Isolated Complete Tubular Duplication Of The Oesophagus In A New Born -A case report

ABSTRACT

Duplications of the alimentary tract are rare and ileum is the most commonly affected site, followed by the oesophagus which accounts for 15% of all digestive tract duplications. The duplicated oesophagus with mucus, submucous and muscular membranes, is adjacent to the true oesophagus without a common wall. This condition leads to dysphagia, vomiting, nausea, retrosteranal pain or respiratory distress and stridor. This condition is commonly seen during the newborn period.

Tubular duplication is commonly seen in the mid ad lower third of the oesophagus where as cystic oesophageal duplication is found in the lower third of the oesophagus. Usuall 70% -90% of the patients are diagnosed before two years of age as they develop symptoms. However few case of oesophageal duplication has been discovered incidently in adult patients.

In this study, we report a rare case of isolated complete tubular oesophageal duplication in a 2 day old male newborn, who presented with excessive salivation, respiratory distress and intolerance to feeds.

Key words: Oesophagus , congenital abnormality, salivation, respiratory distress

INTRODUCTION

Duplication of the Oesophagus is a rare congenital abnormality accounting for 15 % of all digestive tract duplications. Ileum is the most commonly affected site followed by the esophagus.

Oesophageal duplication is of three types:

a. Cystic duplication is most common. It may or may not communicate with the Oesophageal lumen.

b. Tubular
c. Diverticular, is the rarestof the three

Tubular duplication contains mucosal lining derived from the primitive foregut diverticulum. It may or may not have muscular lining.

Mid and lower third of the oesophagus are most commonly affected sites of tubular duplication where as cystic duplication is usually seen in the lower third of the oesophagus. Patients with oesophageal duplication suffer from dysphagia, respiratory distress, recurrent pneumonia and rarely gastro intestinal bleeding. Commonly 70% - 90% of these patients become symptomatic in young age; and are diagnosed before two years of age. Few studies have discovered oesophageal duplication in adult patients incidentally. The authors of this article reported a rare case of tubular oesophageal duplication complicated by adenosquamus carcinoma at its lower end, in a 32 years old man who complained progressive dysphagia[1].

Khvorostov et al published a case of tubular oesophageal duplication in a 3 day old female new born without any associated anomalies. Newborn was having respiratory distress, salivation, dysphagia, tachycardia and tachypnoea.During laryngoscopy, two oesophageal lumen were detected. Two nasogartirc tubes were placed under fluoroscopic guidance. First into the blind ending oesophagus used for suction, and second into the normal oesophagus which was used for feeding. Contrast oesophagram showed a tubular oesophageal duplication extending from the cervical region to the diaphragm without any gastric communication. The duplicated segment was present on the left side in the neck and on the right posterior side of the normal oesophagisin the chest. Computed tomography showed a small duplicated lumen along the right side of the orthotopic oesophagus.

The duplicated oesophagus was excised by thoracoscopy leaving the normal oesophagis in place. Child returned to normal growth and development after follow up period of 20 months[2]

In this study, we report a rare case of isolated complete tubular oesophageal duplication in a 2 day old male newborn, who presented with excessive salivation, respiratory distress and intolerance to feeds.

CASE REPORT

A two days old male newborn was brought to radiology department by a paediatric surgeon for contrast swallow examination. New born was having respiratory distress,excessive salivation and intolerance to feeds. During laryngoscopy,two oesophageallumen were seen and two nasogastric tubes were placed by the paediatric surgeon under fluoroscopic guidance. One tube entered in the normal oesophagusandsecond tube reached blind ending pouch. The nasogastric tube into the normal oesophagus was used for feeding and blind ending duplicated oesophageal pouch tube was attached to suction pump. After obtaining informed consent from parents of new born male, a non ionic contrast was instilled in the oesophagus with the help of syringe and spot films of oesophagus were taken.

The contrast oesophagram showed a tubular oesophageal duplication extending from the cervical region to the diaphragm without any gastric communication. The duplicated segment was posterior and on the right side of the normal oesophagus, separated from it by a septum. After the examination, new born was shifted to the children ward for surgical repair.

In this study, we report a rare case of isolated complete tubular oesophageal duplication in a 2 day old male newborn, who presented with excessive salivation, respiratory distress and intolerance to feeds.Oesophageal duplication was diagnosed on the basis of clinical picture ,direct laryngoscopy and oesophagography .The newborn was referred for surgical repair.

