

ORIGINAL ARTICLE

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Pneumatic dilatation for childhood achalasia

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Abstract Treatment of achalasia by pneumatic balloon dilatation (PBD) is well established in adults. Due to limited experience and the rarity of the condition in children, there are relatively few reports in the paediatric literature. Although PBD has been reported as a primary method of treatment, there are no reports of secondary PBD for childhood achalasia. Between 1995 and 1999, five patients underwent treatment for achalasia (age: 9–14 years; M:F = 4:1). The presenting symptoms were dysphagia (5), vomiting episodes (2), aspiration (1), food-bolus obstruction (1), and failure to thrive (1). In all patients a barium swallow and manometry were used to confirm the diagnosis. Three underwent primary PBD. Two who had previously undergone surgical myotomy underwent secondary PBD for recurrence of symptoms. Dilatation was performed using a 35-mm balloon with the child under general anaesthesia. Technical success was defined as demonstration of a waist under screening at lower pressures followed by abolition of the waist at higher pressures. In addition to reviewing our results, a systematic review of the literature was performed (Medline, Cochrane Library, Pubmed, Embase). Three patients (primary dilatation) showed excellent improvement after a single dilatation. In two cases (secondary dilatation) three and five attempts were required. No complications were encountered. The mean follow-up period was 2 years (1–3.5 years) and four patients remained asymptomatic, an overall success rate of 80%. The literature review revealed similar good results in most of the recent reports. Thus, PBD as a primary treatment for childhood achalasia has a success

rate of 70%–90% with minimal side effects, short hospital stay, and good patient acceptability over an operation. We have also established the usefulness of this method as a secondary treatment when symptoms recur after surgery.

Keywords Achalasia · Pneumatic dilatation · Children

Introduction

Achalasia of the cardia is a disorder of unknown aetiology characterised by failure of the lower oesophageal sphincter (LOS) to relax during deglutition. Surgery has been the traditional treatment, although pneumatic dilatation and botulinum toxin injection are recently becoming successful [1, 2, 5, 13]. Pneumatic balloon dilatation (PBD) of the cardia is a well-established treatment in adults [11]. However there are only a few reports of this procedure in the paediatric literature [6–10, 12, 14], as only 4%–5% of cases present during childhood. Although PBD has been reported as a primary method of treatment, there are no reports of secondary PBD for childhood achalasia. The aims of the present study were to report our results of primary and secondary PBD and to conduct a systematic literature review of PBD in childhood achalasia.

Materials and methods

Between 1995 and 1999, five patients aged 9–14 years were diagnosed with achalasia of the oesophagus. In all patients a barium swallow and manometry were used to confirm the diagnosis. PBD was performed with the patient under general anaesthesia by an experienced radiologist under fluoroscopic control in the operating theatre. A 35-mm Rigiflex balloon (Boston Scientific Corporation, Watertown, MA) was used and the inflation pressures ranged between 1.5 and 2.0 atm. The pressure was maintained for 2–3 min and the definition of technical success was demonstration of a waist under screening at lower pressures, followed by abolition at higher pressures. The patients remained nil by mouth for 6 h and were discharged on return to normal feeds. The clinical details and

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results are reviewed and summarised in Table 1. A systematic review of PBD in childhood achalasia was conducted by a literature search (Medline, Pubmed, Embase, Cochrane Library). All the articles on this subject are reviewed and the technical details/results summarised in Table 2.

Results

There were five patients (four boys and one girl) with ages ranging from 9 to 14 years. Dysphagia (5) was the commonest symptom, followed by vomiting (2), aspiration (1), food-bolus obstruction (1), and failure to thrive (1). Patients 4 and 5 had previously undergone a surgical cardiomyotomy combined with an anti-reflux operation. These two patients presented with recurrent symptoms, and in both contrast studies were performed to prove intactness of the anti-reflux procedure.

Patients 1–3 underwent PBD as the primary treatment as a day-case procedure, and there was excellent improvement of symptoms following a single dilatation. The mean follow-up period was 2 years (range: 1–3.5 years). The patients were not subjected to follow-up investigations, as they remained well and free of medication during follow-up (Table 1).

