Case Report

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Website: www.afrjpaedsurg.org
DOI: 10.4103/0189-6725.181709
Quick Response Code:

Oesophageal pseudodiverticulum after foregut duplication cyst excision: Case report and literature review

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ABSTRACT

Oesophageal pseudodiverticula rarely occur after excision of benign oesophageal neoplasms. While management and outcomes have been reported in the adult leiomyoma literature, sparse data exist on the occurrence and management of pseudodiverticula after foregut duplication cyst excision. We discuss our experience with a paediatric patient and review relevant literature regarding operative techniques and surgical outcomes.

Key words: Bronchogenic cyst, foregut cyst, oesophageal duplication, oesophageal pseudodiverticulum

INTRODUCTION

Foregut duplication cysts are rare congenital masses that require surgical excision in children due to their risk of becoming symptomatic.[1] An infrequent complication after cyst excision is the bulging of mucosa though the muscle fibres, forming a pseudodiverticulum. Only three other cases of pseudodiverticula after duplication cyst excision have been reported in literature.[1-3] We report a paediatric case of pseudodiverticula formation at the site of previous foregut duplication cyst excision and review the literature on the management of this problem.

CASE REPORT

A 4-year-old girl presented with intermittent abdominal pain, constipation and occasional chest pain for 6 weeks. Computed tomography (CT) scan of the abdomen and pelvis showed a right posterior mediastinal cystic structure that was incompletely visualised. The patient was referred to paediatric surgery and a contrasted CT of the chest revealed a discrete 2.6 cm × 2.3 cm right posterior mediastinal cyst adjacent to the oesophagus, most likely representing a foregut duplication cyst [Figure 1]. She was scheduled for elective thoracoscopic resection with oesophagoscopy.

In the operating room (OR), the cyst was dissected bluntly from within the muscular layer of the oesophagus [Figure 2]. No communication between the cyst and oesophageal mucosa was noted on oesophagoscopy. Injury to the mucosa was ruled out by a negative air leak test after endoscopic insufflation. The patient had an uneventful post-operative course and pathology confirmed the presence of a foregut cyst of developmental origin. Histologic features included a respiratory epithelial lining and underlying smooth muscle [Figure 3]. Absence of seromucinous glands and cartilaginous plates excluded bronchogenic cyst.

At 2-month follow-up, the patient complained of chest pain while eating solids. An upper gastrointestinal contrast evaluation with barium revealed an outpouching of the distal oesophagus at the site of cyst excision, suggesting the development of an oesophageal diverticulum or pseudodiverticulum [Figure 4].

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Cite this article as: Bobanga ID, Redline RW, DeRoss AL. Oesophageal pseudodiverticulum after foregut duplication cyst excision: Case report and literature review. Afr J Paediatr Surg 2016;13:50-3.

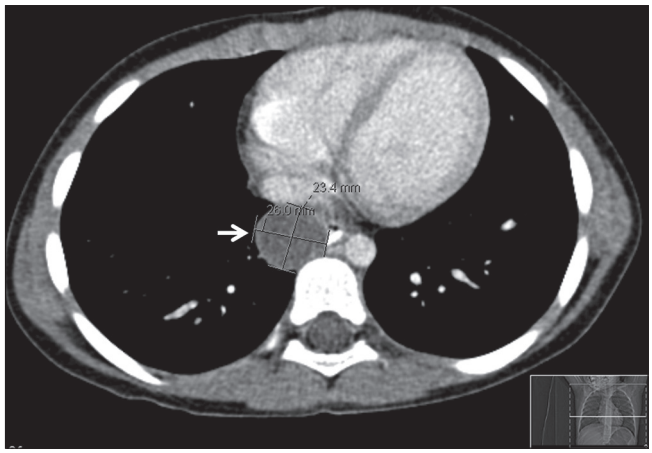

Figure 1: Computed Tomography showing a right posterior mediastinal cyst adjacent to the oesophagus (white arrow) (original)

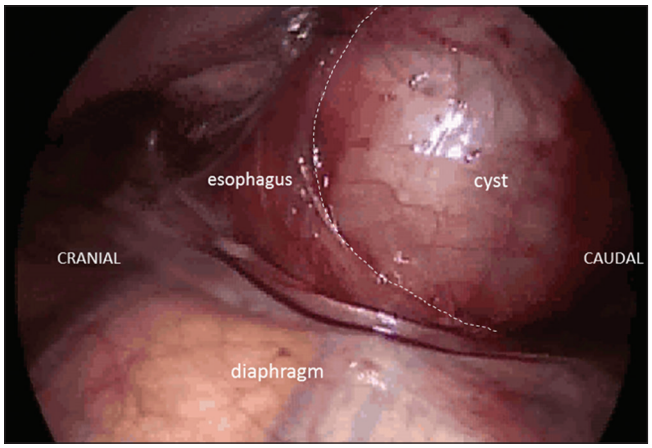

Figure 2: Foregut duplication cyst prior to enucleation (original)

