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Quality outcomes for pediatric colorectal surgery treated during short-term international medical service trips at a dedicated site in Honduras

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A B S T R A C T

Introduction: Short-term international medical service trips (MSTs) provide specialized care in resource-constrained countries. There are limited data on immediate and long-term reported outcomes following specialty MST. We hypothesized that dedicated collaborative MST team and host institution produce outcomes and results comparable to those of high-income settings. Our primary aim was to analyze the long-term surgical and functional outcomes of our specialty-specific MSTs following five years of annual MST in Honduras.

Methods: We performed a single-institution retrospective analysis of 56 children who underwent colorectal and pelvic reconstructive operations between 2014 and 2018. Demographics, diagnosis, comorbidities, type of repair, long-term complications, and functional bowel and bladder results were recorded.

Results: We included a total of 56 children, 47 with ARM and 9 with HD, with a median age of 43.5 months (17–355) at the time of surgery. 25% (22) of the patients were lost to follow-up. Fecal continence was achieved by 23 (60%) patients <5 years who reported toilet training (n = 39) and by 5 (45%) patients >5 years (n = 11). Complications included constipation in 18 (42.9%) children with ARM and in 1 (12.5%) with HD. Eleven (19.6%) patients required revisional surgery for skin level anal stricture. Seventy-five percent of the patients with pediatric colorectal disorders attending the MST were compliant with continued long-term follow-up.

Conclusion: We were able to demonstrate that with organized, dedicated site and surgeon, results achieved can be comparable to those in the high-income countries (HICs). We conclude that this type of specialized care is feasible and beneficial for affected pediatric colorectal patients in resource-limited settings, when a strong partnership with a system of preoperative assessments and peri- and postoperative care can be established.

Level of evidence: Level IV (retrospective cohort study).

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In Latin-America, anorectal malformations (ARMs) and Hirschsprung disease (HD) represent approximately 4.1 cases in 10,000 births for ARMs and 1.86 cases per 10,000 births for HD [1,2]. In low- and lower-middle-income countries (LICs, LMICs), children with pediatric colorectal diseases often remain with an ostomy, as definitive care is delayed or only available to families with resources for additional surgery. This disparity creates a significant economic and social burden [3]. In 2008, Honduras, an LMIC, had eight registered pediatric surgeons to cover a population of 8 million patients (1:1,000,000), compared with 1:284,521 in the United States (US) and 1:108,640 in Europe [4,5].
There are limited data on immediate and long-term reported outcomes following specialty MST in LICs and LMICs. We previously demonstrated that MST specific to pediatric colorectal surgery improves the quality of care following the surgical group’s departure [6]. We hypothesized that a dedicated collaborative MST team and institution partnership produces outcomes and results comparable to those of high-income settings. Our primary aim was to analyze the long-term surgical and functional outcomes of a pediatric colorectal and pelvic reconstruction cohort operated on through a specialized MST following five consecutive years of MST in Honduras.

1. Methods

1.1. Study design, eligibility criteria, and collected data

A retrospective analysis of prospectively collected data of the patients who underwent pediatric colorectal and pelvic reconstruction surgery during the five missions held between 2014 and 2018 was performed. Inclusion criterion was pediatric patients who underwent surgery during the MST for anorectal malformation (ARM), cloaca, and Hirschsprung disease (HD). The study was approved by the Institutional Review Board of the Instituto Hondureno de Seguridad Social (IHSS, ACTA 064-CB-HE).

1.2. Study interventions

We reported the type of colorectal malformations (ARM, cloaca, HD), follow-up, associated comorbidities, type of surgical repair, short-term and long-term complications, and the functional outcome for bowel and bladder function. In addition to functional outcome, anesthetic related complications were obtained from the operative reports and patient records.

1.3. Short-term mission organization

Each MST is a week-long MST which was organized with the hospital “Ruth Paz” in San Pedro Sula, Honduras. Volunteer surgeons from different European and US institutions including surgeons of various disciplines (pediatric general colorectal and pelvic reconstruction surgery, pediatric general surgery, pediatric urologic surgery), surgical residents, and others took part in these MSTs. Surgeries were performed by the same local pediatric surgeon as well as the visiting MST surgeons.

