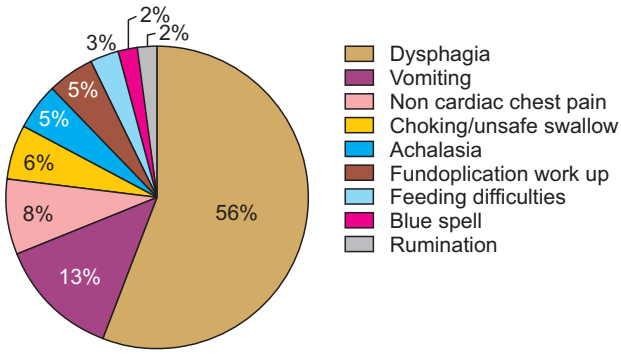

Fig. 1. Indication for procedure.

https://doi.org/10.5223/pghn.2022.25.4.300

Table 2. Results of HRIM/HRM

Manometry results	Study children (n=62)
Diagnosis	
Normal HRIM/HRM	14 (22.6)
Abnormal HRIM/HRM	48 (77.4)
Ineffective esophageal motility	28 (45.2)
Large peristaltic breaks	11 (39.3)
Small peristaltic breaks	17 (60.7)
Esophago-gastric junction obstruction	10 (16.1)
Aperistalsis	4 (6.5)
Achalasia	3 (4.8)
Unknown pressurization	2 (3.2)
Rumination	1 (1.6)

Data are presented as number (%).
Result of HRIM/HRM which categorized by Chicago classification.
HRM: high-resolution manometry, HRIM: high-resolution impedance manometry.

type had pneumatic dilation performed. Patients who had IEM and poor bolus clearance on impedance were treated with a trial of prokinetics and/or dietary texture modification. Of the 48 patients who had abnormal manometry results, seven patients also had abnormal results either in their pH-impedance testing [5] or endoscopy [2]. In these seven patients, the treatment changes were made based on the results of these other investigations rather than the manometry.

Esophageal atresia

In the EA cohort, there were seven female patients (70%) and the median age was 5.5 years (2 to 16 years). Nine patients had Type C, EA and one with a long gap defect had Type A, EA. Five patients (50%) had a history of prior fundoplication. Eight patients fed orally while two remaining had supplemental gastrostomy feeds in addition to oral feeds. Dysphagia was the most common indication (80%), while in the remaining two patients, the indication was a history of cyanotic/blue spells in one and suspected unsafe swallow with aspiration risk in the other. All EA patients had abnormal manometry, with esophageal pressurization seen in 50% [5], aperistalsis in 40% [4] and one patient (10%) with prior fundoplication had distal peristalsis with evidence of EGJ obstruction. Based on the Chicago classification, 5 (50%) of our EA patients had IEM with evidence of pressurization, four (40%) patients had absent contractility and one patient with prior history of fundoplication had EGJ obstruction. Interestingly one patient with Type C, EA had aperistalsis with wet swallow and distal peristalsis with solid swallows. Most of the EA patients were also evaluated with impedance (90%) and endoscopy (100%). There were abnormal results in 22% (2/9) of the impedance results and 20% (2/10) of the endoscopy results were abnormal. The two patients with abnormal endoscopy results had EoE on biopsy and were treated with Budesonide slurry for their EoE. Both of these patients with EoE in their biopsies had only partial improvement of their dysphagia post EoE treatment alone, hence they had HRIM which showed aperistalsis with incomplete bolus clearance. Subsequent to their HRIM, they were also treated with dietary texture modification based on their manometry results in addition to their EoE treatment, which resulted in symptomatic improvement of their dysphagia post this treatment change. Similarly, in the two patients with abnormal impedance results, showing elevated acid reflux index on impedance, but normal endoscopy and biopsies, their dysphagia persisted despite acid suppressive therapy with proton pump inhibitor (PPI). A subsequent HRIM to evaluate their dysphagia showed IEM with esophageal pressurization in one and absent contractility in the other. In these two patients' subsequent to the HRIM, a prokinetic Bethanechol was commenced based on manometry result, which resulted in their dysphagia

https://doi.org/10.5223/pghn.2022.25.4.300

improving post this treatment change. In the remaining 6 patients with normal impedance and endoscopy, the patient with EGJ obstruction had dilation of the fundoplication wrap and the remaining 5 patients with IEM were treated with diet modification and/or prokinetics (Bethanechol). All symptomatically improved post these treatment changes.