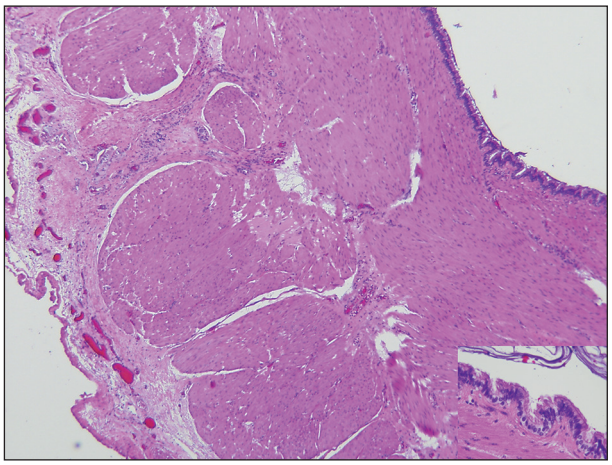

Figure 3: Cross-section of foregut cyst wall showing respiratory epithelium (right) with underlying smooth muscle and fibroconnective tissue serosa in the absence of seromucinous glands or cartilaginous plates (H and E, ×40). Inset: Ciliated respiratory epithelium with focal mucus secretion (H and E, ×400) (original)

The child was taken to the OR 3 months after her first surgery for diverticulum resection via a laparoscopic transabdominal approach combined with oesophagoscopy. Dissection was challenging due to scarring at the previous surgical site. It was decided to imbricate the muscle around the diverticulum instead of resecting it, due to difficulty determining its boarders and concern for causing an oesophagotomy given the surrounding scar tissue. Interrupted sutures were used to imbricate the intact muscle edges of the oesophagus around the diverticulum over a 40 French oesophageal bougie. There was no air leak or bulging at insufflation via oesophagoscopy. Because it was unclear if the aetiology of this lesion was that of a pulsion diverticulum, a myotomy below the level of the previous surgery was done on the lower anterior oesophagus and carried down onto the upper stomach, along with an anterior fundoplication of the stomach to prevent future reflux.

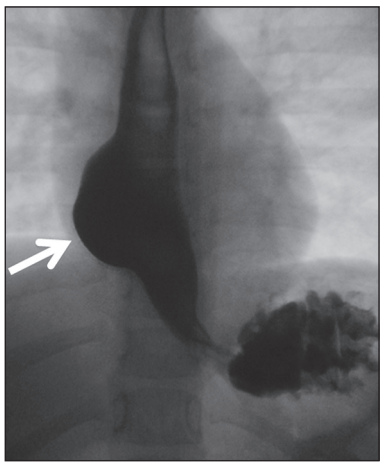

Figure 4: Contrast swallow study showing outpouching of the distal oesophagus representing a pseudodiverticulum (white arrow) (original)

Post-operative swallow evaluation showed no extravasation, but a small outpouching at the surgical site was still noted, with mild narrowing at the gastroesophageal junction. The patient was doing well and tolerated a diet with mild dysphagia. The patient continued to have mild intermittent dysphagia and some chest pain with solid foods in the few weeks after surgery. Subsequently, the patient underwent EGD dilation of the distal oesophagus and a gastrostomy tube placement. Her symptoms were minimal over the next 5 months after two dilations, but some dysphagia did recur, so it was decided that the diverticulum required definitive repair. Approximately, 1 year after initial surgery, the child underwent right posterolateral thoracotomy and diverticulum resection. On long-term follow-up, 2 years after her last surgery, the patient was doing well, tolerating an oral diet and growing appropriately.