1.4. Follow-up and MST daily organization

An annual program for surgical follow-up for all patients operated on during previous MST was organized. On the first day, all patients are seen for a preoperative visit and the follow-up of previously operated patients was performed. A typical operative day consisted of four operating rooms during the day, enuresis, and pain.

1.5. Surgical standardization and definitions

For the standardization of anorectal malformation (ARM) we used the Krickenbeck classification [8]. The operations were classified into posterior sagittal anorectoplasty (PSARP), or posterior sagittal anorectovaginourethroplasty (PSARVUP) for all patients with ARM. In patients with HD, we classified the operations into a Swenson-type transanal endorectal pull-through with or without laparotomy (Swenson-type-TERP, Swenson-type-L-TERP), Soave-type transanal endorectal pull-through with or without laparotomy (Soave-type-TERPT, Soave-type-L-TERPT), or laparoscopic-assisted Swenson- or Soave-type endorectal pull-through. The Soave-type pull-through was defined as the transanal approach with submucosal dissection, whereas the Swenson-type pull-through was defined as the transanal approach with a very short (< 0.5 cm) or nonmucosal dissection [9].

1.6. Assessment of bowel function

To compare the outcomes for bowel function, we used the Krockenbeck International classification for postoperative results [8]. The Voluntary Bowel Movement (VBM) classification consists of a three-part survey and is helpful for toilet-trained children > 5 years of age. Additionally, we subdivided results based on genders and ARM subtype, as these parameters are expected to have different results [10].

1.7. Perianal evaluation

Evaluation of the perianal area included an examination of cleanliness, redness, ulcers, the position of the neoanus and grade of mucosal prolapse. We also recorded the results of our anorectal examination in regard to the presence of stenosis, size of the anus, and perineal muscular contractions. We assessed the perineal muscular via physical examination in the clinic following spontaneous contractions, command or tactile stimulation. We did not use direct electrostimulation in the postoperative evaluation. Patients were interviewed regarding pain during evacuation, spontaneous abdominal pain, intermittent perianal pain, itching, and cramping. In addition, bowel function was evaluated regarding frequency and consistency of stool and the need for laxatives, antie- or retrograde enemas, skincare, and pro- and antibiotic usage.

1.8. Urologic assessment

The urologic outcome in these patients was evaluated by assessing the urological function as suggested by Versteeg et al. in spontaneous voiding, clean intermittent catheterization (CIC), diversion, incontinence, and kidney function [11,12]. Kidney function was assessed by levels of creatinine, and the estimated glomerular filtration rate was assessed using the creatinine-based “bedside Schwartz” equation from 2009 [13]. We also assessed the urinary frequency, urgency, ability to hold during the day, enuresis, and pain.

1.9. Statistical analysis

Analysis and statistics were performed with the cost-free Google®-Spreadsheet tool and the free XLMiner Analysis ToolPak add-on (© Frontline Systems Inc.) with blinded patient data and password-restricted user access containing Health Insurance Portability and Accountability Act (HIPAA) certification.

2. Results

2.1. Demographics

A total of 119 patients were seen and treated with the five years of MST, of which 56 had complete data for analysis. Exclusions included: patients pending colostomy closure (n = 10), treatment for non-ARM and non-HD disorders (n = 31), unable to contact (n = 19), and transfer of care (n = 3). Of those evaluated, the median patient age at the time of their follow-up was 3.6 years (1.4–29.6 years), with 25 (44.6%) female patients and 31 (55.4%) male patients (Table 1). In total 22 (25%) of the 88 ARM/HD patients treated during the 5 years MST were lost to follow-up.

