CASE REPORTS

Tubular duplication of the esophagus

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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 present 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 gas-

troenteritis. He gave history of intermittent dysphagia with both, liquid and solid diets for more than five years. There was no history of cough or choking during swallowing, and the rest of his medical history was noncontributory. Physical examination and routine blood tests were unremarkable.

Gastrografin esophagography 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 1983.⁶ Few other cases were
reported afterword and summarized in Table 1. Esophageal duplications represent 10%-20% of foregut duplications.\cite{5} Tubular duplication of esophagus is rare and is much less common than cystic duplication of the foregut.\cite{6} Unlike cystic duplications, tubular duplications usually communicate with the normal esophagus.\cite{7-9} Tubular esophageal duplication can be associated with other anomalies such ileal duplication cyst and bronchogenic cyst.\cite{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,\cite{12} however, the duplication in this patient involved the proximal esophagus. The lumen of esophageal duplication may show ectopic pancreatic tissue,\cite{13} gastric tissue,\cite{14} or sometimes malignant tissue such as adenocarcinoma carcinoid which was reported in 32-year-old man who presented with dysphagia.\cite{15} Esophageal duplication can be asymptomatic and discovered incidentally.\cite{16} However, the usual presentation of esophageal duplication is dysphagia and chest pain.\cite{7,17,18} Dyspnea has been reported in one case of tubular esophageal duplication and it was associated with bronchogenic cyst and pericardial defect.\cite{11} Anorexia and weight loss have been reported in patient who had developed a malignant transformation of esophageal duplication.\cite{19} Persistent wheezing can be a manifestation of tubular esophageal duplication.\cite{20} Hemoptyis also has been reported to be a feature of esophageal duplication containing ectopic gastric tissue in neonate.\cite{21}

![Figure 1](image1.png)

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

![Figure 2](image2.png)

**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| Choking 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  |     | NA                                          | Tubular duplication of the esophagus           | NA               | 29         |
| 13  | 18 weeks | 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 thoroscopy 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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