Results of other investigations

Impedance testing was performed in 44 (71.0%) patients and was abnormal in half (50.0%) of them. Gastric emptying studies were performed in 26 (41.9%) patients and was delayed in 34.6% of them. Sixty (96.8%) patients had an esophagogastroduodenoscopy that was normal in 38 (63.3%) and demonstrated reflux esophagitis in 9 patients (15.0%) and EoE (>15 eosinophils/HPF) in 3 other patients (5.0%) based on histology. Forty-two (67.7%) of patients had barium swallow and of those that had a swallow study, 17 (40.5%) had an abnormal study, details are given in **Table 3**.

Predictive factors for abnormal HRM/HRIM results

There was no significant association between a history of dysphagia, sex, age of the patient or prior fundoplication and the manometry result. There was also no significant association between having (dysphagia+abnormal barium result), (abnormal biopsy result and barium result) and (dysphagia+abnormal biopsy result) with the manometry results. All the children with either EA or NI had an abnormal manometry result and there was a significant association between having a co-morbidity of EA/NI and an abnormal manometry, p=0.029. In univariate analysis, there was no significant association between an abnormal manometry with results of barium swallow, impedance testing, gastric emptying or endoscopic biopsy. There was a trend towards lower acid reflux index in normal manometry group (1.98%) when compared to the abnormal group (5.61%), but this was not significant, p=0.08. Data is shown in **Table 4**.

Table 3. Impedance, radiologic and endoscopic data

Investigations	Study children (n=62)
Impedance	44 (71.0)
Normal	22 (50.0)
Abnormal	22 (50.0)
Gastric emptying time	26 (41.9)
Normal	17 (65.4)
Abnormal	9 (34.6)
Barium swallow	42 (67.7)
Normal	25 (59.5)
Abnormal	17 (40.5)
Dysmotility	10 (23.8)
Hold up mid esophagus	3 (7.1)
Achalasia	2 (4.8)
Hiatal Hernia	1 (2.4)
Hold up at EGJ	1 (2.4)
Endoscopy	60 (96.8)
Normal	38 (63.3)
Reflux esophagitis	9 (15.0)
Hiatal hernia	4 (6.7)
Eosinophilic esophagitis	3 (5.0)
Stricture	2 (3.3)
External compression by aberrant vessels	2 (3.3)
Tight LES with dilated esophagus	2 (3.3)

Data are presented as number (%).
Data of other investigations including impedance, radiologic, and endoscopic finding.
EGJ: esophago-gastric junction, LES: lower esophageal sphincter.

https://doi.org/10.5223/pghn.2022.25.4.300

Table 4. Predictive factors for abnormal HRM/HRIM results

Variable	Normal HRM/HRIM		Abnormal HRM/HRIM		*p*-value
Sex	9	5	29	19	>0.99
Co-morbid	3	11	17	31	0.52
Co-morbid EA/NI	0	14	13	35	0.02
Dysphagia	7	7	29	19	0.55
Barium swallow	8	1	17	16	0.06
Gastric emptying	6	3	11	6	>0.99
Biopsy results	10	3	31	16	0.52
Stricture	0	13	2	45	>0.99
Hiatal hernia	1	12	3	44	>0.99
pH-impedance	7	5	15	17	0.74
Prior fundoplication	4	10	13	35	>0.99

Data are presented in number and compare using Fisher exact which statistical significance are defined as $p<0.05$.
HRM: high-resolution manometry, HRIM: high-resolution impedance manometry, EA: esophageal atresia, NI: neurological impairment.

DISCUSSION

In recent years, children have often presented to pediatricians and gastroenterologists with symptoms of dysphagia, vomiting, chest pain and food intolerance, which can be due to gastroesophageal reflux disease (GERD), EoE, achalasia, strictures, rumination or functional in etiology [2,4,9]. Investigations such as barium swallow, endoscopy, pH-impedance testing and gastric emptying time are often done to evaluate these symptoms [1,2,10,11]. However sometimes the aforementioned investigations can be non-diagnostic, and if esophageal motor dysfunction is suspected, HRM with or without impedance can be performed to assess esophageal function and bolus transit. Although a manometry can be an invasive test and is not routinely available in all centers, it gives valuable information about esophageal function which is not available from any of the other tests. There is also sparse evidence in literature looking at the correlation between these other traditional testing modalities with esophageal manometry. While a barium swallow gives information about the swallowing mechanism and presence of anatomical abnormalities and an endoscopy gives information about esophageal mucosal pathology, neither of these are sensitive or specific tests of esophageal motor function when compared with HRM [12-16]. The majority of our patients had undergone these tests before they were referred for esophagus manometry. Currently, HRM is the gold standard for diagnosis of esophageal dysmotility [1,4,17-19]. HRM gives valuable information about esophageal peristalsis, integrity of contraction and EGJ relaxation pressure. These data have been well validated in adults using the Chicago classification. However, the Chicago classification [6] has not been validated in the pediatric population and normative values are also lacking. Some recent studies have tried to overcome this limitation by adjusting for age and size in children in order to improve diagnosis accuracy in children [4,9,17,20,21].