In patients 4 and 5 PBD was performed as a secondary treatment. They required multiple dilatations, and the hospital stay varied between 1 and 3 days due to delay in establishment of feeds during some episodes. Each dilatation was followed by a temporary improvement in symptoms. Good improvement was achieved in patient 4 after four dilatations, and she remained free of medications thereafter. Patient 5 is still undergoing secondary dilatations for recurrent dysphagia. Manometry revealed abnormal oesophageal peristalsis, and he required anti-reflux medications for control of symptoms. Four patients remained asymptomatic, a success rate of 80% in the current series.

Discussion

Achalasia, described by Thomas Willis in 1674, was initially treated by dilatation of the lower oesophagus with whalebone. The forceful disruption of the lower oesophagus by this means relieved dysphagia and other obstructive symptoms. Current procedures, PBD and surgical myotomy of the LOS, mimic this earlier approach. The relative merits of these treatments have been

Table 1 Clinical details of pediatric patients with achalasia ($n = 5$)

Patient no.	1	2	3	4	5
Age (years)/sex	12; M	11; M	14; M	9; F	11; M
Symptoms	Dysphagia, failure to thrive	Vomiting, dysphagia	Vomiting, dysphagia	Dysphagia, food-bolus obstruction	Dysphagia, aspiration
Previous operations	Nil	Nil	Nil	Cardiomyotomy + fundoplication	Cardiomyotomy + Thal operation
Diagnosis	Contrast, manometry	Contrast, manometry	Contrast, manometry	Contrast, recurrence of symptoms	Contrast, recurrence of symptoms
No./type of dilatations	1 Primary	1 Primary	1 Primary	4 Secondary	5 Secondary
Complications	Nil	Nil	Nil	Nil	Nil
Hospital stay (days)	1	1	1	1–3 (mean 1.7)	1–3 (mean 1.9)
Follow-up (years)	1	2	2	1	3
Follow-up investigations	Nil	Nil	Nil	Contrast study	Contrast, manometry
Current symptoms	Nil, thriving	Nil	Nil	Nil	Recurrent dysphagia
Current medication	Nil	Nil	Nil	Nil	Cisapride

Table 2 Review of literature on pneumatic dilatation for childhood achalasia (N/A not available)

Author	Year	Balloon size	Pressure/time	Success rate (%)
Present	2000	35 mm	1.5–2.0 atm (2–3 min)	80 ($n = 5$)
Hamza	1999	18 mm (<3 years, <10 kg) 30 mm (4–8 years, 10–20 kg) 35 mm (>8 years, >20 kg)	20psi–35psi (5 min)	90 ($n = 11$)
Hammond	1997	18–30 mm (2–3 balloons in tandem)	1.5–3.0 atm (5 min)	75 ($n = 4$)
Wilkinson	1997	23–30 mm (starting at smaller size)	N/A	100 ($n = 3$)
Perisic	1996	N/A	N/A	83 ($n = 12$)
Emblem	1993	N/A	N/A	50 ($n = 4$)
Samarasinghe	1991	N/A	N/A	100 ($n = 4$)
Nakayama	1987	N/A	7–8 lb/m ² (30–45 s)	73 ($n = 15$)
Berquist	1983	N/A	N/A	62 ($n = 8$)
Boyle	1981	N/A	7–8 lb/m ² (30–45 s)	80 ($n = 10$)
Swenson	1961	N/A	8 lb/m ² (5 min)	66 ($n = 6$)

debated without resolution for more than 40 years [2, 5, 13]. Drug therapies with a long-acting nitrate or calcium-channel-blocking drug and intra-sphincteric injection of botulinum toxin during endoscopy are other modalities occasionally used in patients with achalasia [1].

Surgery for childhood achalasia is effective in relieving obstruction in 86%–100% of cases [2, 5, 13], but results in gastro-oesophageal reflux and post-operative complications in 42% [13]. In adults, PBD is recommended as the initial procedure, but experience in the paediatric population is limited due to the rarity of the condition in children [2, 5]. The results of simple dilatation using a bougie were often disappointing as the calibre of these dilators increased to a maximum of only 15 mm diameter and the stretch was often inadequate, unlike the inform radial stretch achieved with pneumatic balloons [1]. Early reports did not favour PBD in children [5], but some authors recommended it in children older than 9 years [2]. However, many recent publications report high success rates in children [3, 4, 6–10, 14].

