Effect of endorectal pullthrough on external anal sphincter integrity (in cases of Hirchsprung's disease) using EMG

Ehab el-Shafei[a], Abeer K. el Zohiery[b], Rana el Hilaly[b] and Nouran Abaza[b]

Objective **The transanal mucosectomy of the aganglionic segment is a critical step in the transanal endorectal pullthrough procedure for the treatment of Hirchsprung's disease. It exerts considerable traction on the anorectal tissue during dissection. Anal sphincter electromyography (EMG) is an indispensable parameter for the diagnosis of patients with any anorectal dysfunction. The aim of our study was to assess the integrity of the anorectal sphincter after transanal endorectal pullthrough using anal EMG.**

Methods **This prospective study was carried out on 25 infants and children with Hirchsprung's disease who underwent the endorectal pullthrough (soave) procedure. Needle EMG was used to assess the sphincter preoperatively and postoperatively.**

Results **Preoperative EMG showed positive neuropathic changes in 28% of the patients. Postoperative EMG showed neuropathic changes in 60% of the patients, of whom 28% showed preoperative changes and 32% showed absolute postoperative findings, mostly related to difficult operative dissection.**

Conclusion **The functional results of the endorectal pullthrough procedure were acceptable overall.**

Significance **The reduced sphincter function encountered postoperatively was because of a combination of preoperative and intraoperative influences. *Ann Pediatr Surg* 11:13–17 © 2015 Annals of Pediatric Surgery.**

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Departments of [a]Pediatric Surgery and [b]Physical Medicine, Rheumatology and Rehabilitation, Faculty of Medicine, Ain Shams University, Cairo, Egypt

Correspondence to Ehab el-Shafei, MD, FRCSI, Department of Pediatric Surgery, Faculty of Medicine, Ain Shams University, 11331 Cairo, Egypt
Tel: +20 224 870 368; fax: +20 226 366 303; e-mail: ihab.shaf@gmail.com

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Introduction

Single-stage transanal endorectal pullthrough (TEPT), which was introduced by de la Torre in 1998, is a relatively new technique for surgery of Hirschsprung's disease (HD) [1].

Endorectal dissection has become the dominant minimal access procedure because of the ease and reliability in performing this technique [2]. TEPT can be performed abdominally by minilaparotomy [abdominal-assisted transanal pullthrough (AAPT)] or by laparoscopy [laparoscopic-assisted transanal pullthrough (LAPT)], or with a nonadditional procedure, which is total TEPT [3].

Transanal mucosectomy of the aganglionic segment of colon is a critical step in minimally invasive surgery for HD and exerts considerable traction on the anorectal tissue during dissection, thus making assessment of the functional outcome and colonic motility difficult [4].

Accordingly, the question arises as to whether TEPT impairs the integrity of the anorectal sphincter. As most of the children were too young to evaluate for fecal continence and stooling pattern, anorectal manometry, computerized eight-vector manometry, and endorectal ultrasonography were the tools used to answer the question [5,6].

Electrodiagnostic tests may be valuable in the assessment of patients with anorectal dysfunction and are complementary to imaging and manometry. Whereas the latter delineate morphological and functional sphincter changes, respectively, electrodiagnostic methods document, help to localize, and assess the innervation and anatomic integrity of the sphincters as well as the severity and mechanism of injury [7].

Although electromyography (EMG) has been discussed previously in the diagnosis and screening of HD [8–10], no data are as yet available on its use in assessing the integrity of the anorectal sphincter and muscular defects along the anal canal selectively after TEPT. Usually, physiological studies are carried out for an objective assessment of the sphincter musculature and its innervation before and after surgical treatment. The reduced sphincter function was probably related to a combination of muscle and nerve damage following trauma of the surgery [11].

The aim of our study was to assess the integrity of the anorectal sphincter after TEPT using anal EMG.

Patients and methods

This prospective study was carried out at Ain Shams University hospital (pediatric surgery, physical medicine, and rehabilitation departments) on 25 infants and children with HD during the period from January 2009 to July 2013. After obtaining approval of the ethical committee, a written consent was obtained from all the patients' parents following a detailed explanation of the procedure was provided.