Our study describes the clinical utility of esophageal HRM in 62 symptomatic pediatric patients. The cohort size of our study is comparable to other pediatric studies which had between 40–271 patients. The predominant indication for manometry was dysphagia (56%). The referral indication for manometry in our study is similar to what has been described in the earlier studies in children [9,19,22], where dysphagia was the indication in between 18–66% of patients. However, a significant proportion of our cohort (16%) had EA as a co-morbid condition compared other studies where NI was the most commonly associated comorbidity.

https://doi.org/10.5223/pghn.2022.25.4.300

We used the Chicago classification to interpret data and majority of our manometry results were abnormal (77%), which is comparable to results from other pediatric studies using HRM/HRIM (54–90% of abnormal results) [9,19,22,23]. The most frequent esophageal motility disorder was IEM, affecting nearly half of the patients. This finding is similar to that of a recent study on 137 children in the USA and 271 children in France which showed IEM in 23% and 38% respectively [9,19]. Although IEM was the most common manometric abnormality detected in our study, the etiology of dysphagia was varied in our study, which showed IEM (51.4%), EGJ obstruction (11.4%), achalasia (5.7%), absent contractility and unknown pressurization in 5.7%. There was no significant correlation between a specific manometric abnormality and the presence of dysphagia. This highlights the importance of investigating dysphagia in children with an esophageal manometry in order to tailor treatment based on the specific abnormality detected.

In our study, 4.8% and 1.6% had achalasia and rumination syndrome respectively. A manometry is the gold standard to diagnose achalasia and rumination syndrome; without manometry testing, these patients could be misdiagnosed as having GERD or eating disorders and sometimes even referred for fundoplication due to poor response to PPI therapy [24-27]. In our center, achalasia patients were treated by either pneumatic dilation of the lower esophageal sphincter or Heller's myotomy. Patient with rumination were referred for diaphragmatic breathing.

Although other pediatric studies have evaluated the role of esophageal manometry in symptomatic children, ours is the first study which has reported the effect of treatment changes made based on manometry results on patient clinical outcomes. The majority of our cohort (97%) who had treatment changes made based on manometry results demonstrated symptomatic improvement.

Our pediatric study also had 16% of patients with EA. Esophageal dysmotility is universal in EA patients and the HRM findings can range from aperistalsis to distal peristalsis and esophageal pressurization [28-33]. In our EA cohort, an aperistalsis pattern was seen in 40%, pressurization in 50%, and distal peristalsis in 10%, compared to Lemoine study where these were seen in 38%, 15%, and 47%, respectively [29]. Although there have been previous studies evaluating symptomatic EA patients with manometry, ours was the first not only to confirm dysmotility with abnormal HRM/HRIM results in all EA, but also to report the effect of treatment change, including dietary texture modification, prokinetics and dilation of fundoplication wrap on patient clinical outcomes. At follow-up, all patients who had treatment changes instituted demonstrated significant symptomatic improvement. EA patients often have multifactorial etiology for their symptoms of dysphagia, vomiting and feed intolerance, including GERD, EoE, strictures, esophageal dysmotility and abnormal gastric function [28,30]. Therefore, it is important to diagnose the etiology of their symptoms accurately before instituting therapy. This is highlighted by the fact that in our cohort of 10 EA patients, two had treatment changes made based on their impedance testing and manometry and two others with EoE had treatment changes made based on their endoscopy and manometry, and in the remaining six patients, the treatment changes were made only from their manometry results, all with symptomatic improvement. We feel that the treatment changes made based on manometry results in addition to standard treatment of EoE and GERD contributed to additional symptomatic improvement. Our study underscores the importance of evaluating symptomatic EA patients with not only barium

swallow, endoscopy with biopsy and pH-impedance testing, but also HRM of the esophagus if the previous studies are non-diagnostic to optimize clinical outcomes.