Of the cohort analyzed, 47 patients had an ARM and 9 patients were treated for HD. Fifty-one of 56 (91.0%) patients presented with an ostomy. Forty-nine of 51 (96%) patients with ostomies had a colostomy; one patient in the HD group and one patient in the ARM group had an...
ileostomy. The median age for colostomy closure in these patients was 28 months (2–355 months) (n = 45). The five patients without an ostomy had a perineal fistula and the repair was performed without the creation of an ostomy. The mean length of stay for the repair of ARM or HD was 4 days (2–8 days) and for ostomy closure (n = 46) was 5 days (3–7 days).

### Table 1
Baseline demographics.

<table>
<thead>
<tr>
<th>Demographics</th>
<th>All patients (n = 88)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (years) (median, range)</td>
<td>3.6 (1.4–29.6)</td>
</tr>
<tr>
<td>Gender (n (%))</td>
<td></td>
</tr>
<tr>
<td>Female</td>
<td>25 (44.6%)</td>
</tr>
<tr>
<td>Male</td>
<td>31 (55.4%)</td>
</tr>
<tr>
<td>Loss of follow-up</td>
<td>22 (25%)</td>
</tr>
<tr>
<td>Patients analyzed for follow-up</td>
<td>56 (75%)</td>
</tr>
</tbody>
</table>

### Table 2
Voluntary bowel movements in all patients and separated into age groups.

<table>
<thead>
<tr>
<th>Type of ARM/HD</th>
<th>Total</th>
<th>VBM all age groups</th>
<th>VBM &lt; = 5 years</th>
<th>VBM &gt; 5 years</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Yes</td>
<td>No</td>
<td>Yes</td>
<td>No</td>
</tr>
<tr>
<td>No fistula</td>
<td>5</td>
<td>2</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td>Perineal fistula</td>
<td>6</td>
<td>4</td>
<td>2</td>
<td>3</td>
</tr>
<tr>
<td>Perineal fistula female</td>
<td>5</td>
<td>4</td>
<td>1</td>
<td>4</td>
</tr>
<tr>
<td>Vestibular fistula</td>
<td>9</td>
<td>4</td>
<td>5</td>
<td>3</td>
</tr>
<tr>
<td>Rectal atresia/stenosis</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>1</td>
</tr>
<tr>
<td>Bulbar</td>
<td>3</td>
<td>1</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Prostatic</td>
<td>6</td>
<td>3</td>
<td>3</td>
<td>2</td>
</tr>
<tr>
<td>Cloaca</td>
<td>6</td>
<td>2</td>
<td>4</td>
<td>1</td>
</tr>
<tr>
<td>ARM Other</td>
<td>1</td>
<td>1</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td><strong>Total ARM</strong></td>
<td>42</td>
<td>22</td>
<td>20</td>
<td>18</td>
</tr>
<tr>
<td><strong>Total HD</strong></td>
<td>5</td>
<td>6</td>
<td>2</td>
<td>5</td>
</tr>
<tr>
<td><strong>Total MST</strong></td>
<td>50</td>
<td>29</td>
<td>22</td>
<td>23</td>
</tr>
</tbody>
</table>

* Functional follow-up in patients without ostomy or after ostomy closure.

### 2.2. Associated anomalies

Associated anomalies were present in 18 of 47 patients (38.3%) of the ARM group. These associated anomalies included urogenital anomalies (n = 11), sacral anomalies (n = 3), craniofacial anomalies (n = 2), limb anomalies (n = 1), and chromosomal anomalies (trisomy 21) (n = 1). There were no associated anomalies in the HD group.

### 2.3. Perianal examination and muscular contractions

A perianal visual evaluation was performed in all 56 patients. At the time of examination, there were 35 patients (62.5%) clean and without feces on inspection, 14 patients (25%) had minor redness, and 3 (5.4%) had minor ulcers at the perineal area. Seven patients (12.5%, all from the ARM group) had a mucosal prolapse of less than 1 cm, in all cases on the left side of the neoanus. There was one patient (1.8%) with a complete full-thickness rectal prolapse. This patient had a rectobulbar fistula with a sacral anomaly.

