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Epiphrenic esophageal diverticulum in an adolescent with a history of a Nissen fundoplication: A case report

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ABSTRACT

Epiphrenic esophageal diverticuli (EED) are exceedingly rare in children. While esophageal dysmotility is often associated with this diagnosis in adults, the few reports in children implicate retained foreign bodies as the cause. The patient presented here is an 18 year-old female with a distant history of a Nissen fundoplication who developed dysphagia, gastroesophageal reflux, and weight loss, and was found to have an EED. Her symptoms completely resolved following laparoscopic diverticulectomy and hiatal hernia repair. Though the exact etiology of her EED remained unclear, it may have been related to her fundoplication. This potential late complication may be seen more frequently as a large number of children with a history of fundoplication are reaching adulthood.

Epiphrenic esophageal diverticula (EED) are uncommon and have rarely been described in the children [1]. In the adult population, the majority are related to esophageal dysmotility or are associated with achalasia or hiatal hernias. They are generally believed to result from increasing intraluminal pressure of the distal esophagus just superior to the lower esophageal sphincter. Right-sided diverticuli have been more commonly reported. Generally, these are classified as false diverticuli; they form as submucosa and mucosa herniates focally through the muscularis propria. In the few cases reported in children, the diverticulum was thought to result from a retained foreign body [2,3]. The present report documents a case of an EED in an adolescent with a history of a Nissen fundoplication as a toddler.

1. Case

The patient was an 18 year-old female at the time of presentation who had a history of severe epilepsy and who underwent a Nissen fundoplication for significant gastro-esophageal reflux at 2 years of age. She complained of a 3 year history of increasing dysphagia, frequent regurgitation, and early satiety with associated substernal chest pain and weight loss.

A contrast esophagram/upper gastrointestinal series (UGI) revealed an EED originating off of the right lateral aspect of the distal esophagus. The EED was just above the level of the prior Nissen fundoplication (Fig. 1), from which contrast did not clear. Esophagoduodenoscopy demonstrated the diverticulum with normal esophageal mucosa. Esophageal manometry showed appropriate relaxation at the gastroesophageal junction (GEJ) and normal contractility.

Despite initial treatment with proton pump inhibitors, her symptoms persisted over 8 months, and a repeat UGI showed no change. At this point she was referred for surgical evaluation and a laparoscopic resection of the diverticulum was planned. At the time of the operation, no evidence of active distal esophageal obstruction was found; the previous fundoplication was no longer in place and the lumen was widely patent on intraoperative EGD. The diverticulum was evident on EGD which is depicted (Fig. 2). Upon esophageal mobilization, a 2 cm right-sided posterolateral diverticulum was encountered just above the GEJ (Fig. 3) in a position where the prior sutures for the Nissen may have been placed. The diverticulum was stapled along its base using an endo GIA stapler with a size 52 bougie in place under laparoscopic visualization (Fig. 4). She also had a large hiatal hernia that was closed primarily (Fig. 5). We confirmed no staple line leak and no narrowing of the esophagus endoscopically (Fig. 6). An esophagram on

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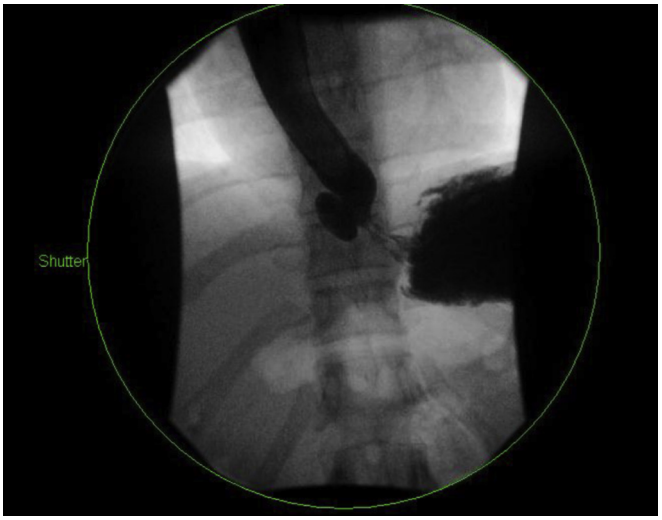

Fig. 1. Preoperative esophagram demonstrates epiphrenic diverticulum.

post-operative day 1 showed a patent lumen without any narrowing or leak (Fig. 7).

The remainder of her post-operative course was uneventful. Despite persistent post-prandial emesis at one month, by the second month all of her symptoms had resolved and she had re-gained weight appropriately. Repeat UGI demonstrated a normal appearing GEJ without evidence of reflux. She remained asymptomatic six months post-operatively.

