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Damaged anal canal as a cause of fecal incontinence after surgical repair for Hirschsprung disease – a preventable and under-reported complication

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ABSTRACT

Introduction: Fecal incontinence after the surgical repair of Hirschsprung disease is a potentially preventable complication that carries a negative impact on patient's quality of life.
Methods: Patients that were previously operated for Hirschsprung disease and presented to our bowel management clinic with the complaint of fecal incontinence were retrospectively reviewed. All patients underwent a rectal examination under anesthesia looking for anatomic explanations for their incontinence.
Results: One hundred three patients were identified. 54 patients had a damaged anal canal. 22 patients also had a patulous anus. The operative reports mentioned the pectinate line in 32 patients, in 12 it was not mentioned, and in 10 patients the operative report was not available. All patients with a damaged anal canal suffered from true fecal incontinence; 45 of them are on daily enemas (41 are clean and 4 are still having "accidents"), 7 are not doing bowel management due to noncompliance and 2 patients have a permanent ileostomy. 49 patients did not have a damaged anal canal, 25 of those responded to changes in diet and medication and are having voluntary bowel movements.
Conclusion: Fecal incontinence may occur after an operation for Hirschsprung disease. When the anal canal is damaged, incontinence is always present, severe, and probably permanent. The preservation of the anal canal may avoid this complication.

Fecal incontinence, inability to control bowel movements, after the surgical repair of Hirschsprung disease is a devastating and potentially preventable complication not frequently mentioned in the literature [1–11]. Few post-operative complications have a greater impact on patient's quality of life [5].

We hypothesized that during the transanal portion of the surgical repair of patients with Hirschsprung, the anal canal can be invaded or completely damaged and this could be an explanation for the fecal incontinence faced by some patients.

We have developed a specific protocol to evaluate such patients. A key aspect is to differentiate between patients that suffer from constipation and overflow pseudo-incontinence from those with true fecal incontinence. The evaluation includes a contrast enema and an examination under anesthesia to specifically look for the integrity of the anal canal [1,6].

1. Methods

Medical records of patients previously operated for Hirschsprung disease at other institutions presenting to our bowel management clinic with the complaint of fecal incontinence, from July 2005 until November 2012, were retrospectively reviewed.

All patients underwent a contrast enema and a rectal examination under anesthesia looking for possible anatomic explanations for their incontinence. Specifically the contrast enema allows for direct evaluation of colonic length and its characteristics (dilated vs. non-dilated colon); the examination under anesthesia evaluates the integrity of the anal canal (anal canal preservation happens when the anastomosis is performed above the pectinate line) and sphincter (presence of a patulous anus indicates major sphincter compromise); and it also rules out stricture. Data was analyzed using univariate analysis with damage anal canal and fecal incontinence set as endpoints, which compared cohort

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Table 1
Comparison between the group of patients with damaged anal canal vs. patients with an intact anal canal.

Endpoint damaged anal canal	Damaged anal canal (54 patients)	Intact anal canal (49 patients)	Univariate analysis	
			P value	OR (95% CI)
Down syndrome	11 (20.4%)	1 (2%)	*0.019	12.3 (1.52–99.1)
Type of procedure:			0.053	
Soave (ref)	42 (84%)	31 (66%)		1.00
Swenson	2 (4%)	5 (10%)		0.3 (0.06–1.63)
Duhamel	1 (2%)	7 (14.9%)		0.11 (0.01–0.91)
Patulous anus	21 (44.7%)	4 (10.5%)	*0.001	6.87 (2.10–22.4)
Operative report mentioned pectinate line	32 (72.7%)	27 (71.1%)	0.78	0.87 (0.34–2.25)
Underwent reoperation	16 (32%)	3 (6.4%)	*0.004	6.90 (1.86–25.6)
Hypermotile vs hypomotile colon	22 (46.8%) vs 25 (53.2%)	11 (23.4%) vs 36 (76.6%)	*0.019	2.88 (1.19–6.98)
Voluntary bowel movements	0 (0%)	25 (55.6%)	*0.0009	0.008 (<0.001–0.14)

Endpoint damaged anal canal	Multivariate analysis	
	P value	OR (95% CI)
Down syndrome	0.17	5.86 (0.86–40.1)
Age at pull-through	0.18	0.98 (0.94–1.01)
Type of procedure	0.09	0.03 (<0.01–1.43) & 0.18 (0.02–1.89)
Underwent reoperation	*0.005	12.8 (2.14–76.6)

* Statistically significant.

demographics and identified potential confounding or impactful factors. Significant differences between groups determined by the univariate analysis were included in a multivariate analysis with logistic regression to provide odds ratios the same endpoint to identify the most significant contributor to set outcome. Data analysis was performed on Statistical Analysis Software (SAS Institute, Inc. Cary, NC).