Deliberate perianal muscular contractions by request were evaluated as ‘good’ in 39 patients (69.6%) and ‘intermediate’ in 15 patients (26.8%, 2 rectobulbar, 1 short common channel (CC) cloaca, 4 long CC cloaca, 2 fistula, 2 rectoprostatic fistulae, 3 vestibular fistulae, 1 HD after Swenson-type L-TERPT), and there were no deliberate contractions visualized in 2 patients (1 patient with long CC cloaca and 1 patient with rectoprostatic fistula).

### 2.4. Postsurgical anal stenosis/stricture or mucosal prolapse

Of the 56 total patients, 11 (19.6%) presented with anal stenosis or stricture. Four patients needed corrective surgery by Heineke–Mikulicz [14] stricturoplasty (n = 3) or minianoplasty (n = 1). The other 7 patients had a minor stenosis that were treated by anal dilations at home with donated Hegar dilators. All of these patients had an ARM. There was no anal or rectal stricture in the group of patients with HD. Minor mucosal prolapse occurred in 12.5% patients with ARM, none of which required operative correction.

### 2.5. Assessment of pelvic pain

Seven of 50 patients (14%) reported daily pelvic pain or discomfort during stool evacuation; 2 patients (4%) reported pelvic pain one to two times a week; 7 patients (14%) noted pelvic pain one to two times a month and 34 of 50 (68%) patients reported no pelvic pain during stool evacuation. Five of 50 (10%) patients reported having abdominal pain once daily; 1 (2%) patient had abdominal pain one to two times per month, and 35 (70%) patients were free of abdominal pain.
Two (4%) patients noted abdominal cramps one to two times a month, and 48 (96%) patients did not report abdominal cramps. Anal pain was noted by 8 (16%) patients once daily, one to two times a week (n = 3, 6%), and one to two times a month (n = 4, 8%).

2.6. Assessment of bowel function — voluntary bowel movements (VBMs), soiling and constipation

For the assessment of voluntary bowel movements (VBMs) ability, we included children with and without a history of ostomy (n = 50). The outcome was separated into patients <5 years (n = 39) or >5 years (n = 11) of age. In those <5 years, 23 (60%) patients had VBM. In patients >5 years, five (45.5%) patients had VBM (Table 2). No soiling was reported in 24 (48%). In the ARM group, there were no soiling in 22 (52.4%) patients, grade 1 soiling in 13 (31.0%), grade 2 soiling in 3 (7.14%), and grade 3 soiling in 4 (18.2%) patients. In the HD group, there were no soiling in 22 (52.38%) patients, grade 1 soiling in 13 (30.95%), grade 2 soiling in 3 (37.5%) patients, grade 2 soiling in 3 (37.5%) patients, and no patient (0%) with grade 3 soiling (Table 3). In the ARM group, 24 (57.14%) patients had no constipation, 1 (2.38%) had grade 1 constipation (treated with dietary modification), 15 (35.7%) had grade 2 constipation (treated with laxatives), and 2 (4.76%) had grade 3 constipation (resistant to dietary changes and laxatives). In the HD group, 7 (87.5%) patients had no constipation and 1 (12.5%) patient had grade 2 constipation (Table 4).

2.7. Bowel management — laxatives and enemas

Twenty-four (48%) patients were treated with laxatives, of which 21 (87.5%) received stimulative laxatives with senna or sodium-picosulfate (Sennax, Contumax), 3 used osmotic laxatives such as polyethylene glycol or lactulose (Miralax, Lactulax), and 2 patients with grade 3 constipation required enemas.

In total, seven patients (2 cloaca, 2 vestibular fistula, 1 prostatic fistula, 1 ARM without fistula and 1 unknown type of ARM) were administered enemas at home, with self-prepared saline solution. Enemas were administered at intervals of monthly (n = 3), daily retrograde rectal enema (n = 1), enemas one to three times a week (n = 1) and daily antegrade colonic enemas via a Malone-appendicostomy (n = 1).

