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Clinical Experience With Performing Esophageal Function Testing in Children

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ABSTRACT

Objectives: Pediatric high-resolution manometry (HRM) and 24-hour pH-impedance with/without ambulatory manometry (pH-MII+/-mano) tests are generally performed using adult-derived protocols. We aimed to assess the feasibility of these protocols in children, the occurrence of patient-related imperfections and their influence on test interpretability.

Methods: Esophageal function tests performed between 2015 and 2018 were retrospectively analyzed. All tests were subcategorized into uninterpretable or interpretable tests (regardless of occurrence of patient-related imperfections). For HRM, the following patient-related imperfections were scored: patient-related artefacts, multiple swallowing and/or inability to establish baseline characteristics. For pH-MII(+/-mano), incorrect symptom registration and/or premature catheter removal were scored. Results were compared between age-groups (0–3, 4–12, and >12 years).

Results: In total 106 HRM, 60 pH-MII, and 23 pH-MII-mano could be fully analyzed. Of these, 94.8% HRM, 91.9% pH-MII, and 95.7% pH-MII-mano were interpretable. Overall, HRM contained imperfections in 78.3% overall and in 8/8 (100%) in the youngest age group, 36/42 (85.7%) in 4 to 12 years and in 37/56 (66.1%) in children above 12 years; $P = 0.011$. These imperfections led to uninterpretable results in 4 HRM (3.8%), of which 3 were in the youngest age group (3/8, 37.5%). Imperfections were found in 10% of pH-MII and 17.4% of pH-MII-mano. These led to uninterpretable results in 5.0% and 4.3%, respectively. No age-effect was found.

Conclusions: Esophageal function tests in children are interpretable in more than 90% overall. In children under the age of 4 years, all patients had imperfect HRM and 3/8 tests were uninterpretable. HRM in older children and pH-MII+/-mano were interpretable in the vast majority.

Key Words: high-resolution manometry, pediatric esophageal function testing, pH-impedance

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What Is Known

- High-resolution manometry and 24-hour pH impedance measurements with and without ambulatory manometry in children are generally performed using adult(-derived) protocols as evidence-based pediatric protocols for the performance of these tests are lacking.

What Is New

- Despite occurrence of patient-related imperfections, nearly all 24-hour pH impedance measurements with and without ambulatory manometry and high-resolution manometry performed in children aged 4 years and older lead to interpretable results. In infants and toddlers, this decreases to approximately two-third of high-resolution manometry.
- Three-quarters of high-resolution manometry in children are imperfect because of occurrence of patient-related imperfections. All children <4 years and two-third of adolescents have imperfect measurements.

Stationary high-resolution manometry (HRM) and intraluminal pH-impedance monitoring (pH-MII) with or without ambulatory manometry (pH-MII-mano) can evaluate esophageal function and monitor gastroesophageal reflux (1). These tests can be used to diagnose gastroesophageal reflux disease, achalasia, rumination syndrome, aerophagia, and supragastric belching. As there are no well-established evidence-based pediatric protocols for these tests, they are generally performed according to adult protocols or adult-derived pediatric consensus-based protocols (2–4). This may, however, be complicated by several factors (5). First, discomfort and fear for the nasogastric catheters may lead to refusal or premature termination of the measurement. Second, younger children and infants may not be able to swallow on command as is required per HRM protocol. They may break up the bolus and perform multiple swallows with smaller volumes instead. This alters physiology, and may thus influence HRM-derived parameters and diagnosis (6). Third, objective symptom association during pH-MII may be difficult in infants and young children, where the accuracy of the symptom diary fully relies on proxy-report. Finally,

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analysis may be complicated by artefacts because of, for example, crying, gagging, coughing, and continuous movement (5).

