# Current management of Hirschsprung's disease in Egypt: a survey of members of the Egyptian Pediatric Surgical Association

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**Background/purpose** Significant progress has been made in the management of Hirschsprung's disease (HD). The choice of the management plan, surgical approach, and operative details is still variable among pediatric surgeons. This survey aims to determine the current preferences of Egyptian pediatric surgeons in the management of HD.

Materials and methods A survey was circulated individually to the members of the Egyptian Pediatric Surgical Association (EPSA) during the General Assembly Meeting. An electronic form of the survey was sent by e-mail to all Egyptian consultant pediatric surgeons registered to EPSA through the 'EPSA online' e-mail group. A second round of e-mails was sent 2 weeks later.

**Results** Responses were received from 112 surgeons; seven responses were excluded (incomplete and duplicate responses), yielding 105 survey charts for analysis. The 105 responses represent 80.7% of the 130 fully trained pediatric surgeons currently working in Egypt. A total of 76.2% of responders use both contrast enema and rectal biopsy for the preoperative diagnosis of HD. Contrast enema alone is used by 13 (12.4%). A further 11.4% combine this with anorectal manometry. Intraoperative frozen section tissue diagnosis is used routinely by only 4.8% of the responders. Surgery is preferred to be performed during the neonatal period by 21.9% of the responders, from 1 to 3 months (28.6%), or after the third month of life (49.5%). A definitive one-stage colonic pull-

# Introduction

Since Swenson [1] first described a surgical intervention for Hirschsprung's disease (HD) in 1948, considerable progress has been made in its management. Operative procedures are now less invasive and are often performed in a single stage [2–5]. The major decision for surgeons appears to be whether to resect the rectum and perform a coloanal anastomosis or bring the ganglionic segment into the posterior aganglionic rectum (Duhamel). Those performing a coloanal anastomosis have many choices on how to handle the muscular cuff of the distal rectum. Initially, these operations were performed in two or three stages depending on whether a colostomy was used to protect the anorectal anastomosis [6–8].

During the past 20 years, there have been reports of satisfactory results after a single-stage primary pull-through for HD. One-stage pull-through has been noted to be particularly beneficial in infants [9]. Some published series suggest earlier primary definitive surgery over multiple stages [10] with increased utilization of laparoscopic [11,12] and transanal approaches [3,13].

through is always used by 59%, whereas 7.6% are still using staged operations. The rest of the responders had no specific predilection. The preferred surgical technique for rectosigmoid HD is transanal endorectal pull-through (68.5%), Soave transabdominal endorectal pull-through (19.0%), laparoscopic-assisted pull-through (9.5%), and Duhamel with GIA staplers (2.9%). The Soave operation is preferred for HD extending to the right side and total colonic HD (45.7%) and redo surgery (37.1%).

**Conclusion** The EPSA survey indicated that most surgeons are moving toward a one-stage pull-through using transanal endorectal pull-through for patients with rectosigmoid HD. There are large variations in preference for the age at pull-through. There is a need for a routine practice of intraoperative frozen section tissue diagnosis, and popularizing laparoscopic surgery for patients with long segment disease. *Ann Pediatr Surg* 11:207–212 © 2015 Annals of Pediatric Surgery.

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The aim of this study is to determine the current preferences of Egyptian pediatric surgeons in the management of HD.

#### Materials and methods

After receiving approval from the Ethical committee of Tanta Faculty of Medicine, a 21-question survey was administered individually to the members of the Egyptian Pediatric Surgical Association (EPSA) during the general assembly meeting held on May 2014 in Cairo (Appendix 1). E-mails were also sent to all Egyptian consultant pediatric surgeons registered to EPSA through the EPSA e-mail discussion forum. A second round of e-mails was sent 2 weeks later with the questionnaire attached.

Questions were designed to cover the key elements of the management of patients with HD in Egypt, such as number of patients managed yearly, methods for preoperative diagnosis, preferred technique for biopsy, type of surgical intervention, use of intraoperative frozen section, age at the time of definitive surgery, type of anastomosis, extent of aganglionic segment resection,

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number of operative stages, length of the muscular rectal cuff, and other technical variables. Other questions included the operative technique for long segment, ultra-short segment, and total colonic aganglionosis, and redo surgery. Data were collected and analyzed using descriptive statistics (mean, median, and range) in predefined subgroups according to the options for each question.

### Results

Responses were received from 112 surgeons; seven responses were excluded (incomplete and duplicate responses), yielding 105 survey charts for analysis. All the responders are involved in the routine management of patients with HD, with 58.1% of responders performing definitive surgery for more than 10 cases per year.