Diagnosis was confirmed by contrast enema in 16 patients with classic reversed rectosigmoid ratio, whereas nine cases were diagnosed by rectal biopsy from above the peritoneal reflexion during exploration for colonic obstruction and leveling colostomy.

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All patients were subjected to thorough physical, neurological, and rectal examinations. They underwent anal EMG both preoperatively and at the fourth postoperative week. Cases diagnosed by transanal rectal biopsy as well as cases that missed postoperative EMG because of any reason were not included in our study.

Eleven cases underwent total TEPT, five cases underwent LAPT, and all nine cases with previous colostomy underwent AAPT.

Surgical technique

AAPT

Abdominal step was started first through a circumferential incision around the colostomy to detach it. We then mobilized and devascularized the colonic segment to be resected. The mesocolon was divided up to a point that allowed adequate mobilization of the colon for the pullthrough procedure.

LAPT

Pneumoperitoneum was obtained using an open technique through the umbilicus (in very small infants, the initial port was placed above the level of the umbilicus). The transition zone was located visually when possible and a seromuscular biopsy was obtained for histologic leveling. The major distal branches of the inferior mesenteric artery and vein were preserved whenever possible to prevent the late scarring of the muscular cuff left.

Transanal dissection (the common step in all patients)

Retraction was achieved using eight perianal retraction 3–0 or 4–0 silk sutures to evert the anus and expose the rectum. A circumferential incision was made in the mucosa 5–10 mm above the dentate line. The endorectal dissection was then carried proximally, remaining in the submucosal. When the submucosal dissection had extended proximally to a point above the peritoneal reflection, the rectal muscle was divided circumferentially and the full thickness of the rectum and the sigmoid was mobilized out through the anus. This required division of rectal and sigmoid vessels, which could be performed under direct vision using cautery or ligatures. When the transition zone was encountered, full-thickness biopsy sections were taken, and frozen section confirmation of ganglion cells was obtained. The rectal muscular cuff was split longitudinally either anteriorly or posteriorly. The colon was then divided and a standard Soave–Boley anastomosis was performed. The anastomosis was performed using absorbable braided suture.

For the electrodiagnosis, patients older than 1 year received chloral hydrate 25 mg/kg body weight 30 min before the test to achieve just a sedative not a hypnotic stage. They were placed on their left side with the hips and knees flexed. Their right thighs were grounded electrically. A local anesthetic spray was used to reduce pain caused by needle insertion. A concentric needle electrode (diameter 0.46 mm) was inserted perpendicularly into the subcutaneous layer of the external anal sphincter (EAS) muscle about 2 cm from the anal orifice. Deeper insertions were made at the anal orifice at an angle of 30° [12]. The EMG activity was measured in four quadrants of the sphincter (sphincter mapping). By moving the position of the electrode, 20 different motor units (MU) were identified. The MU potentials were collected and analyzed during relaxation, crying, coughing, and straining. They were displayedand recorded on the EMG device Toennies Neuroscreen Plus (Toennies Germany). The amplitudes, duration, and polyphasisity of the recorded compound muscle action potentials were studied at several sites to define areas of functioning muscle and identify any site of muscle injury [11]. The cut-off limits for normal motor unit action potential testing were considered according to the study by Del and Entrena [13] (Table 1). Detection of neuropathic MU at one or more of the sphincter quadrants was considered pathological.

Statistical analysis

IBM SPSS statistics (V. 20.0; IBM Corp., Armonk, New York, USA) was used for data analysis. Data were expressed as median percentiles for quantitative nonparametric measures and as both number and percentage for categorized data.

The following tests were performed:

(1) Comparison between two dependent groups for parametric data using a paired *t*-test.
(2) Ranked Spearman correlation test to study the possible association between two variables among each group for nonparametric data.
(3) χ^2-test to study the association between two variables or comparison between two independent groups of the categorized data.

The probability of error at 0.05 was considered significant, whereas at 0.01 and 0.001, they were considered highly significant.

Results

The patient group included 17 boys (68%) and eight girls (32%). Their ages ranged from 5 to 30 months, mean age 17.9 ± 7.6 months.