Patients with achalasia have an increased resistance to flow at the esophagogastric junction. This may make placement of the catheter more difficult and can cause bolus stasis in the distal esophagus with a subsequent increase in symptoms. It is known that adult achalasia patients have more often imperfect HRMs compared with patients without achalasia (7). We hypothesized that HRM performed in children also show more imperfections when performed in (suspect) achalasia cases compared with patients without achalasia. Additionally, we hypothesized that pH-MII-mano measurements would fail more often compared with pH-MII, because of fear and refusal of an additional manometry catheter.

To evaluate how well these tests can be performed in children and how often they lead to interpretable test results, we assessed the occurrence of patient-related imperfections in different pediatric age groups, and their influence on test interpretability by performing a retrospective review of esophageal function tests performed at our center.

METHODS

Study Subjects

Data of patients (0–18 years) who had 1 or more esophageal function measurements (HRM, pH-MII, and/or pH-MII-mano) scheduled at the pediatric motility unit of the Emma Children's Hospital/Amsterdam UMC, location AMC between January 1, 2015 and December 31, 2018 were considered for inclusion in this retrospective cohort study. Patients and/or parents were sent an information brochure in which they were given the opportunity to object against the use of their data for this research project (opt-out procedure). Children were included and their measurements and medical charts were retrospectively reviewed when no objection was received within 6 weeks. Our study protocol was exempted from full ethical review by our local ethical review board because of its observational and retrospective nature (AMC Medical Ethical Review Committee, reference W19_337 # 19.397).

Study Design

HRM, pH-MII, and pH-MII-mano performed between 2015 and 2018 were retrospectively re-analyzed. Retrospective review of data and re-analysis of measurements was performed between October 2018 and February 2019. Catheter specifications, study protocols, and analysis software specifications are detailed in supplemental file 1, *http://links.lww.com/MPG/C57* and Supplementary file 2, *http://links.lww.com/MPG/C58*.

Categorization of Tests

All tests that were scheduled during the study period were retrieved from a clinical database.

These tests were categorized as: first time diagnostic tests; additional diagnostic- or follow-up tests; repeated tests because of previously unperformed or uninterpretable tests. If a child came in for the first time and multiple measurements were performed on the same day, all these were considered to be part of the first time (diagnostic) test.

Patient-related Imperfections

All performed measurements were re-analyzed for the occurrence of patient-related imperfections and for overall test interpretability. The different types of patient-related imperfections are defined below per test. All re-analyses were performed by the same investigator (M.L.). When interpretation was ambiguous, a second reviewer (M.vL.) was consulted. When no consensus was achieved, a third reviewer (J.O.) was consulted to adjudicate.

Subcategorization of Tests Based on Interpretability

A tests was called "*unperformed*" when it was never performed due to: "refusal of catheter" or "inability to position catheter through nasopharynx." All performed tests were subcategorized into *uninterpretable or interpretable* tests (regardless of occurrence of patient-related imperfections).

Measurements were classified as *failed* if they were *unperformed* or if they became *uninterpretable* because of *patient-related imperfections*.

Stationary High-resolution Manometry

In our center, HRMs are performed according to the Chicago Classification (CC) v3.0 protocol, with a small adjustment for its use in children [Supplemental File 1, *http://links.lww.com/MPG/C57* (3)]. In cases where HRM was only performed to establish the LES position relative to the nares, the CC protocol was not carried out and the study was exempted from analysis of patient-related imperfections. If the upper esophageal sphincter (UES) was not visible during HRM because of the use of a relatively short catheter for the size of the patient involved, measurements were also excluded from analysis of patient-related imperfections.

Analysis of stationary high-resolution manometry

First, HRMs were re-analyzed for the occurrence of any of the following *patient-related imperfections*:

1. inability to obtain baseline characteristics during a window of 10 seconds (see Supplemental file 1, *http://links.lww.com/MPG/C57*) before or after the test swallows;
2. double or multiple swallowing during a bolus swallow (ie, more than 1 swallow within 10 seconds of a bolus administration);
3. premature termination of CC protocol (ie, less than 10 boluses administered)
4. artefacts during bolus swallows, either caused by gagging, crying, coughing and/or belching.

