CASE REPORTS

Tubular duplication of the esophagus

Fahad Alenazi[1], Ebtisam Alenazi[2], Ziad Asaad[2], Khaled Alqoaer*[2]

[1] College of Medicine, Tabuk University, Saudi Arabia
[2] Pediatric Department, King Salman Armed Forces Hospital, Tabuk, Saudi Arabia

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ABSTRACT

Esophageal duplication (ED) is a rare congenital anomaly, representing 10%-15% of all foregut duplications. Neonates may present with respiratory distress, while older children usually present with dysphagia. We report here a rare case of tubular duplication of the esophagus presenting with dysphagia in a 12-year-old Saudi boy.

Key Words: Esophageal duplication, Esophageal malformation, Dysphagia

1. INTRODUCTION

Esophageal duplication is a rare congenital anomaly, representing 10%-15% of all foregut duplications.[1] A cyst or a fistula can form from herniation of endodermal gut through a split that occurs in the notochord that is present from 3rd week gestation. It represents either simple epithelial lined cysts or true esophageal duplication bounded by muscularis mucosa, submucosa and muscularis externa that can appear as diverticula or as a tubular malformation. Neonates can present with respiratory distress while older children usually presents with dysphagia.[2] The incidence of congenital esophageal duplication is estimated to be 1:8,200, with male sex predominance 2:1.[3] Esophageal duplication is divided into three types: cystic (the most common type), tubular and diverticular.[4]

In this study, we report a rare case of tubular duplication of the esophagus, in a 12-year-old boy presented with dysphagia.

2. CASE REPORT

A 12-year-old Saudi boy presented to our hospital emergency center with upper respiratory tract infection and acute gastroenteritis. He gave history of intermittent dysphagia with both, liquid and solid diets for more than five years. There was no history of cough or chocking during swallowing, and the rest of his medical history was noncontributory. Physical examination and routine blood tests were unremarkable.

Gastrografin esophagographgy showed a well-defined filling defect measuring about 6 cm in vertical diameter and arising from the proximal left side of the esophagus displacing it to the left side (see Figure 1). Contrast enhanced computed tomography of the chest revealed additional tract of the esophagus with a blind end that measured around six cm in its craniocaudal dimension deviating the esophagus to the left side. Upper GI endoscopy showed two esophageal lumens located about 15 cm from the incisors (see Figure 2). The endoscope could be passed through both lumens; small ulcerations and edematous mucosa were noted in the duplicated segment which was blind ended and measuring about six cm in length. The patient was referred for surgical repair.

Tubular esophageal duplication in children was first described by Granelli et al. in 1983.[16] Few other cases were

*Correspondence: Khaled Alqoaer; Email: dralquaer@hotmail.com; Address: Pediatric Department, King Salman Armed Forces Hospital, Tabuk, Saudi Arabia.

reported afterword and summarized in Table 1. Esophageal duplications represent 10%-20% of foregut duplications.[5] Tubular duplication of esophagus is rare and is much less common than cystic duplication of the foregut.[6] Unlike cystic duplications, tubular duplications usually communicate with the normal esophagus.[7–9] Tubular esophageal duplication can be associated with other anomalies such ileal duplication cyst and bronchogenic cyst.[10,11] However, in this case the additional esophagus was blind ended and there was no accompanied anomaly. The most commonly affected sites of tubular duplication are the mid and lower third of the esophagus,[12] however, the duplication in this patient involved the proximal esophagus. The lumen of esophageal duplication may show ectopic pancreatic tissue,[13] gastric tissue,[14] or sometimes malignant tissue such as adenosquamous carcinoma which was reported in 32-year-old man who presented with dysphagia.[15] Esophageal duplication can be asymptomatic and discovered incidentally.[16] However, the usual presentation of esophageal duplication is dysphagia and chest pain.[7,17,18] Dyspnea has been reported in one case of tubular esophageal duplication and it was associated with bronchogenic cyst and pericardial defect.[11] Anorexia and weight loss have been reported in patient who had developed a malignant transformation of esophageal duplication.[19] Persistent wheezing can be a manifestation of tubular esophageal duplication.[20] Hemoptysis also has been reported to be a feature of esophageal duplication containing ectopic gastric tissue in neonate.[21]

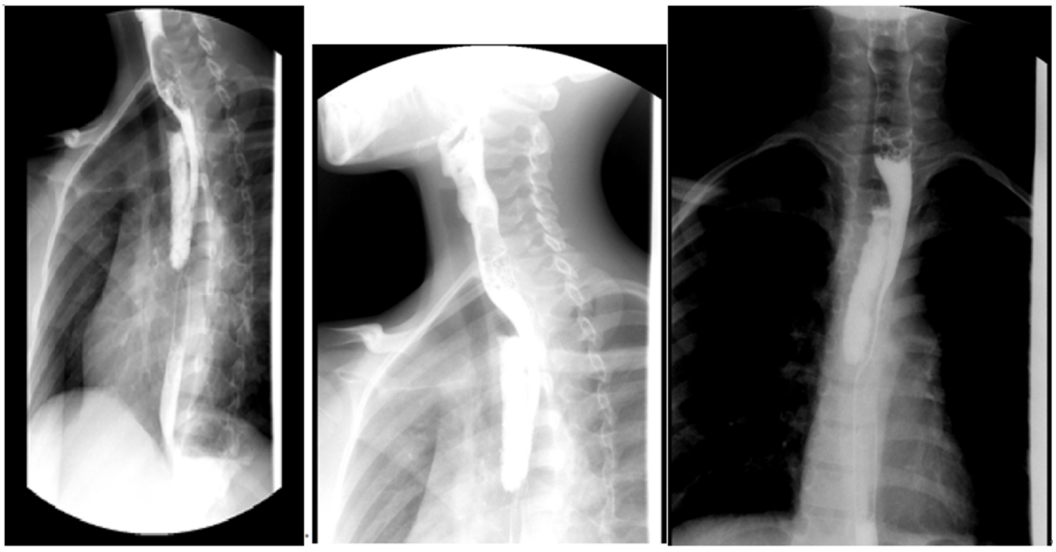

Figure 1. Gastrografin esophagographgy showing an elongated, well defined pouch measuring about 6 cm in the proximal esophagus displacing it to the left