DISCUSSION

Alenazi etal reported a case of tubular duplication of the oesophagus, in a 12 year old boy presenting with upper respiratory tract infection and acute gastroenteritis in a hospital emergency department. He had history of dysphagia with both liquid and solid food for more than five years. Oesophagography with gastrografin showed an elongated, well defined pouch measuring about 6 cms in the proximal oesphagus displacing it to the left. Contrast enhanced

computed tomography of chest showed additional tract of the oesophagus with a blind end that measured about six cm in its cranio caudal dimension and deviated the oesophagus to the left side. Endoscopy showed two oesophageal lumens present about 15 cm from the incisors. The patient was referred for surgical treatment.Author further advised to carry out the surgical excision of the duplicated segment as early as possible to avoid complication like infection or neoplastic transformation[3].

Barbinoetal gave evidence of a possible endoscopic treatment of tubular oesophageal duplication in an 11 year old boy who presented with chest pain, cough, dysphagia and fever.

"Oesophagram with water soluble contrast medium revealed a double oesophageal lumen extending about 5 cm. Endoscopy showed two oesophageal lumens, main lumen was larger with normal mucosa and accessory lumen was narrower and lined with ulcerated tissue. The gastroscope could pass into the accessory lumen till its distal communication with the main lumen. An endoscopic treatment was attempted by using a standard videogastroscope.Starting from the incision, a lengthwise and step by step mixed cutting and cautery of the Intraluminal bridge were carried out. No complications were noted. The child was discharged on 3rd dayafter the procedure and was asymptomatic at 18 months follow up" [4].

Karboubi et al reported two cases of oesophageal duplication diagnosed in the neonatal period. In both cases respiratory distress and vomiting were main symptoms. Diagnosis was confirmed by contrast study of the oesophagus. Total tubular form was seen in the 1st case where as a cystic form in the 2nd case. The authors concluded that oesophageal duplication is a rare abnormality of benign nature, which can be diagnosed in the neonatal period by a noisy compression picture. Diagnosis of this abnormality should lead to search for other digestive duplications as well as associated malformations especially vertebral[5].

VENTURA et al published a case of tubular duplication of the oesophagusin a 6 years old boy who presented to the emergency department with an impacted foreign body in the oesophagus. Boy has mild dysphagia for solid food since 2 years of age.After a coin in the

oesophagus was removed on endoscopy, an orifice in the oesphageal wall andoesophagealstenosis were seen. A barium oesophagram was done which showed tubular duplication of the oesophagus. Oesophageal duplication is a rare congenital anomaly having incidence of 1 in 8200 and represent 10 % of all forgut duplications. Oesophagealduplicataion cysts are seen in lower third (60% to 95%) and an right side and tubular form are present in the middle and lower oesophagus. 70% to 90% of the cases of duplication are diagnosed before 2 years of age but these have also been discovered incidentally in adult hood. Surgical treatment has been recommended by some authors due to the risk of malignancy especially for tubular forms; however a laproscopic approach for resection of cystic forms has also been described recently.[6]

FAMILIARIet al reported "successful endoscopic treatment in a symptomatic tubular oesophageal duplication in 24 years old man. A mentally retarded patient has dysphagia since childhood which had worsened recently and was associated with abdominal pain and regurgitation. EGD (esophagogastroduodenoscopy) showed presence of a tubular duplication of the distal tract of the oesophagus, 7 cm long, starting 35 cm from the upper incisors. A cap assisted septotomy was done endoscopically under general anesthesia with endo tracheal intubation. In less than 20 minutes, procedure was completed. Oral feeding was started on the second post operative day and patient was discharged 2 days later. Follow up after one year showed patient in good clinical condition" [7].

TOMAR et el published "a case of asymptomatic thoracic oesophageal duplication cyst, diagnosedalong with bronchiectasis incidentally, in a 18 years old female. She presented with high grade fever, cough and breathlessness for 1-2 months. She had productive cough with expectoration with non-foul smelling sputum. Radiograph of chest showed bronchiectatic changes in left lung lower zone and surprisingly showed a cystic lesion on left side of trachea, just below the carina. Contrast enhanced computed tomography of chest showed a tubular cystic lesion measuring 9.1x3.1x2.9 cm in the left para tracheal region with thin enhancing wall having minimal fluid, Trachea and oesophagus were deviated slightly to the right side by the cyst. Endoscopic ultrasound showed an elongatedtubular structure parallel to oesophagus with

common serosal lining likely to be an oesophageal duplication cyst. Oesophageal duplication cysts are indication of anomalies of the foregut along with bronchogenic cysts and consist of up to 1 out of 15 cases of mediastinal cysts. Oesophageal cysts are mostly asymptomatic but complications can develop and surgery may be needed" [8].