#### **Diagnosis of HD**

Contrast enema alone is used by 13 of 105 (12.4%) responders to diagnose HD. Combined contrast enema and rectal biopsy was used by 76.2% of the responders. A further 11.4% of the responders combine this with anorectal manometry.

Partial rectal biopsy that includes mucosa, submucosa, and only part of the muscle layer was performed by either suction rectal biopsy (9.5% of the responders) or snip biopsy forceps or other techniques (44.8%). A full-thickness biopsy is performed under anesthesia; a specimen that included all layers was preferred by 45.7% of the responders.

#### Timing of definitive surgery

In a generally well-stable neonate with left-sided HD, the definitive surgery is planned in the neonatal period by 21.9% of the responders, 28.6% would operate on an infant 1 to 3 months of age, and a further 49.5% postpone surgery until the child is older than 3 months.

#### Choice of definitive surgery

A definitive one-stage colonic pull-through is always used by 59.0%, whereas 7.6% are still using staged operations. The rest, 33.4%, have no specific predilection.

In terms of the optimal operative procedures, transanal endorectal pull-through (TEPT) is performed by 68.5% of responders, Soave transabdominal endorectal pullthrough is still practiced by 19.0%, laparoscopic-assisted pull-through is preferred by 9.5%, and Duhamel with GIA staplers is used by 2.9% of the responders (Table 1).

### **Technical surgical details**

The Lone Star retractor (Cooper Surgical Inc., Trumbull, Connecticut, USA) is used for anorectal exposure and dissection by 17.1% of responders, whereas 19.0% are using it in conjunction with sutures and about 1.9% are using other retractors. Sutures only are used for retraction by 61.9% of the responders.

The point of initiation of anorectal dissection and coloanal anastomosis is less than 1 cm above the top of the anal columns (7.6% of responders); 73.4% of the responders start the anorectal dissection 1-2 cm above

the anal columns, whereas the remaining 19.0% of responders start more than anorectal higher than 2 cm.

Handling of the muscular cuff by surgeons is variable. The majority (55.6%) incise the cuff anteriorly to prevent later stenosis and recurrence of symptoms, 41.7% of the responders shorten the cuff circumferentially to less than 5 cm, whereas 2.7% leave it as it is.

There was a difference in the length of the ganglionated bowel excised proximal to the transition zone; 27.6% of responders excise less than 5 cm, 56.2% excise 5-10 cm, whereas 16.2% excise more than 10 cm above the transition zone.

Intraoperative frozen section tissue diagnosis is used as a routine practice by 4.8% of responders, whereas 29.5% use it during surgery whenever it is possible; 65.7% of the responders stated that it is not available in their institutions.

Absorbable sutures are used for the coloanal anastomosis by 88.6% of responders; 4.8% use them in conjunction with nonabsorbable sutures. Nonabsorbable sutures alone are used by 6.6%.

In a well neonate with HD extending to the right side or total colonic HD, 46.6% of responders adopt a staged approach, whereas 30.6% recommend a one-stage approach. The remaining responders stated that they have no experience with these types of HD. Many surgical options were reported for the management of this subset of patients. Transabdominal endorectal pull-through (Soave) is preferred by 45.7% of responders, whereas 27.6% advocate the Duhamel procedure. Laparoscopicassisted pull-through is recommended by 19.0% of responders and 7.3% do not have a specific preference.

In redo surgery, most of the responders stated that the choice of the operation will depend on the previous technique and the cause for redo. TEPT, if feasible, was recommended by 23.8% of responders, whereas the Soave technique was preferred by 37.1%, Duhamel with or without staplers was recommended by 27.6%, and the Swenson operation was recommended by 5.7%. The remaining responders stated that they have no experience with redo surgery.

In terms of the ultra-short segment HD (referring to patients with persistent constipation without an evident transition zone at the contrast enema, and an absent rectoanal inhibitory reflex on anorectal manometry), myectomy only was preferred by 50.5% of the responders, TEPT was preferred by 28.6%, and 19.0% recommended conservative treatment and follow-up. Only two responders advocate starting with conservative treatment and then myectomy in case of failure.

#### **Postoperative management**

Routine postoperative anal dilatation is recommended by 56.2% of responders at a mean time of  $4 \pm 1$  weeks, whereas 43.8% do not recommend routine regular dilatation at all.

