Original Article

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Intractable chronic constipation in children: Outcome after anorectal myectomy

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ABSTRACT

Background: Many children with constipation fail to respond with conventional medical therapy. Surgery can produce a good result in dysfunction of the colon secondary to aganglionosis. However, its role in treating idiopathic constipation is more controversial. **Patients and Methods:** A consecutive series of 44 patients with chronic idiopathic intractable constipation were included in this study. All children were investigated by barium enema and anorectal manometry. Due to inadequate response to medical therapy, all of these patients were selected for internal sphincter myomectomy. Patients were followed-up from 3 to 12 months. **Results:** Short-term (3 months) and long-term (6 months) follow-up was available for all patients. The histology examinations showed normal ganglion cells in 32, hypoganglionosis in eight and aganglionosis in four patients. In short-term, regular bowel habits, without the need for laxatives or low dose drugs were recorded in 35 patients (79.5%). Overall there was an improvement in 68.2% of the children after 6 months follow-up. There was not any correlation between histopathological findings, duration of symptoms, age and sex of operation and response to myectomy. **Conclusion:** anorectal myectomy is an effective procedure in patients with intractable idiopathic constipation. It relieves symptoms in 68.2% of patients with chronic refractory constipation.

Key words: Anorectal myectomy, children, idiopathic constipation

INTRODUCTION

Bowel problems in childhood are common, but often difficult to manage. This disorder has led to many controversies between paediatricians, surgeons and psychiatrists over managing these patients. Many children with constipation fail to respond or rapidly relapse with conventional medical treatment. Surgery can usually produce a good functional result in dysfunction of the colon secondary to lesions like aganglionosis. However its role in treating idiopathic constipation is more controversial. Internal anal sphincter achalasia (IASA) is a clinical condition with a presentation similar to Hirschsprung's disease. The diagnostic criteria of IASA are based on anorectal manometry (ARM), which shows the absence of the recto-anal inhibitory reflex (RAIR) and normal rectal biopsy. [1-3]

Posterior internal sphincter myectomy (ISM) has been recommended as the treatment of choice for patients with ultra-short Hirschsprung's disease and IASA.[2,4] Nevertheless, the choice of surgical procedure in children with chronic constipation and hypoganglionosis is not agreeable.

The aim of this study was to assess the outcome of children who have been diagnosed with intractable chronic constipation and histopathologic condition. All of whom have been treated with posterior ISM.

PATIENTS AND METHODS

This was a prospective study of 44 consecutive children admitted to Booali's Hospital, Sari, for investigations and treatment of chronic constipation between January 2010 and September 2012. All children presenting with intractable constipation that did not respond to classic conservative treatment (diet, laxatives or enema) over a period of 3 months or more were included in the study. The age at operation ranged from 1 to 12 years (median: 4.6 years) and post-operative follow-up time ranged from 3 to 12 months (median 5 months).

A full history was taken, including duration of symptoms, age of onset, gender and type of medical

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treatment. Patients were excluded from the study if they had an underlying anorectal anomaly, documented metabolic and endocrine disease and contrast enema showing classical transition zone. All children were investigated by barium enema followed by ARM. Anal manometry was performed without muscle relaxants and with the patient lying in left lateral position by a method based on a balloon system. The presence of rectoanal relaxation reflex was examined by inflation of the rectal balloon with air (10-60 ml). The rectoanal reflex was considered as habitual constipation when a fall of at least 25% from the basal resting pressure level occurred after inflation of the balloon on three consecutive measurements.[4]

Due to an inadequate response to medical treatment, all of these patients were selected for surgery. A total of 44 children had myectomy in addition to the medical treatment of their chronic constipation. Under general anaesthesia and in lithotomy position, an incision was placed on the dentate line at 6 O'clock and the internal sphincter muscle was identified. The dissection was extended proximally to include the internal sphincter muscle. The myectomized muscle strips were 1 cm in width and the length of the strips ranged from 4 to 6 cm. The myectomy specimens were examined histologically by eosin and haematoxylin staining. Patients were followed-up for a period ranging from 3 to 12 months. Follow-up protocol included questionnaire regarding bowel habits, complications and symptomatic improvement. The Research Ethics Committee of Hospital approved this research project.

RESULTS

All patients suffered chronic constipation averaging about 28.4 months in duration (range: 3-71 months). They were refractory to conventional medical treatment consisting of the daily use of laxatives, cathartics or enemas. All children had finding consistent with idiopathic constipation on manometry. Anal manometry showed a weakly response of RAIR by 40-60 ml pressure of balloon in all patients.