All NI patients in our cohort were diagnosed with IEM and commenced on prokinetic medication. One of NI who was being considered for fundoplication had IEM, and due to high risk of dysphagia post fundoplication was recommended to have jejunal feeding, which showed a good response [34-36].

Our study has some limitations. Due to its retrospective methodology some of the follow up data was missing. However, we tried to overcome this limitation by doing a thorough search of electronic and paper medical records of the patients. Although observer bias was another limitation of this study, the effect of this was limited by the fact that there was only one gastroenterologist (observer) involved who did all the manometry procedures during this ten years period. The gastroenterologist who performed the procedure was not blinded to the patients' symptoms, or to treatment changes made based on result of the manometry, the manometry results were not influenced by any observer bias and the reported change in symptoms was based on what was reported by the parents and children during clinic visits. Another limitation was the fact that a validated symptom, quality of life (QOL) and dysphagia questionnaire was not administered at baseline at time of manometry and at clinic review post treatment changes being made. In addition, no repeat manometry was done post treatment change to determine if the symptom improvement correlated with manometric changes. We were able to ascertain the improved symptoms from information ascertained from patient records which contained details about history and symptom evaluation which was done during clinic visits which occurred at least a month post treatment changes being made based on manometry results. Although a high percentage (77.4%) of abnormal manometry results in our cohort could potentially be attributed to a selection bias with all EA and NI patients (21% of our cohort) having abnormal results, even in the remaining 49 normal children, an abnormal manometry was seen in 71.4%, highlighting the importance of motility testing not only in symptomatic children with a co-morbidity but also normal children with symptoms. For the EA patients, although a validated questionnaire was not used, we have developed our own detailed GERD/QOL symptom questionnaire which is administered to all these patients at the time of visit to the multidisciplinary EA clinic at our institution. Chicago classification version 3.0 was used to interpret our patients' data. We realize that the Chicago classification is based on adult normative data, which is a limitation of the study. However, in the absence of a pediatric classification for motility disorders, the Chicago classification is routinely used for pediatric manometry studies for children of all ages including those under 10 years of age in all the pediatric motility centres around the world including in ours. Studies have shown reliability of software-based Chicago Classification diagnosis of pediatric HRM recordings to be high overall [37].

In conclusion, HRM, ideally with impedance, has an important role in the evaluation of both esophageal and extra-esophageal symptoms encountered by the general pediatrician, gastroenterologist, and surgeon. Our study is one of the largest pediatric studies to date which evaluated the role of HRM of esophagus in the evaluation of symptomatic pediatric patients and in guiding the selection of optimal treatment. A majority of our study results were abnormal and treatment changes made based on manometry results resulted in symptomatic improvement and improved clinical outcomes in a majority of our cohort, validating the role of HRM/HRIM of the esophagus in symptomatic children. Future larger prospective studies need to be done to confirm our findings.

ACKNOWLEDGEMENTS

The authors acknowledge Dr. Steven T Leach PhD, School of Women's and Children's Health UNSW Medicine, Sydney Australia for his help with the statistical analysis of this study. In addition, we would like to thank Ms. Sam Ormond, a native English speaker with a BA in Communication Studies for her editorial assistance in improving the written English in this manuscript.