#### Table 1 Summary of key findings

	Number of responders (%)
Diagnosis	
Contrast enema	13 (12.4)
Contrast enema + rectal biopsy	80 (76.2)
Contrast enema + rectal biopsy + manometry	12 (11.4)
Definitive surgery	
Routine one stage	62 (59.0)
Routine multiple stages	8 (7.6)
Either	35 (33.4)
Preferred surgical technique	
Transanal endorectal pull-through	72 (68.5)
Soave transabdominal endorectal pull-through	20 (19.0)
Laparoscopic-assisted pull-through	10 (9.5)
Duhamel with GIA staplers	3 (2.9)
Intraoperative frozen section	
Routine	5 (4.8)
Sometimes	31 (29.5)
N/A	69 (65.7)
Length of the excised proximal ganglionated bowel	(cm)
<5	29 (27.6)
>5	17 (16.2)
5-10	59 (56.2)
Right sided/total colonic: definitive surgery	
Soave transabdominal endorectal pull-through	48 (45.7)
Duhamel procedure	29 (27.6)
Laparoscopic-assisted pull-through	20 (19.0)
Redo surgery	
Transanal endorectal pull-through	25 (23.8)
Soave transabdominal endorectal pull-through	39 (37.1)
Duhamel with or without staplers	29 (27.6)
Swenson operation	6 (5.7)
Ultra-short segment	
Myectomy	53 (50.5)
Transanal endorectal pull-through	30 (28.8)
Conservative management	20 (19.0)

A postoperative rectal tube is recommended routinely by 21.0% of responders, whereas 3.8 and 27.6% recommend using it only in neonates and in difficult cases, respectively, and 47.6% do not recommend it at all.

## Discussion

The current survey of the practice patterns of a large group of pediatric surgeons in Egypt reflects the worldwide approach to HD with some distinctions. A number of pediatric surgeons in many countries have local resources, challenges, and practice patterns similar to those in Egypt, and will probably be interested in this study, which addresses the problems that they are probably facing.

There have been few reports on national surveys of HD in the literature [14,15]. This report is considered the first national survey conducted in Egypt despite a previous multicentric study evaluating the management of HD [13].

Our study highlights the diagnostic modalities for HD in Egypt. Unlike the Bradnock and Walker series [14], preoperative partial rectal biopsy obtained either by suction or by other techniques is practiced by 54.3%. Suction cup is available in few places. Pediatric surgeons working in centers with well-experienced pediatric pathologists available feel quite comfortable making a definitive diagnosis from a partial-thickness biopsy, whereas nonpediatric pathologists insist on a full-thickness biopsy.

The current study confirms the transition from using a staged approach to wide acceptance of primary pullthrough for left-sided HD. The safety and efficacy of primary pull-through has been shown and has been accepted by our survey respondents; 59.0% perform a one-stage repair as a routine practice in contrast to 7.6%, who are still using staged operations. The remaining surgeons perform either one-stage or multistage repair according to the working situations, although this percentage decreases to 51.0% in cases with long segment and total colonic aganglionosis. The two-stage operation was initially used because most patients presented later in life and had significant colonic distension or enterocolitis at the time of diagnosis. Now, more patients are diagnosed early in life, less colonic distension and healthier patient population allowed the development of one-stage procedure in the newborn period. In a retrospective concurrent series, the one-stage and twostage coloanal anastomotic approaches were found to result in equal complications in terms of the outcome of the operation [6]. However, stomal complications with the two-stage approach have been reported to be as high as 26.0%, and half required a stomal revision operation [6]. This persistent problem of stomal complications with the two-stage repair has been reported by Santos et al. [16].

The current study showed that more than 78.0% of surgeons are still not comfortable operating on patients in the neonatal period. In addition to some technical difficulties in starting the submucosal dissection, the uncertainty of precise localization of the transition zone in the absence of available intraoperative frozen section pathology dictates postponing definitive treatment until the transition zone is quite obvious during the preoperative contrast study as well as during surgery.

Before the past two decades, the use of an intraoperative frozen section biopsy for confirmation of the site of the transition zone was lacking in almost all hospitals in Egypt. However, there has been a steady increase in the number of pediatric surgical centers that have introduced this service. The availability of an intraoperative frozen section biopsy in the future will encourage many pediatric surgeons to perform a definitive surgery at an earlier age and to confidently resect less segments of the bowel when the transition zone is not clearly evident, especially in neonates.

There has been a change in the surgical option of leftsided colonic HD, with a predominance of the TEPT technique (68.5% of responders). Soave TEPT is still practiced by 19.0% of the responders. We believe that this change is because of the fact that previous experience with the traditional endorectal pull-through is not mandatory to perform TEPT safely.