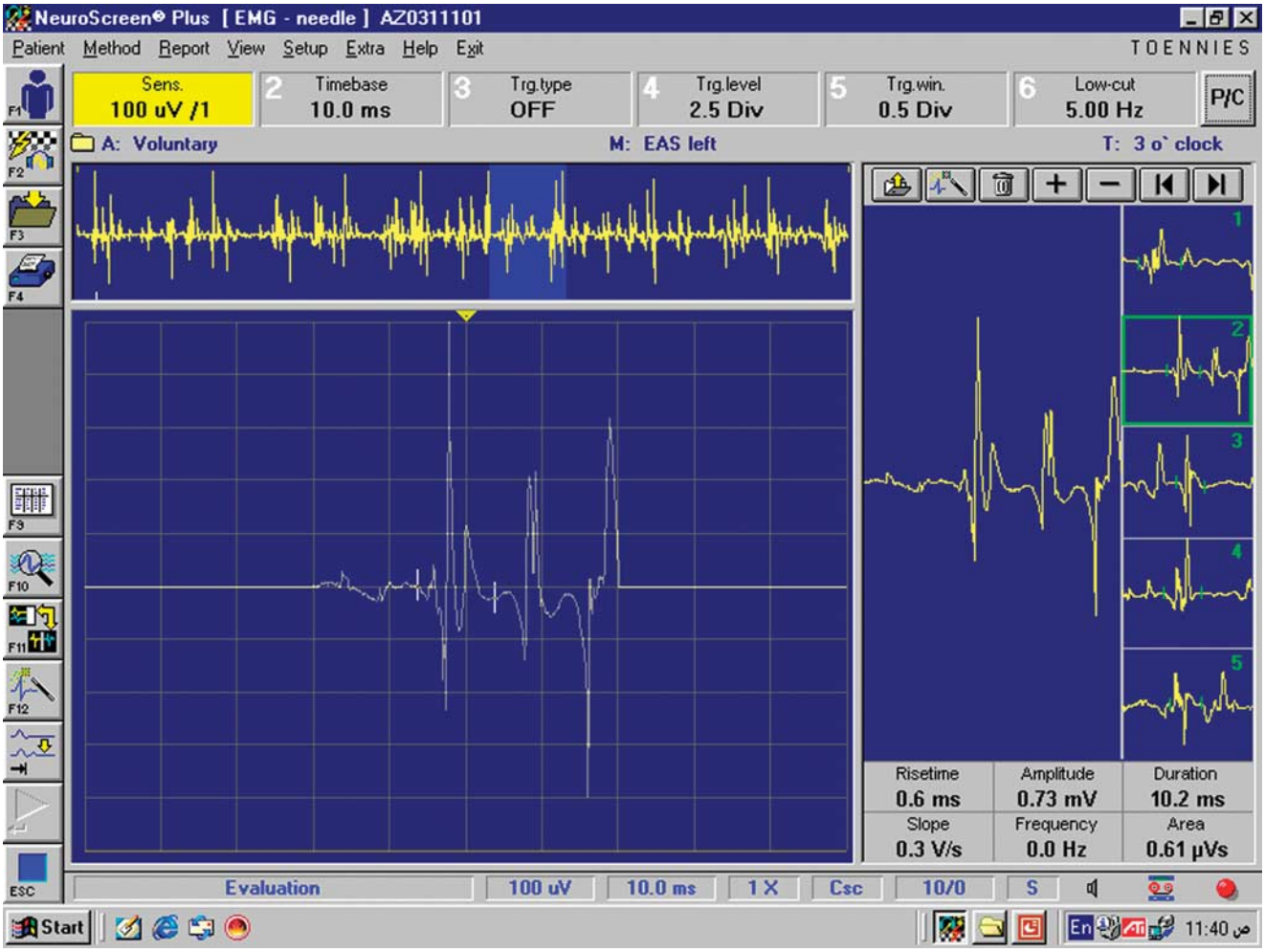
Preoperative anal EMG showed neuropathic changes in the form of large amplitude and/or wide polyphasic MU in seven patients (28%) (Fig. 1).

Table 1 Cut-off limits for normal MUAP

	MU analysis		
Age	Duration (ms)	Number of polyphasic potentials	Amplitude (mv)
Less than 1 year	3.94 ± 0.29	1.16 ± 0.14	0.22 ± 0.07
From 1 to 3 years	4.39 ± 0.41	1.32 ± 0.22	0.32 ± 0.1

MU, motor unit; MUAP, motor unit action potential.