DISCUSSION

Oesophageal duplication cysts are part of the spectrum of congenital foregut duplication cysts

often found in the mediastinum.[1] They can be classified as oesophageal duplications if they are close to the oesophageal wall, are covered by two muscle layers, and if the lining is squamous columnar, cuboid, pseudostratified or ciliated epithelium[2] while bronchogenic cysts are characterised by ciliated columnar epithelium and by the presence of cartilage. While they are rare, oesophageal duplications represent up to 20% of all alimentary tract duplications[4] and 10% of all mediastinal tumours in children,[3] with a prevalence of 1:8200 reported in autopsy studies.[5] Embryologically, they are believed to develop due to a defect of vacuolisation of the developing oesophagus.[6,7] Some authors advocate the general nomenclature of foregut cyst for both 'bronchogenic cyst' and 'oesophageal duplication' due to the many similarities they share: Common embryologic origin, anatomic proximity, histologic similarity and overlapping clinical features.[8,9] Complete excision is recommended for all paediatric cases of foregut duplication cysts, as most will develop symptoms and result in a higher rate of intraoperative complications.[1-3,6,8] Like other benign oesophageal lesions, most intramural foregut duplication cysts are excised via video-assisted thoracoscopic surgery, without opening the oesophageal mucosa.[2,3,6] Particular attention must be paid to maintaining mucosal integrity, preserving the oesophageal muscle layer, and identifying and preserving both vagal nerves.[2]

A pseudodiverticulum, or outpouching of the oesophageal mucosa at the site of cyst excision, is a rare complication after foregut cyst excision. Until date, only three other cases have been reported in the literature [Table 1].[1-3] Oesophageal pseudodiverticula have more commonly been reported after leiomyoma enucleation[10-12] and achalasia treatment at the proximal end of myotomy not covered by fundoplication.[13] Unusual cases have been reported following high-dose intracavitary irradiation for oesophageal cancer[14] and alcohol abuse and chronic esophagitis.[15]

The need to repair the oesophageal muscle wall after excision of intramural oesophageal duplication cysts is controversial.[1] Several case reports that encountered pseudodiverticulum formation after oesophageal duplication or leiomyoma excision advocate for interrupted suture closure of the muscular defect[3,6,10-12] to preserve the propulsive activity of the oesophagus. However, pseudodiverticula have been reported to develop in a few cases in which the muscular defect was repaired after duplication cyst[2] and leiomyoma excision.[12]

The management of an oesophageal pseudodiverticulum that occurs after the excision of a benign oesophageal neoplasm varies based on patient symptoms. Those pseudodiverticula found incidentally on routine barium oesophagram may only need monitoring. In the case discussed by Bratu *et al.*, the pseudodiverticulum was of 'questionable significance' and resolved spontaneously.[1] Perger *et al.* also described a case that was incidentally discovered, remained asymptomatic and required continued patient follow-up.[3] In patients with symptoms of dysphagia, dyspepsia or regurgitation, it is important to evaluate further them for other potentially contributing oesophageal disorders such as gastrointestinal reflux, incompetent lower oesophageal sphincter or a concomitant oesophageal motility disorder.

The need for surgical intervention varies based on the size of the diverticulum and the presence and severity of symptoms. Our patient had a relatively large and symptomatic diverticulum requiring surgical intervention. In literature discussion of pseudodiverticula occurring after leiomyoma excision, several cases required thoracotomy and diverticulectomy for dysphagia.[10,11] The need for myotomy and fundoplication is dictated based on suspicion or findings of an oesophageal motility disorder and risk for developing reflux. While the closure of the muscular wall around a bulging mucosa may seem to be an option, it may not prevent

Table 1: Listing of reported cases of oesophageal pseudodiverticula after intramural foregut duplication cyst excision

References	Age group	Symptoms	Mode of dx	Findings	Treatment	Outcome
Bratu *et al*.[1]	Child	Unknown	Unknown	Unknown	None	Resolved spontaneously
Cioffi *et al*.[2]	Adult	Dyspepsia, regurgitation, vomiting	Barium oesophagram Manometry pH probe Scintigraphy	Pseudodiverticulum at previous resection site, incompetent LES, GERD	Histamine-2 blocker, Cisapride	Good results
Perger *et al*.[3]	Child (age 8)	None	Barium oesophagram	Pseudodiverticulum at site of cyst excision	None	Remained asymptomatic
This paper	Child (age 4)	Dysphagia	Barium oesophagram	Pseudodiverticulum at site of cyst excision	Resection via thoracotomy	Tolerating oral diet

LES: Lower oesophageal sphincter; GERD: Gastroesophageal reflux disease

pseudodiverticulum recurrence and could cause stricturing of the oesophagus at an already scarred and inflamed area, as in our case. Thus, definite resection of the pseudodiverticulum is recommended.

CONCLUSION

Foregut duplication cysts require surgical resection in children. The authors believe that the muscular layer should be approximated with interrupted sutures after excision to decrease the risk of pseudodiverticulum formation. If a pseudodiverticulum occurs and is symptomatic, definitive treatment of oesophageal pseudodiverticulum is surgical resection.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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