The type of anastomosis is also transitioning toward the simpler coloanal anastomotic technique, with only 2.8% preserving the aganglionic rectum (i.e. Duhamel). This transition was also reported in other countries: the survey of pediatric surgeons published in 1979 in USA documented that 30.0% favored the Duhamel operation [16]

and three decades later, use of this technique has decreased to 5.4% [17].

We cannot ascertain whether the continued popularity of the Soave and Duhamel pull-through in Egypt is region specific or whether surgeons within the same institution use different approaches. Continued exposure to these techniques is potentially useful as they are popular approaches in redo surgery, 37.1 and 27.6%, respectively, and most responders in this survey utilize the Soave technique, 45.7%, and the Duhamel technique, 27.6%, in HD extending to the right side and total colonic aganglionosis.

Recent advances in minimal access surgery have led to the successful applications of laparoscopic-assisted techniques for the surgical management of diseases of the colon. Because the endorectal dissection is facilitated by laparoscopic mobilization of the rectosigmoid colon, there is less potential for overdilating the internal anal sphincter and thereby weakening the patient's fecal continence mechanism during the transanal dissection. The laparoscopic approach also provides greater versatility in fashioning the ganglionated pedicle proximal to the aganglionic colon and allows for completion of the pullthrough in patients with a longer aganglionic segment [9]. Our survey found limited popularity of laparoscopy in the management of left-sided HD and redo surgery (9.5%), whereas this percentage increased to 19.0% in HD extending to the right side and total colonic HD; this limited popularity is perhaps because of the limited number of pediatric surgeons with advanced laparoscopic skills as well as the availability of advanced laparoscopic facilities in many governmental hospitals. The relatively higher cost of laparoscopic-assisted pull-through compared with conventional surgery is another important factor, especially in the private health care sector. On the basis of the current survey, we cannot recommend a certain technique. However, we encourage greater use of laparoscopic-assisted pull-through to avoid laparotomy in patients with a longer ganglionic segment and those with no clearly identified preoperative transition zone.

The current survey found a variation in some technical operative details, for example the point of initiation of anorectal dissection; about 80.0% of responders prefer starting dissection less than 2 cm above the top of the anal columns. The concern is that dissection and subsequent anastomosis higher than 2 cm may be responsible for recurrence of postoperative obstructive symptoms, whereas dissection less than 0.5 cm close to the dentate line may damage the delicate nerve endings required for continence. Our results are in agreement with the Keckler *et al.* [17] series, where the mean distance for initiating the coloanal dissection was 7 mm.

The initial Soave repair reported leaving a portion of aganglionic rectal muscle. This portion of dysfunctional muscle has been hypothesized to contribute toward complications such as constipation, cuff abscess, and enterocolitis. One retrospective analysis compared patients with a 10–15 cm cuff with those with a less than 2 cm cuff and found that those with a short cuff had

significantly decreased length of hospitalization, with fewer patients requiring postoperative dilation. Although not significant, the incidence of enterocolitis was lower (9.0 vs. 30.0%) in the short cuff group [18]. In this survey, 97.0% of the respondents either shorten the muscular cuff or incise it to avoid complications.

A significant disparity in opinion exists in the amount of ganglionated bowel that should be resected above the transition zone, with 27.6% excising it to less than 5 cm, 16.2% excising it to more than 10 cm, and most of the responders excising about 5-10 cm above the transition zone. Surgeons, who resect longer segments to ensure reaching a normally innervated bowel, usually have no intraoperative frozen section biopsy service. In a previously published study, we found obstructive symptoms because of residual or acquired aganglionic segments in 15.0% of patients, who required redo surgery [19]. Apart from aganglionosis, some surgeons believe that there may be an area of hypoganglionosis in the bowel proximal to the transition zone, which may increase the risk of postoperative colonic dysfunction. Conversely, removal of excessive normal colon may decrease the absorptive mucosa without benefit. A recent retrospective analysis suggests that extensive resection does not decrease the rate of postoperative enterocolitis [20]. However, this may be an important area for future prospective studies.

### Limitations of this study

The present survey is limited by the inability to correlate the clinical practice with the outcome results and the fact that not all members of EPSA responded to e-mails or personal communication.