All HRM studies were subsequently classified based on their overall interpretability:

Perfect measurement: 10 bolus swallows *without* any of above-mentioned patient-related imperfections.

Imperfect measurement(3): <10 *perfect* bolus swallows (3). We categorized imperfect measurements into 2 groups:

1. <10, but ≥7 *perfect* swallows *without* any of abovementioned patient-related imperfections, which is considered acceptable to identify a CC diagnosis in adults (7).
2. <7 *perfect* swallows.

Twenty-four Hour Studies (pH-impedance Monitoring and pH-impedance Monitoring With Ambulatory Manometry)

Measurements were classified as *imperfect* if they contained any of the following (4):

1. incorrect use of event button;
2. incomplete registration of symptoms, meals and/or body position (upright vs supine) in the diary;
3. premature termination because of catheter removal by patient.

Statistical Analysis

Descriptive analysis was performed using SPSS (IBM Statistical Package for the Social Sciences [SPSS] for Windows, v 25.0 Armonk, NY: IBM Corp). Data are shown as median and range.

Results were subdivided between 3 age groups: pre-school children, school children, and adolescents (0–3, 4–12, and 13–17 years). Subgroup analysis was performed in these 3 age groups and in patients with achalasia versus a group of age-matched patients without a diagnosis of achalasia. Additionally, differences between number of failed and imperfect pH-MII and pH-MII-mano were compared.

Chi-square and Fisher exact test were used to compare age-groups in terms of: percentage of failed measurements; percentage of imperfect measurements containing <10 but ≥ 7 perfect swallows; percentage of imperfect measurements containing <7 perfect swallows; percentage of overall adherence imperfections. A $P < 0.05$ was considered statistically significant.

Spearman correlation coefficient was used to calculate the correlation between age and the following: percentage of failed HRM and pH-MII; percentage of imperfect HRM and pH-MII; and number of perfect swallows performed. The strength of a (positive or negative) significant ($P < 0.05$) correlation was described using the following classification: 0.00 to 0.19 ‘‘very weak’’; 0.20 to 0.39 ‘‘weak’’; 0.40 to 0.59 ‘‘moderate’’; 0.60 to 0.79 ‘‘strong’’; 0.80 to 1.0 ‘‘very strong’’ (8).

RESULTS

Between January 2015 and December 2018, 123 children were referred to our motility center for esophageal motility testing. Parents of 1 patient objected against the use of its data.

A total of 122 children (52% boys, median age 12 [0–17] years) had 201 esophageal function tests scheduled: 116 HRM, 62 pH-MII, and 23 pH-MII-mano.

Categorization of Tests

One-hundred thirteen patients visited our motility center for a first time diagnostic test (102 HRM; 46 pH-MII, and 19 pH-MII-mano, see Table 1), 22 patients for *additional diagnostic- or follow-up tests* (13 HRM, 13 pH-MII, and 3 pH-MII-mano) and 5 patients had a *repeated test* after a previously failed test (1 water-perfused HRM, placed under anesthesia during esophagogastroduodenoscopy because of extreme fear; 3 pH-MII and 1 pH-MII-mano). Of all patients visiting our center for the first time, 9/113 (8%) had 1 or more failed measurements (see Table 1 for details). All 22 patients who came for an additional diagnostic- or follow-up test, had interpretable measurements.

One out of 5 children who had a repeated test because of a previously failed test, failed again (pH-MII failure because of premature catheter removal as a result of vomiting).

Stationary High-resolution Manometry

Of all 116 scheduled HRM, 110/116 (94.8%) were *interpretable*, regardless of patient-related imperfections. Two out of 116 (1.7%) scheduled measurements were *unperformed* (n = 1 refusal of catheter; n = 1 inability to position catheter). Out of 114 *performed* HRM, 8 (interpretable) measurements were exempted from analysis: 1 HRM was performed for determination of LES position only and no CC protocol was carried out. Seven measurements showed no UES visualization.