Nakayama et al. [8] were the first to report good results after PBD and favoured it as a logical first step. Reviewing the paediatric series reported between 1980 and 1987, they identified a success rate of 49%. Hammond et al. [6] reviewed the results up to 1997 and found a success rate of 74%. With the advent of low-compliance balloons, the success rate has increased to 90% in the latest review by Hamza et al. [7]. Many earlier reports did not include the balloon size or the pressures used. We use a 35-mm balloon, although in our series all patients were above 9 years of age. Hamza et al. [7] recommended balloon sizes as follows: 18 mm (<3 years, <10 kg); 30 mm (4–8 years, 10–20 kg); and 35 mm (>8 years, >20 kg). Wilkinson et al. [14] used balloons of increasing size starting at 23 mm and Hammond et al. [6] used 2–3 balloons in tandem to achieve better dilatation. As in our experience, most of these series have reported good results with PBD in children (Table 2).

Although adult series report oesophageal perforation in approximately 4% of PBDs, there are no documented perforations in the paediatric literature. The reported complications include symptomatic reflux, postoperative retrosternal pain, and prolonged epigastric pain [6–10, 14]. Advantages of PBD include ease of performance, shorter recovery time, and decreased hospital stay, resulting in decreased cost. It has better patient accept-

ability over an operation and if needed can be repeated. In several reviews multiple dilatations have resulted in increased overall success [6, 7]. Our results show that PBD can also be useful as a secondary treatment if an operation fails.

Based on the literature review and our results, we conclude that PBD of achalasia is an effective method of treatment in children of all age groups; it can be used safely as a primary or secondary treatment; and repeated dilatations should be considered before accepting it as a failure and proceeding to surgical myotomy.

References

1. Annese V, Basciani M, Lombardi G, Frusciante V, Simone P, Andriulli A, Vantrappen G (1996) Controlled trial of botulinum toxin injection versus placebo and pneumatic dilatation in achalasia. *Gastroenterology* 111: 1418–1424
2. Azizkhan RG, Tapper D, Eraklis A (1980) Achalasia in childhood: a 20-year experience. *J Pediatr Surg* 15: 452–456
3. Berquist WE, Byrne WJ, Ament ME, Fonkalsrud EW, Euler AR (1983) Achalasia: diagnosis, management, and clinical course in 16 children. *Pediatrics* 71: 798–805
4. Boyle JT, Cohen S, Watkins JB (1981) Successful treatment of achalasia in childhood by pneumatic dilatation. *J Pediatr* 99: 35–40
5. Emblem R, Stringer MD, Hall CM, Spitz L (1993) Current results of surgery for achalasia. *Arch Dis Child* 68: 749–751
6. Hammond PD, Moore DJ, Davidson GP, Davies RP (1997) Tandem balloon dilatation for childhood achalasia. *Pediatr Radiol* 27: 609–613
7. Hamza AF, Awad HA, Hussein O (1999) Cardiac achalasia in children. Dilatation or surgery? *Eur J Pediatr Surg* 9: 299–302
8. Nakayama DK, Shorter NA, Boyle JT, Watkins JB, O'Neill JA (1987) Pneumatic dilatation and operative treatment of achalasia in children. *J Pediatr Surg* 22: 619–622
9. Perisic VN, Scepanovic D, Radlovic N (1996) Nonoperative treatment of achalasia. *J Pediatr Gastroenterol Nutr* 22: 45–47
10. Samarasinghe DA, Nicholson GI, Hamilton I (1991) Dilatation of achalasia in the young: a useful alternative. *Gastrointest Endosc* 37: 568–569
11. Stark GA, Castell DO, Richter JE, Wu WC (1990) Prospective randomised comparison of Brown-McHardy and microvasive balloon dilators in treatment of achalasia. *Am J Gastroenterol* 85: 1322–1326
12. Swenson O, Oeconomopoulos CT (1961) Achalasia of the esophagus in children. *J Thorac Cardiovasc Surg* 41: 49–59
13. Vane DW, Cosby K, West K, Grosfeld JL (1988) Late results following esophagomyotomy in children with achalasia. *J Pediatr Surg* 23: 515–519
14. Wilkinson AG, Raine PAM, Fyfe AHB (1997) Pneumatic dilatation in childhood cardio-achalasia. *Pediatr Radiol* 27: 60–62