Reoperations were defined as redo pullthrough due to previous complications such as: stricture, dehiscense, persistent aganglionosis or transition zone.

Bowel control was defined as the ability to voluntarily pass stool combined with the ability to retain stool thereafter. To have a voluntary bowel movement requires an individual to be capable of feeling the urge to defecate, the ability to avoid the immediate passage of stool, and the coordination to pass stool into the toilet. The capacity to retain should be maintained without fatiguing [12].

Institutional review board approval was obtained for this study (#2012–3443).

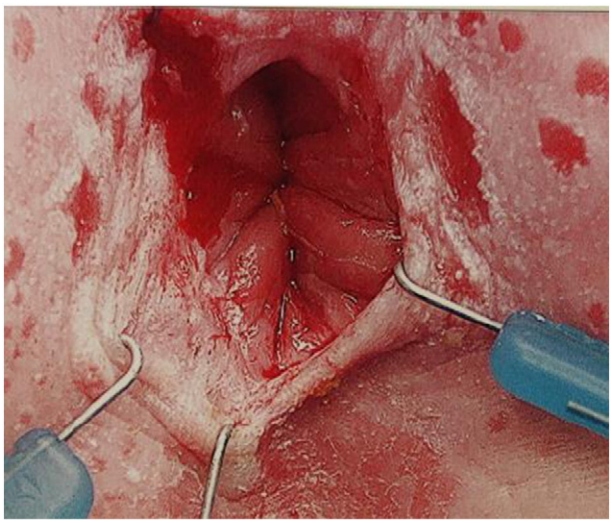

Fig. 1. Damaged anal canal in a patient previously operated for Hirschsprung (there is no visible pectinate line).

2. Results

One hundred three patients were identified as meeting our inclusion criteria. The mean age at initial Hirschsprung pull-through operation (6 months vs. 13 months, $P = 0.054$) and presentation for bowel management (6.1 years vs. 7.2 years, $P = 0.093$) were similar between damaged and intact anal canal cohorts respectively. Gender was not different between groups (76% male with damaged and 78% male with intact anal canal, $P = 0.85$). Similarly the mean follow-up duration was nearly 2 years (1.9 years with damaged and 1.8 years with intact anal canal, $P = 0.69$).

In this population, 54 (52%) patients had a damaged anal canal and 49 (48%) had an intact anal canal (Table 1).

In 30 (56%) of the 54 patients with a damaged anal canal, the damage was complete, it affected the entire circumference, with absence of a pectinate line (Fig. 1). Additionally, in some cases, rectal mucosa was anastomosed to the anal skin (Fig. 2). The anal canal was partially damaged in 24 patients, meaning that there was absence of a pectinate line in some portions of the anal canal. Of the damaged anal canals, 22 patients also had a patulous anus (Fig. 3). Upon reviewing the operative reports, the pectinate line was mentioned in 32 of the 44 available reports. In 2 reports the surgeons actually described the incision being made at the muco-cutaneous junction.

The incidence of rectosigmoid and total colonic aganglionosis was similar between groups 81% and 7% (damaged anal canal) and 75% and 4% (intact anal canal) respectively. Soave pull-through was the most common operation in both cohorts (Table 1).

After correcting for multiple differences between cohorts including age at surgery, Down syndrome and type of surgery performed, the need for reoperation was the only significant factor that increased the odds of having a damaged anal canal ($P = 0.005$). Knowing that undergoing reoperation affects the integrity of the anal canal, we explored the most influential factors to achieve fecal continence in this population. Patient demographics were similar between patients with and without voluntary bowel movements including the segment of bowel involved with Hirschsprung disease and type of initial surgery. Univariate analysis with fecal incontinence as the endpoint revealed associations with fecal incontinence to include having a reoperation, a patulous anus, a non-dilated colon and a destroyed anal canal (Table 2). When these most influential factors were collectively analyzed by multivariate analysis, the only significant factor leading to fecal incontinence was having a damaged anal canal ($P = 0.015$).