SUPPLEMENTARY MATERIAL

Supplementary Table 1
Abnormal manometry with treatment detail

Click here to view

REFERENCES

1. Nurko S. Gastrointestinal manometry: methodology and indication. In: Kleinman RE, Goulet OJ, Mieli-Vergani G, Sanderson IR, Sherman PM, Shneider BL, eds. Walker's pediatric gastrointestinal disease: pathophysiology, diagnosis, management. 6th ed. Raleigh: People's Medical Publishing House, 2018.
2. Chumpitazi B, Nurko S. Pediatric gastrointestinal motility disorders: challenges and a clinical update. Gastroenterol Hepatol (N Y) 2008;4:140-8.
PUBMED
3. Rommel N, Rayyan M, Scheerens C, Omari T. The potential benefits of applying recent advances in esophageal motility testing in patients with esophageal atresia. Front Pediatr 2017;5:137.
PUBMED | CROSSREF
4. Rommel N, Omari TI, Selleslagh M, Kritas S, Cock C, Rosan R, et al. High-resolution manometry combined with impedance measurements discriminates the cause of dysphagia in children. Eur J Pediatr 2015;174:1629-37.
PUBMED | CROSSREF
5. Hsing TY, Tsai IJ, Hsu CT, Wu JF. Role of esophageal manometry in children with refractory gastroesophageal reflux symptoms. Pediatr Int 2019;61:807-11.
PUBMED | CROSSREF
6. Kahrilas PJ, Bredenoord AJ, Fox M, Gyawali CP, Roman S, Smout AJ, et al. The Chicago Classification of esophageal motility disorders, v3.0. Neurogastroenterol Motil 2015;27:160-74.
PUBMED | CROSSREF
7. Papadopoulou A, Koletzko S, Heuschkel R, Dias JA, Allen KJ, Murch SH, et al. Management guidelines of eosinophilic esophagitis in childhood. J Pediatr Gastroenterol Nutr 2014;58:107-18.
PUBMED | CROSSREF
8. Pilic D, Fröhlich T, Nöh F, Pappas A, Schmidt-Choudhury A, Köhler H, et al. Detection of gastroesophageal reflux in children using combined multichannel intraluminal impedance and pH measurement: data from the German Pediatric Impedance Group. J Pediatr 2011;158:650-4.e1.
PUBMED | CROSSREF
9. Edeani F, Malik A, Kaul A. Characterization of esophageal motility disorders in children presenting with dysphagia using high-resolution manometry. Curr Gastroenterol Rep 2017;19:13.
PUBMED | CROSSREF
10. Gyawali CP, Bredenoord AJ, Conklin JL, Fox M, Pandolfino JE, Peters JH, et al. Evaluation of esophageal motor function in clinical practice. Neurogastroenterol Motil 2013;25:99-133.
PUBMED | CROSSREF
11. Zerbib F, Omari T. Oesophageal dysphagia: manifestations and diagnosis. Nat Rev Gastroenterol Hepatol 2015;12:322-31.
PUBMED | CROSSREF

12. Halland M, Ravi K, Barlow J, Arora A. Correlation between the radiological observation of isolated tertiary waves on an esophagram and findings on high-resolution esophageal manometry. Dis Esophagus 2016;29:22-6.
PUBMED | CROSSREF
13. von Arnim U, Kandulski A, Weigt J, Malfertheiner P. Correlation of high-resolution manometric findings with symptoms of dysphagia and endoscopic features in adults with eosinophilic esophagitis. Dig Dis 2017;35:472-7.
PUBMED | CROSSREF
14. Ott DJ, Richter JE, Chen YM, Wu WC, Gelfand DW, Castell DO. Esophageal radiography and manometry: correlation in 172 patients with dysphagia. AJR Am J Roentgenol 1987;149:307-11.
PUBMED | CROSSREF
15. Yamasaki T, Tomita T, Mori S, Takimoto M, Tamura A, Hara K, et al. Esophagography in patients with esophageal achalasia diagnosed with high-resolution esophageal manometry. J Neurogastroenterol Motil 2018;24:403-9.
PUBMED | CROSSREF
16. Schima W, Stacher G, Pokieser P, Uranitsch K, Nekahm D, Schober E, et al. Esophageal motor disorders: videofluoroscopic and manometric evaluation--prospective study in 88 symptomatic patients. Radiology 1992;185:487-91.
PUBMED | CROSSREF
17. Singendonk MM, Kritas S, Cock C, Ferris L, McCall L, Rommel N, et al. Applying the Chicago Classification criteria of esophageal motility to a pediatric cohort: effects of patient age and size. Neurogastroenterol Motil 2014;26:1333-41.
PUBMED | CROSSREF
18. Hong J. Clinical applications of gastrointestinal manometry in children. Pediatr Gastroenterol Hepatol Nutr 2014;17:23-30.
PUBMED | CROSSREF
19. Juzaud M, Lamblin MD, Fabre A, Alessandrini M, Baumstarck K, Bazin C, et al. Correlation between clinical signs and high-resolution manometry data in children. J Pediatr Gastroenterol Nutr 2019;68:642-7.
PUBMED | CROSSREF
20. Singendonk MMJ, Ferris LF, McCall L, Seiboth G, Lowe K, Moore D, et al. High-resolution esophageal manometry in pediatrics: effect of esophageal length on diagnostic measures. Neurogastroenterol Motil 2020;32:e13721.
PUBMED | CROSSREF
21. Goldani HA, Staiano A, Borrelli O, Thapar N, Lindley KJ. Pediatric esophageal high-resolution manometry: utility of a standardized protocol and size-adjusted pressure topography parameters. Am J Gastroenterol 2010;105:460-7.
PUBMED | CROSSREF
22. Staiano A, Boccia G, Miele E, Clouse RE. Segmental characteristics of oesophageal peristalsis in paediatric patients. Neurogastroenterol Motil 2008;20:19-26.
PUBMED | CROSSREF
23. Rosen JM, Lavenbarg T, Cocjin J, Hyman PE. Diffuse esophageal spasm in children referred for manometry. J Pediatr Gastroenterol Nutr 2013;56:436-8.
PUBMED | CROSSREF
24. Reas DL, Zipfel S, Rø Ø. Is it an eating disorder or achalasia or both? A literature review and diagnostic challenges. Eur Eat Disord Rev 2014;22:321-30.
PUBMED | CROSSREF
25. van Lennep M, van Wijk MP, Omari TIM, Benninga MA, Singendonk MMJ. Clinical management of pediatric achalasia. Expert Rev Gastroenterol Hepatol 2018;12:391-404.
PUBMED | CROSSREF
26. Hallal C, Kieling CO, Nunes DL, Ferreira CT, Peterson G, Barros SG, et al. Diagnosis, misdiagnosis, and associated diseases of achalasia in children and adolescents: a twelve-year single center experience. Pediatr Surg Int 2012;28:1211-7.
PUBMED | CROSSREF
27. Nikaki K, Rybak A, Nakagawa K, Rawat D, Yazaki E, Woodland P, et al. Rumination syndrome in children presenting with refractory gastroesophageal reflux symptoms. J Pediatr Gastroenterol Nutr 2020;70:330-5.
PUBMED | CROSSREF
28. Krishnan U, Mousa H, Dall'Oglio L, Homaira N, Rosen R, Faure C, et al. ESPGHAN-NASPGHAN guidelines for the evaluation and treatment of gastrointestinal and nutritional complications in children with esophageal atresia-tracheoesophageal fistula. J Pediatr Gastroenterol Nutr 2016;63:550-70.
PUBMED | CROSSREF