Of the remaining measurements, 83/106 (78.3%) contained *patient-related imperfections.* Seventy-six (71.7%) of these measurements became *imperfect* and 4/106 (3.8%) measurements were *uninterpretable* because of patient-related imperfections (see Table 2). There was no correlation between *failure* of HRM and age ($r = -0.070$; $P = 0.408$).

TABLE 1. Reasons for failed measurements in children visiting the motility center for the first time

First measurements all patients	0–3 y n = 16	4–12 y n = 45	13–18 y n = 52	Total n = 113
One or more failed tests, %	3 (18.8)	3 (6.7)	3 (5.8)	9 (8.0)
Patient related, %	3 (18.8)	2 (4.4)	2 (3.8)	7 (6.2)
Inability to position catheter, %	–	1 (2.2)	1 (1.9)	2 (1.8)
HRM	n = 7	n = 45	n = 50	n = 102
Failed, %	3 (42.8)	1 (2.2)	2 (4.0)	6 (5.9)
Refusal of catheter, %	–	1 (2.2)	–	1 (1.0)
Inability to position catheter, %	–	–	1 (2.0)	1 (1.0)
Patient-related artefact, %	3 (42.8)*	–	–	3 (2.9)
premature termination, %	–	–	1 (2.0)†	1 (1.0)
pH-MII (+/- mano)	n = 15	n = 22	n = 28	n = 65
Failed, %	1 (6.7)	2 (9.1)	1 (3.6)	4 (6.2)
Refusal of catheter, %	–	1 (4.5)‡	–	1 (1.5)
Inability to position catheter, %	–	1 (4.5)	–	1 (1.5)
Premature termination, %	1 (6.7)§	–	1 (3.6)‖	2 (3.1)

Patients could undergo 1 or more measurements during first visit. HRM = high-resolution manometry.
*All because of continuous crying.
†Because of fear.
‡Patient with autistic spectrum disorder.
§Because of self-removal (same patient continuously cried during HRM).
‖Because of vomiting.

TABLE 2. Patient-related imperfections leading to imperfect or failed measurements