2.8. Bladder and urinary function

For bladder function and urinary pattern, all patients were included. Forty-seven (83.9%) patients had spontaneous voiding and urinary continence. Six (10.7%) patients had a urinary diversion with a vescostomy (all cloaca). One (1.8%) patient with vescostomy reported spontaneous voiding in addition to the vescostomy. No patient performed CIC via the urethra. One patient performed CIC via the vescostomy orifice for complete emptying of the bladder.

2.9. Urinary incontinence

Urgency and incontinence were present in 26 (46.4%) patients. The frequency of incontinence was daily (n = 8, 14.3%), weekly (n = 7, 12.5%), two to three days each week (n = 9, 16.1%), and five days per week (n = 1, 1.8%). Urgency was present in 30 (53.6%) patients. Five of 11 (45.5%) patients >5 years (all patients had a cloacal malformation) noted being unable to hold urine during the day. Three of 11 (27.3%) patients had problems every day, one (9.1%) patient for 2 to 3 days per week, and one (9.1%) patient 1 day per week. Six of 11 (54.5%) patients older than 5 years had no problems with urgency. Creatinine and renal function were not obtained for the majority of the patients unless there were known urinary tract anomalies (n = 6) with measured creatinine level 0.53 mg/dL (± 0.09) and estimated glomerular filtration rate (eGFR) was calculated with 89.86 mL/min/1.73 m² (± 17.45) in this cohort.

2.10. Skincare and frequency of follow-up

Local skincare was necessary in 7 of 50 (14%) patients; 43 (86%) patients did not use local skincare ointments. All of them noted the use of homemade ointment for the perianal region. No patient used probiotics. No patient was treated with loperamide to reduce colonic motility.

Regarding frequency of follow-up with the local surgical team following the operation, 17 of 56 (30.4%) patients indicated one to two times per year; 33 (58.9%) patients three to six times per year; and 6 (10.7%) patients consulted them more than six times per year.

3. Discussion

This study presents our long-term results following 5 years of the consecutive site dedicated MSTs for pediatric colorectal patients with ARM and HD in Honduras, one of the most resource-constrained Latin American countries.

From these data, we can conclude that most patients in this disease group are compliant with follow-up and will embark on long trips and overcome difficult logistic and economic situations to be able to attend follow-ups. When a lack of feasible examinations under anesthesia exists, digital stimulation in the hands of an experienced surgeon can provide feedback on the function and location of the sphincter. Education is essential and can be implied from the good results obtained from simple bowel management regimens adapted to the local environment.

3.1. Comparison with previous literature

Surgical subspecialty MST appears to be a valuable approach for reducing the burden of surgical disease in countries with limited resources.
resources. However, long-term operative and functional outcomes, long-term host program setup and mechanism of patient follow-up are often unknown. Collaboration with the same host institution on our MST allowed multidisciplinary and adaptive collaborations. Visits to the same institution allowed for shared program goals, development and design [15,16].

Many studies have noted that the logistical challenges of locating former patients, varying degrees of patient compliance and coordinating follow-up with local professions have also been noted as barriers to follow-up [17–19]. Data on the immediate and short-term outcomes from these missions are limited [20–22]. Compared to the published 10-year follow-up data of a comparable group from an NIC-center (United Kingdom) with 53 patients, we achieved comparable results. The reported continence was 90% with the group of perineal fistula, 57% with vestibular fistula, and 58% with rectourethral fistula; soiling was present in 14% with vestibular fistula and 5% in rectourethral fistula; and grade 2 or 3 constipation was present in 21% with perineal fistula, 28% with vestibular fistula and 42% in rectourethral fistula [23]. Our follow-up showed VBM in 52.4% of all ARM patients and in 56.3% patients less than 5 years of age. Notably, in the group of children older than 5 years, half of the patients (50%) had a cloaca. Grade 3 soiling was present in 18.2% of our patients (3 of 4 patients had a cloaca). Grade 2 or 3 constipation was present in 40.5% in our ARM patients. Overall, our long-term follow-up rate was significantly better than those reported for most MSTs. This is likely because an established surgeon at the host institution provided seamless accessible ongoing follow-up at an establishment recognized by the families. The number of patients with stricture, soiling, and constipation and the perceived frequency of follow-up underline the importance of postoperative care systems and highlight the importance of ensuring follow-up care when doing this type of surgery. Our missions were also complemented by collaborative visiting and host institution nurses to deliver bowel management education while surgery was performed. In the review of guidelines and checklists for MST [24], our success was based on having the proper facilities for perioperative care and follow-up. There were additional nurses and a dedicated surgeon who follow-up with patients and addressed any complications or bowel management between MST. Institutional capacity and capability were also assessed both by the host surgeon as well between MST familiar with the site. For example, length of stay averaged 4.1 days (2–8 days) following ARM/HD repair and length of stay for ostomy closure was 5.0 days (3–7 days). These resources required for this length of inpatient stay during an MST should be clarified with the host institution. One of the greatest challenges is reliable histological results in patients with HD. In some cases, laparoscopic colonic mapping was performed and resulted in permanent pathology with a surgery performed the following year.