29. Lemoine C, Aspirot A, Le Henaff G, Piloquet H, Lévesque D, Faure C. Characterization of esophageal motility following esophageal atresia repair using high-resolution esophageal manometry. J Pediatr Gastroenterol Nutr 2013;56:609-14.
PUBMED | CROSSREF

30. Faure C, Righini Grunder F. Dysmotility in esophageal atresia: pathophysiology, characterization, and treatment. Front Pediatr 2017;5:130.
PUBMED | CROSSREF

31. Gottrand M, Michaud L, Sfeir R, Gottrand F. Motility, digestive and nutritional problems in Esophageal Atresia. Paediatr Respir Rev 2016;19:28-33.
PUBMED | CROSSREF

32. Tong S, Mallitt KA, Krishnan U. Evaluation of gastroesophageal reflux by combined multichannel intraluminal impedance and pH monitoring and esophageal motility patterns in children with esophageal atresia. Eur J Pediatr Surg 2016;26:322-31.
PUBMED | CROSSREF

33. Aspirot A, Faure C. Esophageal dysmotility: characterization and pathophysiology. Dis Esophagus 2013;26:405-9.
PUBMED | CROSSREF

34. Gockel I, Rabe SM, Niebisch S. Before and after esophageal surgery: which information is needed from the functional laboratory? Visc Med 2018;34:116-21.
PUBMED | CROSSREF

35. Loots C, van Herwaarden MY, Benninga MA, VanderZee DC, van Wijk MP, Omari TI. Gastroesophageal reflux, esophageal function, gastric emptying, and the relationship to dysphagia before and after antireflux surgery in children. J Pediatr 2013;162:566-73.e2.
PUBMED | CROSSREF

36. Gariepy CE, Mousa H. Clinical management of motility disorders in children. Semin Pediatr Surg 2009;18:224-38.
PUBMED | CROSSREF

37. Singendonk MM, Smits MJ, Heijting IE, van Wijk MP, Nurko S, Rosen R, et al. Inter- and intrarater reliability of the Chicago Classification in pediatric high-resolution esophageal manometry recordings. Neurogastroenterol Motil 2015;27:269-76.
PUBMED | CROSSREF