A case report of double oesophageal duplication cysts, with ectopic gastric mucosa has been published by ZHANG et al.Patient was a 3 year old Chinese boy who had intermittentfever and dry cough.Computed to mography of the thorax showed an oval shaped cyst like tumour of size 10x5.4x5.8 cm located in the extrapleural space. It was extending along the right paraverbral gutter and compressing the trachea forward.In addition to this, a small (1 cm) sized, oval shaped cyst was noted in the posterior mediatinum, between the oesophagusand the spinal colum at T_1 level. Larger cyst was resected in an enbloc manner and smaller cyst was left untreated.Oesophageal cyst was neither communicating with the oesophageal lumen nor the trachea, computed tomography done at 6month follow up showed no recurrence and small sized cyst remained unchanged in size[9].

Garge and Samujh reported a case of isolated complete tubular oesophageal duplication in a neonate, 2 day old male who presented with excessive salivation and intolerance to feeds. Nasogastric tube could not be passed beyond 17 cm. Plain radiograph showed tube had stopped at the level of 5^{th} thoracicvertebra. Normally nasogastric tube could be inserted into the stomach.Oesophagogram with contrast showed tubular oesophageal duplication extending from cervical region to diaphragm. There was no associated gastric duplication. The duplicated segment was lying posterior and on the right side of the normal oesophagus. An infant feeding tube was inserted in the pouch under flouroscopic control and used for suction. The nasogastric tube was placed in the stomach and used for feeding. During laryngoscopy two oesophageal lumen were detected. Patient condition was deteriorated and he expired due to pneumonitis and sepsis.Tubular oesophageal duplication is a rare congenital anomaly seen in about 10% of all foregut duplications. Differential diagnosis of tubular duplication are pharyngeal perforation and long upper pouch of a trachealoesophageal fistula. The autheros further opined, that the

rare diagnosis of tubular duplication should be considered in patients presenting with typical history of excessive salivation and atypical chest radiographs[10].

Ozcan et al described endoscopic septum division of tubular oesophageal duplication in two children and did a systemic review of 14 studies in the literature having 16 pediatric cases of tubular oesophageal duplication. Surgery was performed in 10 cases and two cases were treated endoscopically. In follow up, 9 patients who had surgical treatment and two endoscopic, were healthy. Author of this article presented two cases of tubular oesophogeal duplication in whom endoscopic septum division was doen using an electrosurgical knife. Both showed satisfactory radiological and clinical response to treatment. Authors further opined that endoscopic septum division was minimally invasive procedure with satisfactory therapeutic resposnse[11].

Kim et al published a case of communicating tubular esophageal duplication combined with bronchoesophageal fistula. Oesophageal duplication is rarely diagnosted in adults as they are usually asytomatic. It is further extremely rare that oesophageal duplication is connected to the oesophagus through a tubular communication and combined with bronchoesophageal fistula and has never been reported in the English literature. It is very difficult to diagnose this condition even with combinations of several modalities like computed tomography, magnetic resonance imaging, endoscopic ultrasonogrphy,oesophagography etc. Authors reported a 49 years old male patient having communicating tubular Esophageal duplicataion combined with bronchoesophagealfistula (BEF) that was diagnosed incidentally on the basis of endoscopy and oesophagography finding during the postoperational evaluation of bronchoesophageal fistula (BEF)[12].

Huang et al reported a case of communicating oesophageal tubular duplication in new born infant without any other associated abnomalties. Diagnosis of duplication of oesophagus was made by contrast study of the oesophagus with water soluble contrast medium and computed tomographic scan. Surgical excision of the duplicated oesophagus was done and at 1 year follow up examination, the patient was healthy[13].

Takemura et al published a case of thoracic oesophagealdulplication cyst in adult, which had ectopic pancreatic tissue in the solid portion. Cyst was resected under the thoracosopic approach in adult, instead of traditional thoracotomy approach. Authors recommended thoracoscopic approach for mediastinal diseases as it was minimally invasive for patients[14].