The perianal visual evaluation was performed during one visit at the follow-up within this study and therefore only represents the condition at that time of examination and is highly subjective in the observer interpretation. In addition, the majority of our patients were <5 years of age and not toilet-trained. Compliance with anal dilations following ARM repair was uncertain. However, routine dilations in the postoperative period have failed to demonstrate advantage over selective dilations [25]. This underscores the importance of postsurgical assessment.

As the outcome differs by type of ARM [26], we tried to separate the functional outcome according to subtype and gender. However, we were limited by age (the majority of our cohort was <5 years) as well as small sample size. The functional outcome regarding VBM, soiling, and constipation was comparable to published literature, although the follow-up criteria in many reports vary [23,27]. The poor total outcome regarding VBM in the group of patients older than 5 years might be explained by the high number of patients with cloaca belonging to the group of older patients. Patients with a cloaca were reported to have a less favorable functional outcome [11].

For the treatment of constant soiling, we suggested daily enemas for social continence. Bladder or rectal catheterization was also discussed. Enemas, however, carry a negative social stigma and parental fear that these would influence their children’s sexual development. Additional education regarding this misbelief is being provided. Of importance, the host institution teaching stoma-care techniques and bowel management regimens to local nurses and physicians will help aid continuity of care as well as allow postsurgical support. Following our departure, we remained in communication for questions regarding postoperative care, long-term follow-up, feedback and case assistance.

The results of this study may have potential implications for groups that are planning multidisciplinary or pediatric surgery specialty-specific MST and can be used to assist in the follow-up of patients long-term. The results can also be utilized to set up a single site with a host surgeon in order to improve care and long-term follow-up.

3.2. Limitations

There are several limitations to our study inherent to a retrospective cohort review. The resource-constrained environment and setting in Honduras and comparable LICs and LMICs make a complete preoperative functional and diagnostic assessment very difficult. We could not always measure the sacral-ratio owing to missing or poor-quality radiographs. No patient had preoperative pelvic magnetic resonance images or standardized radiographs for a complete evaluation of the spinal anatomy. Ultrasound of the kidneys and distal urinary tract was very infrequently performed preoperatively and had to be assessed at the time of the MST. The evaluation of the creatinine in all of our patients was not possible within our MST. We managed to evaluate those patients with known urogenital disorders, but formal urological laboratory and imaging-based annual evaluations were not completed. Finally, sexual function was not performed as our cohort was too young. Organizing ways to provide this additional information during continued patient follow-up and long-term management may provide a valuable addition.

4. Conclusion

We were able to demonstrate that with organized, dedicated site and surgeon, results achieved can be comparable to those in the high-income countries (HICs).

We conclude that this type of specialized care is feasible and beneficial for affected pediatric colorectal patients in resource-limited settings, when a strong partnership with a system of preoperative assessments and peri- and postoperative care can be established.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jpedsurg.2020.06.040.

References