2. Discussion

Epiphrenic esophageal diverticula (EED) are rare with an incidence of 1:500,000/year in the adult population [1,4]. They are likely caused by a combination of a motility disorder (achalasia, diffuse esophageal spasm, etc), creating a relative distal obstruction (or an atomic obstruction) coupled with a weakness in the muscularis propria [1]. Symptoms are related to the location of the diverticulum and include dysphagia, odynophagia, regurgitation, chest pain, and aspiration [5]. Diagnosis is usually made via a barium swallow with supplemental tests including an upper endoscopy to rule out malignancy and esophageal manometry to delineate any underling motility disorder [1,5–7].

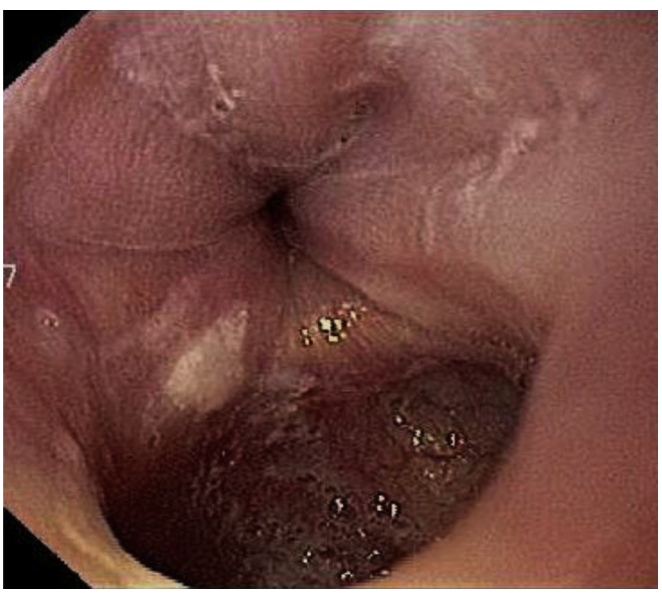

Fig. 2. Endoscopic demonstration of the epiphrenic diverticulum.

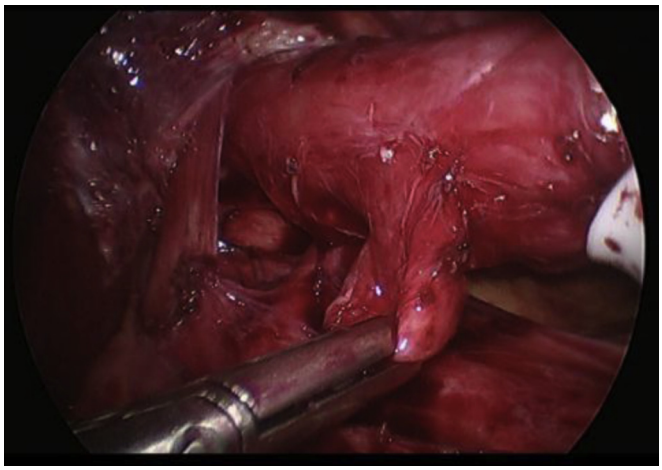

Fig. 3. Intraoperative view of right postero-lateral diverticulum (held by grasper).

Most mildly symptomatic diverticula are initially treated with H_2-blockers or proton pump inhibitors [1,6]. Surgery is reserved for those patients with large diverticula, severe dysphagia, or a significant risk of aspiration [1–3,5,6,8–10]. In previously described cases, diverticulectomy, myotomy, and fundoplication were completed laparoscopically [1,5,6,11–14]. For symptomatic patients unwilling or unfit to undergo an operation, botulinum toxin injections or endoscopic esophageal dilation can relieve the distal obstruction and improve symptoms [1]. Laparoscopic diverticulectomy and myotomy with fundoplication has been successful in adult case series [13,15].

Most case reports in children describe an EED associated with an impacted foreign body [2,3]. The pathophysiology in the patient presented here remains somewhat unclear. With normal manometry and no intraluminal foreign body, it is possible that the prior fundoplication caused a relative distal obstruction, similar to some documented cases in adults [5,8], though the wrap was not intact at the time of laparoscopy. It is also possible that the diverticulum resulted from traction on the esophagus from a fundoplication

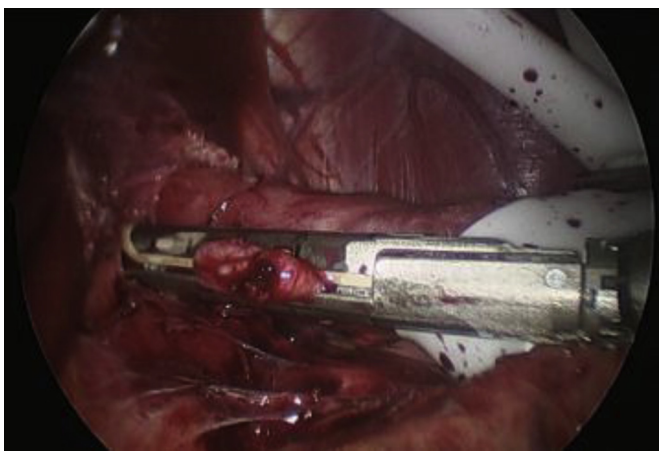

Fig. 4. Dividing diverticulum across its base with endo-GIA stapler.