Soiling was present in 14 (31.8%), distress in defecation in 28 (63.6%) and rectal bleeding in 3 (6.8%) patients. Short-term (3 months) and long-term (6 months) follow-up was available for all patients.

The histology examinations of internal anal sphincter (IAS) myectomy in 32 (72.7%) children showed normal ganglion cells. Furthermore, we had hypoganglionosis and aganglionosis in 8 (18.2%) and 4 (9.1%) patients. The patients with evidence of the ultra-short segment Hirschsprung had cured in 2 and partial improvement in others. In hypoganglionosis group, 2 (25%) patients had regular bowel motions without the use of laxatives. 4 (50%) patients had regular bowel motions, but remained on small doses of laxatives [Table 1].

In short-term, regular bowel habits, without the need for laxatives or low dose drugs was recorded in 35 patients (79.5%), whereas 9 (20.5%) achieved no improvement.

Overall there was an improvement in 68.2% of the children after 6 months follow-up, with 25% of those showing an excellent and 43.2% partial improvement result. Fourteen (31.8%) of the patients continued to have severe constipation 6 months after posterior myectomy. There was not any correlation between histopathological findings, duration of symptoms, age and sex of operation and response to myectomy. Table 2 summarises the results in 44 patients in short and long-term follow-up post-operatively. Complication included only in one case of rectal bleeding that spontaneously stopped after 12 h.

DISCUSSION

Nutritional support, laxatives and enemas are the first line of treatment of chronic constipation and approximately 85% of cases could improve or cure by conservative medical therapy.[5] Surgical intervention for patients with severe idiopathic constipation is rarely necessary. It is acceptable only once medical management has proved unsuccessful.

Table 1: Histopathological findings of anorectal myectomy from 44 patients with intractable constipation and clinical outcome

Neuronal dysplasia	No. of patients (%)	Clinical improvement (partially or complete) (%)
Aganglionosis	4 (9.1)	4 (100)
Hypoganglionosis	8 (18.2)	6 (75)
Normal	32 (72.7)	25 (78.1)
Total	44 (100)	35 (100)

Table 2: Clinical outcome of anorectal myectomy for intractable idiopathic constipation

Bowel pattern achieved	No. of patients	
	After 3-month (%)	After 6-month (%)
Excellent*	18 (40.9)	11 (25)
Partial improvement**	17 (38.6)	19 (43.2)
No improvement	9 (20.5)	14 (31.8)
Total	44 (100)	

*Regular bowel habits, no laxatives; **regular bowel habits, small dose of laxative

In most children with chronic constipation, the aetiology is unknown. In a reported series of children with idiopathic constipation, Keshtgar *et al.* [6] had shown underlying abnormalities in these children. These include higher threshold to rectal distension, lower contractility of the rectum and thickened IAS. They reviewed 144 children with idiopathic constipation and found a correlation between the thickness of IAS and severity and duration of symptoms, size of megarectum and age of patient. Therefore, children with a longer history of constipation had thicker IAS. The usefulness of diagnostic myectomy to provide muscle biopsy specimen is well understood. In addition, the myotomy therapy is not a new surgical technique. It was successfully applied for achalasia of oesophagus and hypertrophic pyloric stenosis.

Anorectal myectomy which was developed from a technique described by Lynn and van Heerden[7] for a short segment Hirschsprung is suggested for treatment of intractable constipation.

In Holschneider's[8] review of the literature, 144 (76%) of 189 patients had excellent results after myectomy. De Caluwé *et al.*[2] in their study have reported 2-6 years follow-up of 15 patients after ISM. Seven of these 15 patients had regular bowel motions without any medication and six needed small doses of laxatives. In a similar study Doodnath and Puri[9] and Redkar *et al.*[10] have reported 87.5% and 93% success rate. In our study, normal bowel habit with a small dosage of drug or without the need for laxatives was recorded in 79.5% patients after 3 months. However, it decreased to 68.2% after 6 months. On the other hand, the present study shows that bowel habit improvement occurs in most patients who undergo ISM for chronic intractable constipation and there is not any correlation between histopathological findings and success rate. Furthermore, there was not any correlation between the response to myectomy therapy and duration of constipation and age of children. Although, this might be due to a small number of patients in our series.

CONCLUSION

Anorectal myectomy is an effective and technically simple procedure in selected patients with severe idiopathic constipation. It therapeutically relieves symptoms in 68.2% of patients with chronic refractory constipation.

One study limitation was that there was no group-controlled comparison. Subsequent studies will need to evaluation in a large series of two groups (normal and hypoganglionosis) separately and with longer duration of follow-up. Because, this beneficial effect may be due to a transient decreasing tension in IAS.

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