Fig. 1



Neuropathic motor units in anal sphincter electromyography (large, polyphasic, and wide).

The preoperative MU analysis is shown in (Table 2).

Preoperatively, there was no statistically significant difference between patients with and those without neuropathic MU in terms of patients' mean age ($t = 0.125$, $P > 0.05$) or sex distribution ($\chi^2 = 0.47$, $P > 0.05$) among the two groups.

During the surgery, the operative transanal dissection time (TADT) ranged from 40 to 135 min, mean 72 ± 25.9 min.

This step was difficult in five cases, in which dissection was bloody, adherent, and time consuming.

In the postoperative period, two patients had leakage on the fifth day, thus requiring colostomy, whereas four patients had enterocolitis and were managed conservatively.

Postoperative EMG showed neuropathic changes in 15 of the 25 cases (60%) in the form of large amplitude and/or wide polyphasic MU, of whom 7 (28% of the total patients) had preoperative findings and 8 (32% of the total patients) showed only postoperative MU changes. Postoperative MU analysis is shown in (Table 3).

Among the seven cases who showed preoperative pathological MU, there was no statistically significant difference between pre-EMG and post-EMG MU analysis in the amplitude and duration of the MU (Table 4).

Table 2 Preoperative motor unit analysis

Preoperative MU	Amplitude	Duration	Percent of polyphasicity
Number of cases	25	25	25
Mean	0.398	4.6668	4.528
SD	0.26412	3.57233	4.6961
Minimum	0.11	0.27	0.8
Maximum	1.1	12.2	20

MU, motor unit.

Postoperatively, there was still no statistically significant difference between patients with and those without neuropathic MU in patients' mean age ($t = 1.329$, $P > 0.05$) or sex distribution among the two groups ($\chi^2 = 0.29$, $P > 0.05$) (Table 5).

There was no statistically significant difference between the mean TADT among the two groups ($P > 0.05$).

Out of those eight cases with only postoperative EMG findings, in five patients, had operative dissection was difficult and in three patients, it went smooth. In the

Table 3 Postoperative motor unit analysis

Postoperative MU	Amplitude	Duration	Percent of polyphasicity
Number	25	25	25
Mean	0.6056	7.256	7.684
SD	0.39435	3.5321	6.3125
Minimum	0.18	2.6	1
Maximum	1.7	13	22

MU, motor unit.

Table 4 Comparison between preoperative and postoperative motor unit analysis among cases with pathological preoperative EMG

	Mean	Number	SD	*t*	*P*	Significance
Preoperative amplitude	0.7686	7	0.19222			
Postoperative amplitude	0.7871	7	0.23300	−0.782	0.464	NS
Preoperative duration	9.7286	7	1.81081			
Postoperative duration	10.3	7	2.1087	−2.326	0.059	NS

EMG, electromyography; NS, nonsignificant.

Table 5 Comparison between patients with postoperative neuropathic EMG and those with normal postoperative study in the transanal dissection time

	NMU ($n=15$)	Normal ($n=10$)	*t*	*P*	Significance
TADT (minutes)	75 ± 28.4	67.5 ± 22 5	0.7	0.491	NS

EMG, electromyography; NMU, neuropathic motor units; NS, nonsignificant; TADT, transanal dissection time.

remaining 10 patients, no postoperative electrodiagnostic changes were detected.

Discussion

The patients' mean age was 17.9 ± 7.6 months, with a relatively narrow range (from 5 to 30 months), in an attempt to minimize the myths of interpretation of the EMG results.

EMG study with a concentric needle electrode of the EAS muscle continues to be a fully reliable and reproducible, fast, and almost painless method to differentiate a healthy from a diseased muscle [13]. It has been proved to be superior even to ultrasonography and manometry in recognizing anal sphincter damage [14].

In HD, the possible causes of postoperative fecal incontinence may be intraoperative sphincter damage and/or existing associated anomalies in sphincter muscles or their innervations. Even though numerous studies have been attempted to clarify the pathophysiology of the aganglionic segment, little attention has been paid to the striated muscle function in HD [10].