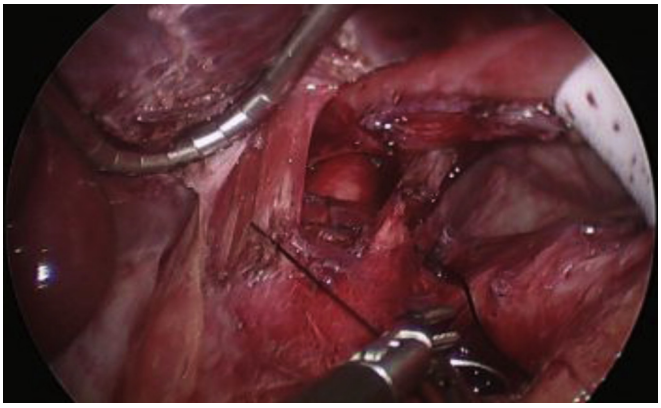

Fig. 5. Primary closure of hiatal hernia.

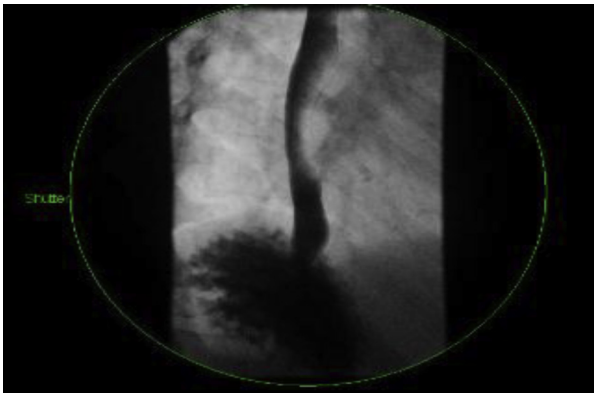

Fig. 7. Post-operative esophagram demonstrates resolution of diverticulum with normal luminal diameter and no hiatal hernia or leak.

stitch. There was no apparent relationship of the patient's epilepsy or pharmacologic therapy to her EED.

Given the substantial number of fundoplications currently performed in children in the US, the findings in his patient may be seen more frequently as the contemporary cohort ages. The ideal treatment in these particular circumstances (and in particular, the role of repeat fundoplication or myotomy) remains unclear. In this patient, a laparoscopic approach provided excellent visualization of the distal esophagus. Here, hiatal hernia repair without fundoplication was effective in ameliorating her reflux. Given normal preoperative manometry and the absence of hypertrophic muscle typically seen with achalasia, myotomy was not performed, consistent with the limited data guiding therapy in these circumstances [16].

3. Conclusion

Epiphrenic esophageal diverticula are uncommon and are mainly described in adult patients. They most often occur secondary to a functional obstruction and result in symptoms including dysphagia, chest pain, and regurgitation. This adolescent patient's diverticulum may have been related to previous fundoplication and thus may represent one sequelae of this commonly performed operation. As in adults, laparoscopic diverticulectomy with or without myotomy and/or repeat fundoplication may be appropriate, though more data are needed to identify the optimal treatment strategy.

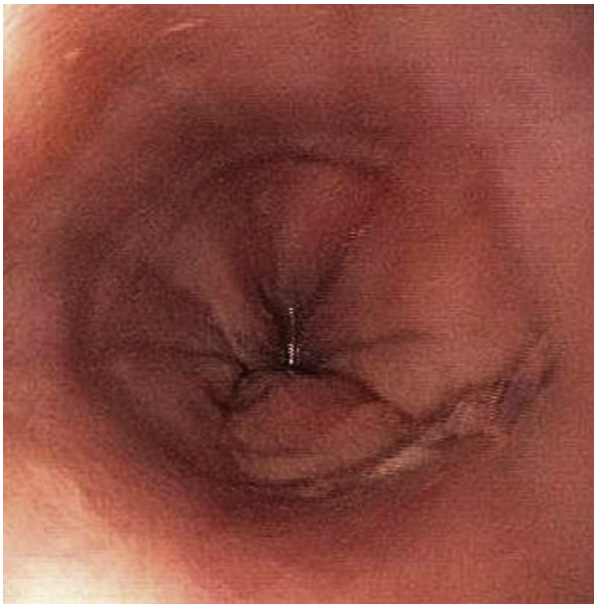

Fig. 6. Endoscopic demonstration of repair.

Disclosure statement

The authors have received no financial support, and do not have any potential conflicts of interest to report.

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