High-resolution manometry (HRM)	0–3 y	4–12 y	13–17 y	Total	*P* value
Scheduled HRM, n	8	49	59	116	
All scheduled HRM that led to a meaningful test result	5 (62.5)	48 (97.6)	57 (96.6)	110 (94.8)	<0.001
Unperformed tests, n (%)	–	1 (2.0)	1 (1.7)	2 (1.7)	n/a
Refusal of catheter, n (%)	–	1 (2.0)	–	1 (0.9)	n/a
Inability to position catheter, n (%)	–	–	1 (1.7)	1 (0.9)	n/a
Performed HRM, n	8	48	58	114	
UES not visible because of catheter-related imperfection, n (%)	–	6 (12.5)	1 (1.7)	7 (6.1)	
Only LES determination, no CC protocol performed, n (%)	–	–	1 (1.7)	1 (0.9)	
Analyzed measurements, n	8	42	56	106	
Measurements with any patient-related imperfection, n (%)	8 (100.0)	38 (90.5)	37 (66.1)	83 (78.3)	0.011
No baseline established, n (%)	1 (12.5)	0 (0.0)	0 (0.0)	1 (0.9)	n/a
Double/multiple swallowing, n (%)	8 (100.0)	36 (85.7)	33 (58.9)	77 (72.6)	0.008
Patient-related artefacts, n (%)	5 (62.5)	9 (21.4)	6 (10.7)	20 (18.9)	ns
Imperfect measurements, n (%)	5 (62.5)	36 (85.7)	35 (62.5)	76 (71.7)	0.025
≥7 but <10 perfect swallows, n (%)	0 (0.0)	13 (30.9)	30 (53.6)	43 (40.6)	0.002
<7 perfect swallows, n (%)	5 (62.5)	23 (54.8)	5 (8.9)	33 (31.1)	<0.001
Uninterpretable because of imperfections, n (%)	3 (37.5)	0 (0.0)	1 (1.8)	4 (3.8)	n/a
Artefacts (crying) throughout measurement, n (%)	3 (37.5)	–	–	3 (2.8)	n/a
No bolus swallows performed, n (%)	–	–	1 (1.8)	1 (0.9)	n/a
Number of bolus swallows performed per test					
Median number bolus swallows per study [range]	8 [0–11]	10 [9–14]	10 [5–17]	10 [0–17]	<0.001
Median number perfect bolus swallows [range]	0 [0–3]	6 [0–10]	9 [0–10]	9 [0–10]	<0.001
24 hour pH-impedance measurement (pH-MII)	**0–3 y**	**4–12 y**	**13–17 y**	**Total**	***P* value**
Scheduled pH-MII	17	21	24	62	
All scheduled HRM that led to a meaningful test result	16 (94.1)	17 (80.9)	24 (100)	57 (91.9)	ns
Unperformed tests, n (%)	–	2 (9.5)	–	2 (3.2)	n/a
Irefusal of catheter, n (%)	–	1 (4.8)	–	1 (1.6)	n/a
Inability to position catheter, n (%)	–	1 (4.8)	–	1 (1.6)	n/a
Performed pH-MII	17	19	24	60	
Measurements with any patient-related imperfection, n (%)	2 (11.8)	2 (10.5)	2 (8.3)	6 (10.0)	ns
Imperfect measurements, n (%)	1 (5.8)	–	2 (8.3)	3 (5.0)	ns
incorrect event button use, n (%)	1 (5.8)	–	–	1 (1.7)	n/a
incorrect diary registration, n (%)	1 (5.8)	–	2 (8.3)	3 (5.0)	n/a
Uninterpretable due to premature catheter removal, n (%)	1 (5.8)	2 (10.5)	–	3 (5.0)	n/a
24 hour pH-impedance-manometry (pH-MII-mano)	**0–3 y**	**4–12 y**	**13–17 y**	**Total**	***P* value**
Scheduled and performed pH-MII-mano	1	8	14	23	
All pH-MII-mano that led to a meaningful test result	1 (100)	8 (100)	13 (92.9)	22 (95.7)	ns
Measurements with any patient-related imperfection, n (%)	–	1 (12.5)	3 (21.4)	4 (17.4)	n/a
Imperfect measurements, n (%)	–	1 (12.5)	2 (14.2)	3 (13.0)	n/a
Incorrect event button use, n (%)	–	1 (12.5)	1 (7.1)	2 (8.7)	n/a
Incorrect diary registration, n (%)	–	–	1 (7.1)	1 (4.3)	n/a
Uninterpretable because of premature catheter removal, n (%)	–	–	1 (7.1)	1 (4.3)	n/a

CC = Chicago Classification; UES = upper esophageal sphincter. One measurement can contain more than 1 adherence imperfection. *P* value (χ^2 or Fisher exact test in case of n <5) significance level <0.05; n/a = analysis not applicable <3 patients; ns = not significant.

Of 76 *imperfect tests,* 43 were imperfect as they contained <10 but ≥7 *perfect* swallows, whereas 33 contained <7 *perfect* swallows. Pre-school and school-children had significantly more *imperfect* HRM containing <7 *perfect* swallows compared with adolescents (100% vs 54.8% vs 9.1%, respectively; $P = < 0.001$). Number of perfect swallows performed had a strong correlation with age ($r = 0.623$). The number of *patient-related imperfections* had a very weak, negative correlation with age ($r = -0.289$; $P = 0.011$).

High-resolution Manometry in Patients With Achalasia

There were no differences in the number of *imperfect* or *failed* tests for achalasia versus nonachalasia age-matched controls (Table 3). Thirteen (46%) HRM measurements of patients with suspect achalasia versus 12 (43%) of control patients contained less than 7 perfect swallows.