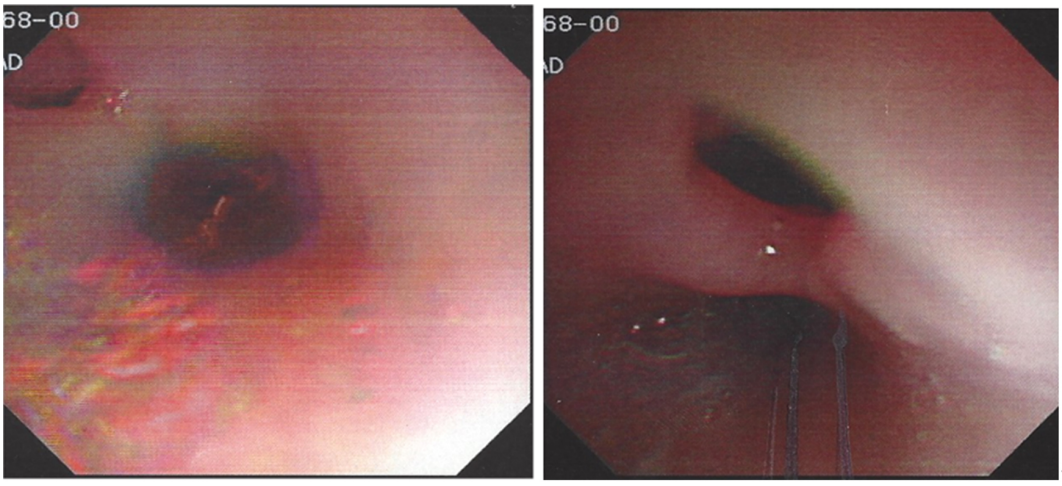

Figure 2. Upper GI endoscopy showing two esophageal lumens located about 15 cm from the incisors

Table 1. Reported cases of tubular esophageal dilatation in children (age < 19 years)

NO.	Age	Sex	Clinical features	Radiological finding	Surgical finding	References
1	11 years	Male	Chest pain Dysphagia, Cough and Fever	Double esophageal lumen with Intraluminal bridge	NA	17
2	2 days	Male	Excessive Salivation + Intolerance To feeds	Tubular esophageal duplication from cervical region to diaphragm	NA	23
3	3 days	Male	Chocking and cyanosis	Proximal tubular esophageal duplication	NA	8
4	10 months	Male	Cough and persistent wheezing	Hypodense oval mass in posterior mediastinum displacing esophagus to the right	Large cystic mass in posterior mediastinum	20
5	14 years	Male	Dysphagia and retrosternal chest pain	Cystic duplication with upper esophageal stricture	Two esophageal lumens with thick intraluminal bridge	18
6	Newborn	Male	NA	Esophageal atresia and tubular non communicating esophageal duplication	NA	24
7	22 months		Congenital stridor.	Esophageal cervical duplication	NA	25
8	6 years	Male	Dysphagia to solid	Tubular duplication of esophagus	NA	26
9	neonate	Male	Respiratory distress and vomiting	Total tubular esophageal duplication	NA	27
10	16 months	Male	Dysphagia to solid, stridor and mass underneath his tongue	Intraluminal filling defect from upper to middle 1/3 of esophagus	Intraluminal tubular esophageal duplication	28
11	18 years	Male	Dry cough and mild dyspnea	Cystic mass with air fluid level connected to esophagus in middle mediastinum and left pericardial defect	The pleural mass invested accessory lobe connected with esophageal wall by tubular structure	11
12	1 month old		NA	Tubular duplication of the esophagus		29
13	18 weeks gestation	Male	NA	Tubular cystic mass in posterior mediastinum and multiple cystic masses in abdomen	Esophageal duplication cyst adherent to serosa of esophagus and ileal duplication cyst	10
14	17 year-old	Male	Retrosternal pain during eating + fever	Double intramural channels separated by mucosal layer + two communications were present between duplication and lumen	Close contact between duplication and esophagus	7
15-16	2 cases	NA	Respiratory and digestive signs	Tubular esophageal duplication	NA	30
17	Neonate	Male	Respiratory distress	Esophageal duplication	NA	31
18	Newborn	Male	Peripheral cyanosis and pulmonary crepitations	Short tubular blind pouch projecting from the left posterolateral aspect of the upper esophagus	NA	9
19	One day	Male	Irregular respiration and blood stained secretions from mouth after feeds	Short tubular blind pouch projecting from the left posterolateral aspect of the upper gullet	Communicating esophageal duplication	9
20	10 years	Male	Sudden dysphagia, fever and pharyngeal pain	Double esophageal lumen	No surgical intervention	18

Note. NA: Not Available.

The treatment of choice is surgical excision via thoracotomy or video-assisted thoracoscopy which has the advantages of reduced postoperative pain, short recovery period, early hospital discharge and minimal skin scarring.[22] Surgical excision should be performed as early as possible even if the patient is asymptomatic to avoid occurrence of complications such as infection,[13] or neoplastic transformation.[15]

Conflicts of Interest Disclosure

The authors have no competing interests to declare.

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