#### Conclusion

In summary, the current survey reflects the diversity in the practice patterns of the management of HD among pediatric surgeons in Egypt. Some surgeons have wellequipped neonatal ICUs, advanced laparoscopic facilities, frozen section biopsy service; so, they perform one-stage repair confidently in the neonatal period, and use liberally laparoscopy. Others, working under less favorable conditions, tend to postpone definitive surgery beyond the neonatal period; they depend on preoperative contrast studies for localization of the transition zone, and in some instances, do not hesitate to use a multiple-staged approach to ensure safe practice.

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#### **Conflicts of interest**

There are no conflicts of interest.

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# Appendix: Hirschsprung's disease: Egyptian Pediatric Surgical Association Survey form

Annex I. Hirschsprung's Disease: EPSA Survey form
Name (optional) Position: Hospital:
I- How many patients with HD do you manage (as the PRINCIPLE surgeon) per year A. <5 B. 5-10 C. 10-15 D. 15-20 E. >20
2- How many patients with HD do you manage (as an ASSISTANT) per year B. <5 B. 5-10 C. 10-15 D. 15-20 E. >20
<ul><li>3- Preoperative diagnostic tool?</li><li>A. Contrast only</li><li>B Biopsy only</li><li>C. Contrast and biopsy</li><li>D. Contrast, biopsy and ARM</li></ul>
<ul><li>4- What is your preferable technique for biopsy?</li><li>A. Partial thickness rectal biopsy. B. Full thickness rectal biopsy C. Suction rectal biopsy</li></ul>
5- Do you use intraoperative frozen section tissue diagnosis?         A. Not available       B. Yes as a routine       C. Sometimes
<ul><li>6- What is the preferable age at time definitive surgery?</li><li>A. Neonatal period (if definitive diagnosis was made)</li><li>B. I-3 month</li><li>C. &gt; 3 months</li></ul>
7- Do you do ONE stage repair? A. Yes as a routine B. Sometimes C. I prefer two stage repair D. I still do 3 stage repair
<ul> <li>8- What is your preferable definitive surgical technique for the standard rectosigmoid HD:</li> <li>A. Entirely Transanal ERPT B. Soave C. Duhamel with GIA stapler D. Duhamel without GIA stapler E. Swenson F. Lap assisted pull-through G. Others</li> </ul>
9- What is your preferred definitive technique for the very long segment or total colonic HD       A.       Straight trans abdominal ERPT B. Duhamel with pouch       C. Duhamel without pouch.       D.         Lap assisted pull-through       D. Others (please specify)       E. No experience with long segment       D.
<ul> <li>10- What is the preferred patient's position during Transanal ERPT</li> <li>A. Prone B. Supine C. I do not do transanal ERPT</li> </ul>
<ul> <li>I I- What is your choice for anal exposure during pull-through:</li> <li>a- Lone Star retractor</li> <li>B. Suture</li> <li>C. Both</li> <li>D. Other retractors</li> </ul>
12- Point of initiation of anorectal dissection and coloanalanastmosis above "dentate line"A.<1 cm
I 3- What the type of suture do you prefer to use during coloanal anastomosisa- Absorbable SutureB. Non absorbableC. Any of them
I4- How do you deal with the muscular rectal cuff during transanal ERPT:         A.       Incised interiorly       B. Shortening circumferentially to < 5cm
15- What is the length of ganglionated bowel do you excise proximal to the transition zoneA.<5 cm
<ul><li>I 6-Do you prefer putting a rectal tube postoperatively</li><li>A. Yes as a routine Only in neonates Only in difficult cases D. No</li></ul>
<ul> <li>I7- Do you recommend routine regular dilatation?</li> <li>A. Yes for &lt; two weeks</li> <li>B. 2-4 weeks</li> <li>C. 4-8 weeks</li> <li>&gt;8 weeks</li> <li>D. No routine regular dilatation</li> </ul>
<ul> <li>18- What is the median post-operative hospital stay</li> <li>A. &lt;3 days B. 3-5 days</li> <li>C. 5-7 days</li> <li>D. &gt;7 days</li> </ul>
<ul> <li>I9- What is your preferred technique for REDO pull through:</li> <li>A. Entirely Transanal ERPT if feasible</li> <li>B. Soave trans abdominal ERPT</li> <li>C. Duhamel with</li> <li>GIA stapler</li> <li>D. Duhamel without GIA stapler</li> <li>E. Swenson</li> <li>F. Others</li> </ul>
20 - What is your preferred approach for long segment and total colonic aganglionosis A. One stage B. Multiple stages
21- What is your preferred approach for ultra short HD         A.       Myectomy only B. Trananal ERPT       C. Conservative TTT       Others