Twenty-four Hour Studies (pH-impedance Monitoring and pH-impedance Monitoring With or Without Ambulatory Manometry)

A total of 62 pH-MII and 23 pH-were *interpretable*, regardless of patient-related imperfections. Two out of 62 (3.2%) scheduled pH-MII were *unperformed.* All scheduled pH-MII-mano were performed.

Patient-related imperfections occurred in 6/60 (10%) performed pH-MII versus 4/23 (17.4%) pH-MII-mano. These imperfections led to *imperfect tests* in 3/60 (5.0%) pH-MII versus 3/23 (13.0%) pH-MII-mano and to *uninterpretable tests in* 3/60 (5.0%) pH-MII and 1/23

TABLE 3. Age-matched comparison of high-resolution manometry outcome of achalasia versus nonachalasia patients

	Achalasia n = 28	No achalasia n = 28
Median age [range]	13 [8–17]	14 [6–17]
Uninterpretable measurements, %	0 (0.0)	0 (0.0)
Imperfect measurements n (%)	19 (67.9)	18 (64.3)
Patient-related imperfections n *(%)*	18 (64.3)	16 (57.1)
Double/multiple swallowing n (%)	18/27 (66.7)*	16/27 (59.3)*
No baseline established n (%)	0 (0.0)	0 (0.0)
Premature stop of measurement n (%)	0 (0.0)	0 (0.0)
Patient-related artefacts n (%)	4/27 (14.8)*	4/27 (14.8)*
Imperfections leading to <10 perfect swallows		
<10 but ≥7 perfect swallows (%)	6 (21.4)	6 (21.4)
<7 perfect swallows n (%)	13 (46.4)	12 (42.8)
Number of bolus swallows performed		
Median total no. of bolus swallows [range]	10 [10–12]	10 [1–14]
Median no. of *perfect* bolus swallows [range]	9 [3–10]	9 [0–10]

Differences between achalasia and nonachalasia group calculated using chi-square or Fisher exact test. $P < 0.05$ significant. None of the imperfections led to a significant difference.
*n = 1 not analyzable as UES was not visible.

(4.3%) pH-MII-mano, respectively. There was neither correlation between age and *uninterpretable measurements* nor with the occurrence of patient-related *imperfection*s ($r = -0.70$; $P = 0.179$ and $r = 0.78$; $P = 0.406$, respectively). There was no significant difference between percentage of *uninterpretable* or *imperfect* pH-MII versus pH-MII-mano ($P = 0.668$ and $P = 0.617$, respectively).

DISCUSSION

Our study shows that more than 90% of all esophageal motility tests performed (HRM, pH-MII, and pH-MII-mano) in the pediatric age led to a meaningful test result. Age was a factor in how well HRM could be performed but this effect was not seen in 24 hour studies (pH-MII or pH-MII-mano). No difference was found between patients with (suspected) achalasia and age-matched controls in terms of patient-related imperfections or failed HRM.

In 95% of scheduled HRM tests, a meaningful result could be generated. A large proportion of these tests (78%) nevertheless contained patient-related imperfections. Age was clearly a factor influencing these numbers. In fact, all HRM performed in infants and toddlers showed patient-related imperfections to some extent and in this age group, a meaningful result was generated in 63% only. Contrary to the youngest age group, tests of school children and adolescents were interpretable despite occurrence of patient-related imperfections in the majority of tests.

Roman et al (7) argued that a minimum of 7 well performed swallows, instead of 10 swallows recommended by the CC protocol, seemed reasonable to clinically interpret a HRM. When applying these criteria to our cohort, the percentage of *imperfect* HRM remained 100% in the youngest age group, but dropped from 53.6% to 8.9% in adolescents, which is similar to the percentage of imperfect tests in adults (7).