To investigate the possibility of iatrogenic trauma during operation, preoperative and postoperative electrophysiologic assessments were performed. Preoperative neurogenic affection of the sphincter muscles was confirmed by the marked complexity of the MU potentials as this parameter had been proved to be reliable with most physicians [7,10,11,15].

In the current study, preoperative anal EMG showed neuropathic changes in 28% of our cases, a finding that was supported previously by Gadallah and colleagues They detected preoperative neuropathic findings in 20% of their patients. They even suggested that if original sphincter affection was detected preoperatively in a child with HD, he or she is more likely to develop postoperative fecal incontinence irrespective of the surgical intervention performed [10]. This unexpected finding of original sphincter affection was also suggested by Springall *et al.* [16].

Postoperative anal EMG showed neuropathic changes in the majority of the cases (60%). Although it seems a high percent, it did not statistically reflect a true defect in the surgery. Almost half of them (28%) had shown these changes preoperatively. Moreover, among those seven patients (28%), there was no statistical difference between preoperative and postoperative MU analysis in amplitude and duration, thus eliminating the effect of the surgery on the anal sphincter in those patients. Only 8 (32%) of our cases exclusively had postoperative affection of the anal muscle integrity, of whom five cases (20%) had a definitely recognized intraoperative reason and three cases (12%) had unexplained postoperative anal pathology unrelated to either preoperative or intraoperative clear reasons. During surgery, endorectal dissection was difficult in those five cases as it was bloody, adherent, and time consuming, with considerable anal stretch.

Although statistically nonsignificant, the mean TADT was higher in those who had postoperative neuropathic EMG than those without, highlighting the effect of prolonged intraoperative manipulation on anal sphincter integrity. Thus, postoperative pathological EAS findings could be attributed to the surgical procedure itself in only 32% of our cases.

We did not perform the pudendal nerve terminal motor latency (PNTML) for many reasons, mainly because many physicians have indicated only a limited role for PNTML testing in the management of anal sphincter dysfunction [17,18]. Moreover, Sentovich and his colleagues reported that it was often difficult, with failure to determine one or both PNTMLs because of patient discomfort or anxiety, or because of the inability to obtain a reproducible MU potential. In addition, among electrodiagnostic tests, concentric needle EMG of the EAS is the most important. It shows muscle denervation, quantitatively estimates muscle reinnervation, estimates the level of motor neuron excitability, and assesses several kinesiological parameters [7]. However, PNTML is a measurement of the conduction in the fastest-conducting nerve fibers. As the fastest latency is not influenced by the presence of increased numbers of slowly conducting damaged axons, the PNTML does not provide a quantitative estimation of the extent of abnormality in the nerve. Therefore, normal pudendal latency does not rule out abnormal innervation [19]. The clinical usefulness of this test is still controversial and for this reason, the guidelines provided by the American Society

of Gastroenterology do not recommend PNTML for the evaluation of patients with anal incontinence [20]. Finally, to reduce the time and burden on our cases, being so young and easily exhausted, we spared our cases a test that might be difficult, unreliable, or unnecessary.

Hence, the analysis of the mean values of the isolated motor unit action potentials is reliable enough [13]. Moreover, MU analysis showed injuries to the sphincters that were not detected clinically and was successful in mapping the sites. The parameters were indicative of a continuing process of denervation and reinnervation usually associated with anal damage [11,14,15].

Therefore, we did not depend on interference pattern analysis as voluntary squeeze is difficult to request and assess in young children, especially in the precontinence age, as in our cases.

The postoperative neuropathic EAS injury that we found was detected previously by Springall and his colleagues in incontinent children following surgery for HD. They even recommended modification to the clinical management of such patients. In fact, their cases were assessed by EMG postoperatively only and were compared with other age-matched and sex-matched control patients [16]. Ignorance of the preoperative elements that influence the post-operative results usually leads to false interpretation of the data. In fact, unifying the surgical procedure and minimizing the age difference among the cases reduce misreading of the results. However, in our study, continence tended to be unpredictable taking into account the preawareness age of our patients in comparison with the elder, incontinent cases that the surgeons had selected in their study.

Conclusion

Overall, the functional results of the endorectal pull-through procedure were acceptable. The reduced sphincter function postoperatively was probably related to a combination of congenital preoperative muscle and/or nerve damage, as well as excessive intraoperative manipulation.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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