

Case report

Esophageal transverse chord following repair of esophageal atresia

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SUMMARY

Within a period of one year two children were treated for a transverse intraluminal chord of the esophagus, which developed following repair of the esophageal atresia and peripheral tracheo-esophageal fistula. The chord was located at the level of the anastomosis in the upper third of the esophagus. In the first child, the chord was partially obstructing the esophageal lumen, causing severe dysphagia and requiring dilatation. In the second child, the chord was causing only mild stenosis and minimal symptoms. There were no available instruments suitable for the transection of the chord and a special snare was improvised on site. The etiology of this singular formation is unknown. A possible explanation for this formation is discussed.

Key words: Esophageal atresia, stenosis, transverse chord

INTRODUCTION

Esophageal stenosis following correction of esophageal atresia is encountered with a frequency of 30-40%.^{1,2} However the formation of a transverse intraluminal esophageal chord causing symptoms is an exceptional occurrence and only one reference³ to a comparable finding could be found. This is the report of two such children with stenotic symptoms, who were referred for dilatation. The etiology of this formation is unknown and no such parallel could be found in the literature. The findings are described and the possible etiology, as well as contributing factors, are discussed.

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SUBJECTS

Case 1

A three-year-old boy was referred to our department for esophageal stenosis following repair of the esophageal atresia and tracheo-esophageal fistula. The operative repair was reported to have been straight forward. During endoscopy for dilatation, a transverse chord was seen at the level of the anastomosis (Fig. 1), dividing the lumen in two unequal parts, and causing severe obstruction. The endoscope could not be forced through the larger opening. Therefore, only dilatation was performed, which cured the stenosis. Due to the small caliber of the working channel of the pediatric endoscope, no suitable instrument could be found for the transection of the chord. At a subsequent procedure, a wire loop was improvised by feeding a guide wire through one side of the chord and retrieving it through the other. This was then covered with the sheath of a normal polypectomy snare and the chord was severed with the diathermy current. At a later follow-up, the severed stump regressed

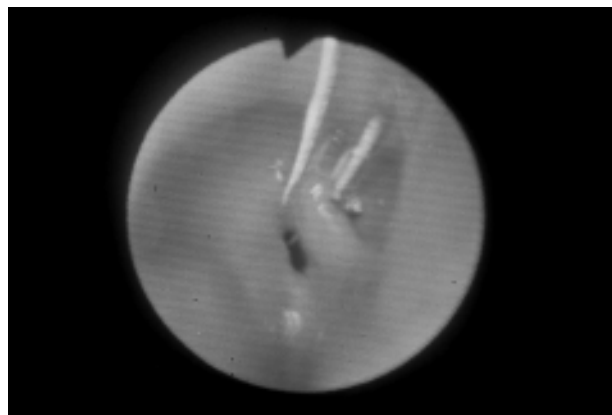


Figure 1. The thick transverse chord of case 1. The esophageal lumen divided in two unequal sections and obstructed. Two separate guide wires have been introduced through the two sections. The larger section was then dilated.

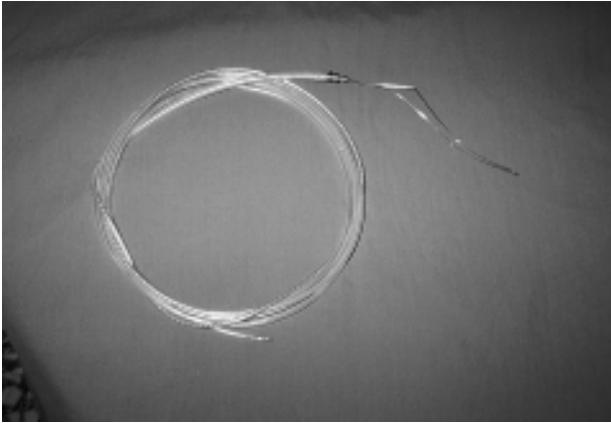


Figure 2. Photograph of the improvised wire loop. The arrow shows the actual loop.

completely. There was only mild esophagitis at the gastroesophageal junction, requiring mild antireflux therapy. The stenosis did not recur.

Case 2

A four-year-old boy was referred for mild dysphagia and gastroesophageal reflux following repair of the esophageal atresia and tracheo-esophageal fistula. During endoscopy a transverse chord was seen at the level of the anastomosis, dividing the lumen in two sections and causing mild obstruction. The pediatric endoscope could be forced through the stenosis with minimal effort and a balloon dilatation was performed. The above mentioned snare was again constructed on site and the chord transected. The remaining stump was then resected with a normal polypectomy snare and sent for histologic examination, which revealed changes of mild esophagitis. Further passage of the endoscope revealed moderate esophagitis at the gastroesophageal junction. The stenosis did not recur and reflux was treated with antireflux therapy.

DISCUSSION

Esophageal stenosis following repair of the esophageal atresia is an unwelcome but well described complication in children. It is encountered with a frequency of 30-40%.^{1,2,4} By contrast, the formation of a transverse intraluminal esophageal chord, causing symptoms, is an exceptional occurrence. The mechanism of formation of such a chord cannot be easily explained. No parallel case could be found in the literature, except one referred to in Czech, of which only the summary could be read. The description is consistent with a similar formation. The most likely

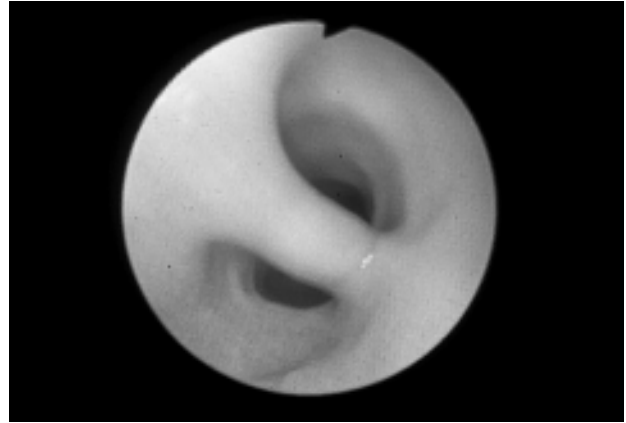


Figure 3. The transverse Chord of case 1 dividing the esophageal lumen in two unequal parts.

cause of this chord was a granuloma forming a bridge across the lumen, which was then intensified and thickened in the postoperative course by gastroesophageal reflux. The formation of a fibrin bridge could facilitate this development. Another, less likely mechanism may be the initial formation of such a granuloma around an absorbable suture protruding across the lumen.

The endoscopic maneuver invented for the transection is thought to be quite resourceful and can be used in similar situations. In the first case, the dilatation was performed to alleviate the stenosis and provide time to find or design a suitable instrument for the transection. It also allowed the passage of the endoscope for the recovery of the tip of the guide wire and the formation of the loop. In the second case this was not necessary and excision could be performed during the same session. The histology, which disclosed mild esophagitis, is consistent with the formation of polypoid lesions as a reaction to gastroesophageal reflux^{5,6}. Both children had esophagitis in addition to the changes found at the site of the anastomosis and chord formation, as is common in children with esophageal atresia.²

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