Anastomosis instead of resection: An unusual approach for the treatment of a cervical esophageal duplication cyst

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A R T I C L E   I N F O

Article history:
Received 25 October 2013
Received in revised form 29 January 2014
Accepted 30 January 2014

Key words:
Esophageal cyst
Cervical approach
Surgical stapler

A B S T R A C T

We report of a 5 year old boy with severe dysphagia. He was operated upon a tubular esophageal duplication located in the upper right mediastinum. Due to the existing severe esophageal stricture the malformation was not resected but unified with the originary lumen of the esophagus using a stapled anastomosis. Access was chosen via low collar incision. During follow up no further stricture occurred. The resection of esophageal duplications is the therapy of choice. In our case, union with the originary organ was performed due to its distinctive anatomical conditions. Besides the collar incision, as an access to an upper mediastinal mass, is a rather uncommon but safe technique to manage such malformations in children. Follow up investigations to eliminate formation of malignancies are recommended. The operative technique in this case is uncommon and has not been described in esophageal duplications yet. It should be considered as an operative strategy for specific anatomical conditions.

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Esophageal duplication cysts are rare, mostly asymptomatic, congenital anomalies. Most of them are located in the distal thoracic part of the esophagus. Localization in the upper part of the esophagus is rather unusual and is a challenge to pediatric surgeon. Symptoms are caused by the compressive effect on mediastinal structures or due to local infection [1–4]. Localization in the upper part of the esophagus can lead to dyspnea, particularly in the infant [2]. Dysphagia, as in our case, represents the most common symptom in the older child or adolescent [1]. Surgical resection is indicated in all symptomatic lesions and has to be considered for asymptomatic cysts to prevent complications [2,4–6].

1. Case report

A five year old boy was referred to our hospital with a history of dysphagia since birth. He regurgitated while swallowing lumpy food so an esophageal stricture was suspected. The child was hospitalized in a different institution where an endoscopy and an MRI were carried out. The endoscopy showed an esophageal stricture of unknown origin. Biopsies showed inflammation of the mucosa but the etiology remained unresolved. An MRI-study showed a mass in the upper dorsal mediastinum. The patient was then transferred to the department of radiology of our institution. An additionally conducted CT-scan (Fig. 1) verified the upper esophageal stricture with a shift of the trachea to the right caused by a mass of the upper mediastinum. No concomitant malformations were seen in other sites. The radiological aspects lead to the suspect of a foregut duplication cyst or a lyomyoma. A contrast study showed a double lumen of the esophagus (Fig. 2), so that the diagnosis of a tubular esophageal duplication was most likely. Thus an intervention under general anesthesia was arranged. Performing inversion in esophagotomy (Fig. 3) a septum in the distal esophagus was detected and the diagnosis of a tubular duplication cyst of the esophagus with distal communication was verified.

To clarify anatomy of the proximal esophageal wall in the cervicothoracic junction, where the duplication was assumed, the operation was planned via a low transverse collar (Kocher’s) incision. Identifying the trachea which was put aside with hooks, the esophagus could easily be exposed and armed. The esophagus appeared slender in the very proximal part and thicker in the subsequent (duplicated) part in the thoracic inlet. The outer esophageal wall appeared homogenous. This finding was compatible with a tubular esophageal duplication. In the esophagus the nasogastric tube was palpable. The esophagus was opened and the gastric tube was identified. Since the right sided esophagus appeared to be thick walled, a second esophagotomy on the right side of the esophagus was carried out. Thus a second lumen could...
be identified. A further probe was introduced easily for the length of the distal esophagus (Fig. 4). Both, esophagus and duplication, were dilated. Then a stapler was introduced and the two structures were unified. Intraoperative endoscopy showed a continuous, normal wide esophagus lined with normal mucosa without signs of leakage. Esophagotomy was then closed using an absorbable running suture. A drain was left in the operative field.

After extubation on the day of surgery, the boy showed severe emphysema of the collar skin indicating anastomotic insufficiency. The emphysema and signs of inflammation declined under addi-
tional antibiotic therapy with cefazolin and meropenem. On day 6 after operation the patient showed salivation via the drain which was fixed to skin. An esophagocutaneous fistula was confirmed by a contrast study. Since the patient showed no signs of general infection we decided on an observant therapy. The fistula closed keeping the patient nil per os 12 days after the primary operation.

In a subsequent contrast study we could not verify any remaining fistula or stenosis.

A gastroscopy showed a well healed, normal configured esophagus with no stricture or residual septum at four weeks postoperatively. 4 months postoperatively the boy gained 4 kilos of weight and had neither dysphagia nor regurgitation.

A contrast study (Fig. 5) showed a regular esophagus with a slight declination to the left but no signs of a stricture. Follow up endoscopies are planned once a year screening for malignancies.

2. Discussion

Esophageal duplications — tubular or cystic — constitute a relatively seldom congenital anomaly, which is thought to arise as an error of foregut budding during the third to sixth week of gestation [2]. Its incidence is approximately 1:8200 with predominance of male sex (2:1) [7]. Associated anomalies are spinal malformations or additional duplications of the alimentary tract [4].
Terminology is still under discussion considering either the organ of origin or histology as the focus of classification [4]. Generally a differentiation of cystic malformations of foregut into esophageal duplication cyst, neuroenteric cyst and bronchogenic cyst is stated [2]. There are two types of esophageal duplication cysts described: cystic and tubular.

Most esophageal duplications are asymptomatic [8] even though symptoms vary on their localization and origin [3,9]. Besides infection or rupture, cysts become symptomatic by local compression on mediastinal or abdominal structures [1]. There are few reports that postulate a “wait and see” strategy or an “internal drainage” between duplication cyst and the gastrointestinal tract in case of delicate anatomy. In the literature, resection of gastrointestinal duplication cysts, even in asymptomatic cases, is recommended due to the risk for infection, bleeding, rupture, malignancy and its space occupying effect [2,4–6].

Operation via collar incision is described as an access for resection of upper mediastinal masses but is rarely described for duplication cysts [2,3,5,6]. We decided on a cervical approach with the opportunity of a sternotomy appearing to be the safest and less extensive access to this mass.

Since severe dysphagia was the main symptom in this patient and endoscopy showed a communication of the duplication to the healthy esophagus unification of the two lumina using a stapler was the technique of choice to realize a sufficient width of the upper part of the esophagus. In addition the duplication in our patient was part of the regular esophageal wall. We therefore do expect a normal function of the esophagus after unification. The patient showed no signs of dysphagia and does not require any further therapy up to now. Routine follow up endoscopies are planned to screen for malignant transformation or recurrence of stricture.

3. Conclusion

We herein describe a case of esophageal duplication cyst presenting with the main symptom of severe dysphagia. Correction was performed by a limited access to these benign lesions via cervical approach. To improve the clinical signs of the esophageal stricture, resection of the lesion was avoided in favor to unifying the lumina via staple anastomosis.

Conflicts of interest and source of funding
None.

References