Bielen et al published a case of double barreled oesophagus caused by intramural dissection. The authors opined that this condition should be differentiated from oesophageal duplication, intramural abscess and diverticulum. The patient presented with a history of dysphagia for solid food, odynophagia and minor heametemesis for 2-3 years. Complaints had worsened recently with progressive dysphagia even for liquids. Computed to mography of chest showed a pneumomediastinum and a double barrelledoesophagus due to air in wall and oesophageal lumen. Water soluble contrast study after one week showed longitudinally oriented linear filling defect in the oesophagus extending from the level of aortic ARCH nearly to the level of the gastro-oesophageal junction giving double barrel appearance. Conservative management was thought to be sufficient in most of cases[15].

CONCLUSIONS

oesophageal duplication is a rare congenital anomaly, seen in 15% of all foregut duplications. Neonates can present with respiratory distress, excxessive salivation and intolerance to feeds and older children usually present with dysphagia. The incidence of congentialoesophageal duplication is approximately 1:8200 with male sex predominance.

Tubular type of oesophageal duplication seen in approximately 5-10% of all foregut duplications and commonly reported in middle and lower third of the oesophagus. The lumenof oesophageal duplication can show gastric tissue, ectopic pancreatic tissue and some times malignant tussue. Tubular oesophageal duplication may be associated with ilealduplicataion cyst and bronchogenic cyst.

Tubular duplications without communication with the normal esophagus are more commonly seen than cystic duplications. Althouth diagnosis of esophagus duplication can be made by contrast study of the esophagus, CT scan can prove useful by showing a second tubular structure adjacent to the true esophagus.

Consent

As per international standard or university standard,parental(s) written consent has been collected and preserved by the author(s).

Ethical Approval:

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

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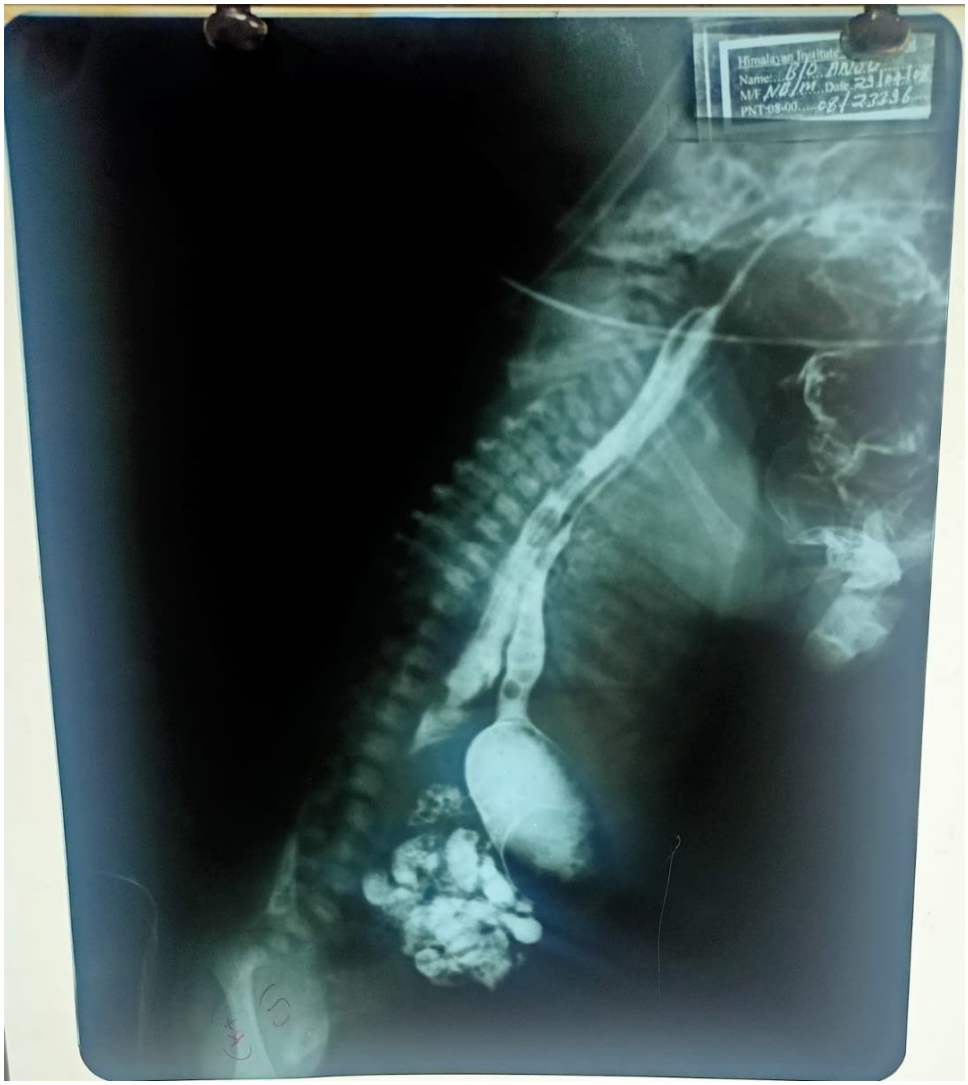
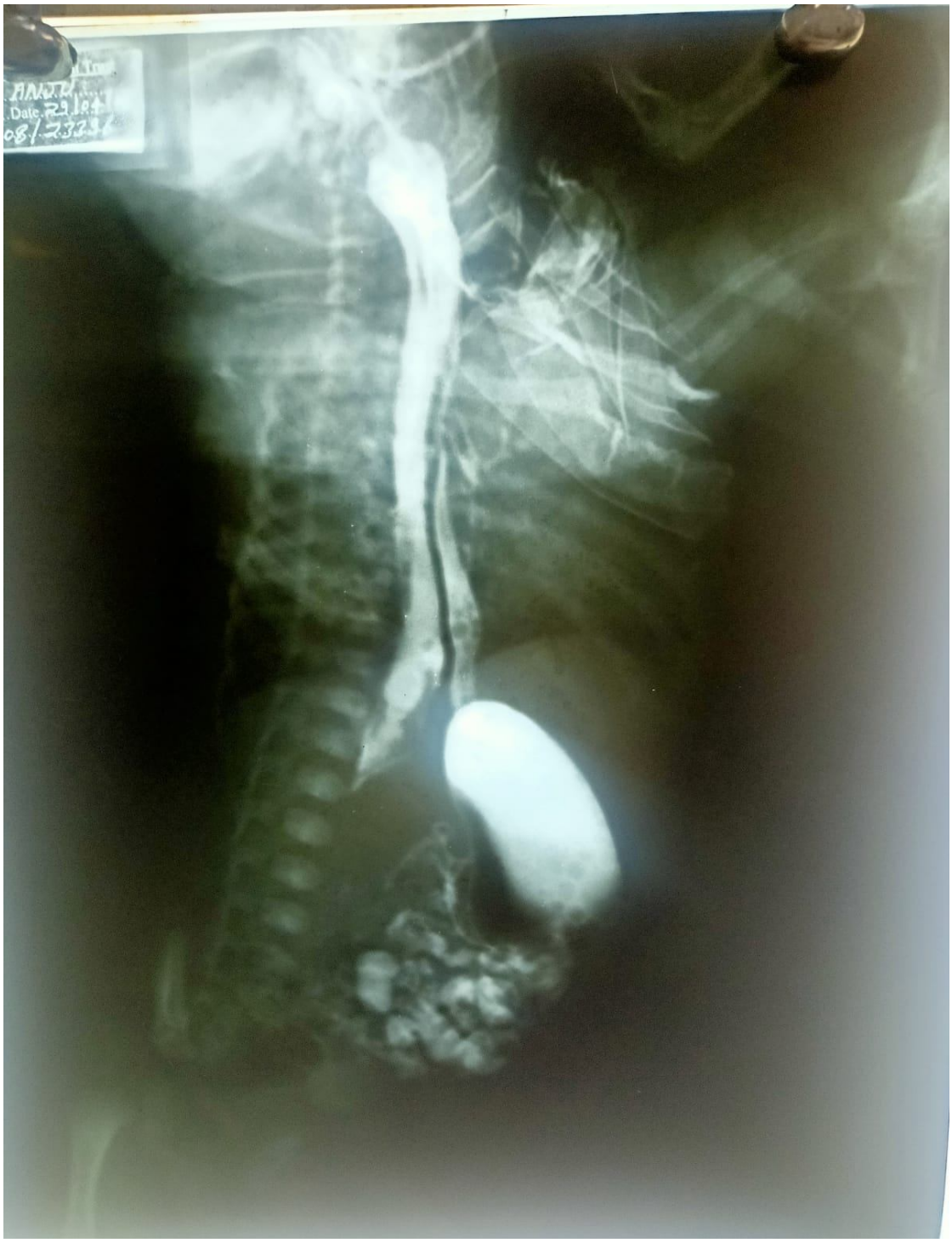
Fig.1 and Fig.2

Contrast oesophagram showing tubular oesophageal duplication extending from the cervical region to the diaphragm without any gastric communication. The duplicate segment is posteritor to the normal oesophagus, separated from it by a septum.



(a)



(b)

Fig 1 a,b : oesophagram reports