Although patient-related imperfections do not necessarily hamper HRM analysis (7), they may influence accuracy of the diagnosis. For example, infants and young children are unable to refrain from swallowing after a bolus swallow, leading to multiple closely spaced swallows (6). It is known that this ''piecemeal deglutition'' pattern alters pharyngeal high-resolution impedance manometry (HRIM) parameters in infants and young children (up to 4 years old) (6). Similarly, esophageal motility may alter as well during piecemeal deglutition. When using the adult CC criteria in HRMs containing double/multiple swallows, healthy subjects may incorrectly be diagnosed with ineffective- or absent esophageal motility, while in fact their motility is normal (3). These double/multiple swallows may hypothetically also hamper HRIM analysis in achalasia suspects (6).

We hypothesized that achalasia patients would have more imperfect tests because of an increased resistance to flow at the EGJ (7); however, this was not seen in our cohort. Achalasia patients often deal with long-term symptoms, and may therefore, be more cooperative and motivated to undergo a HRM, even if the circumstances are more difficult (9).

Our results implicate that a pediatric tailored protocol for performing HRM in young children is needed to improve feasibility and clinical utility. For example, piecemeal deglutition should be taken into account in such a protocol (6). Additionally, in patients with achalasia, pattern recognition is often sufficient to establish a diagnosis. This is also reflected in our cohort, almost half of achalasia suspects had 7 *perfect* swallows on HRM and still a diagnosis of achalasia could be made. Although strict criteria are not always met, a shorter pediatric protocol may be sufficient to establish a clinical diagnosis in some cases (11). Similarly, in esophageal atresia patients who all exhibit disordered motility, it may clinically be more relevant to describe the distinct patterns of the residual proportion of peristalsis (eg, absent-, distal, or a pressurization contraction pattern) as recently proposed, rather than to derive all HRM metrics and apply the CC algorithm (10). On the other hand, prolonged protocols for the use of HRIM might be of added value in specific cases (11,12). By administering additional liquid, semi-solid, and solid boluses of various volumes, the so called 'pressure flow analysis' can enhance understanding of the underlying motility problem and symptom etiology.

We hypothesized that pH-MII-mano measurements would fail more often compared with pH-MII, because of fear and refusal of an additional manometry catheter. In our study, however, none of the patients who had a pH-MII-mano scheduled refused the test and only 1 catheter was removed prematurely. This might partly be explained as our pH-MII-mano cohort was older than the pH-MII only cohort. On the other hand, placement of an additional catheter only takes a few extra seconds. Once these catheters are placed and secured patients may, contrary to our hypothesis, not experience any additional burden compared with pH-MII measurements.

Our study has several strengths. We managed to build a large cohort by including 122 patients over 4 years and our experienced pediatric motility team performed all measurements according to the current (adult or adult-derived) protocols, which enabled comparison between the tests. Limitations include the retrospective design of the study. Additionally, low incidence rates of rare diseases, such achalasia and small patient groups, such as preschool children led to small samples in subgroups. In a small minority of HRMs performed, the UES was not visible as a result of catheter-related (technical) imperfections. This made analysis of patient-related imperfections impossible in these children.

CONCLUSIONS

In summary, esophageal function tests in children were interpretable in more than 90%.

Patient-related imperfections occurred often in HRM and were more common in infants and young children. HRMs performed in (suspect) achalasia cases did not differ from those performed in controls without achalasia. In contrast to HRM, patient-related imperfections occurred in only a minority of 24 hour studies (pH-MII and PH-MII-mano). Therefore, we conclude that pH-MII and pH-MII-mano are well tolerated and can be performed in all age groups according to the currently used protocol. HRM is well tolerated in older children and adolescents but we believe that specific pediatric protocols could improve feasibility and test interpretability especially in infants and (young) children. Future studies assessing the effect of interventions by psychological or pedagogical staff, such as medical hypnotherapy or mental support before and during the measurement may further improve the performance of esophageal function tests in children.

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