ORIGINAL ARTICLE

Acquired Tracheoesophageal Fistula in Infancy and Childhood

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Abstract

Acquired tracheoesophageal fistula in infancy and childhood is a very rare condition with no epidemiological data available in the literature for it. There is a huge similarity of the condition to the congenital acquired tracheoesophageal fistula. Management of children with this condition can be difficult with a high risk of complication. To provide a successful management of the condition, there are some points that need to be addressed about acquired tracheoesophageal fistula. In this report, the author presents fourteen different cases of acquired TEF with different etiologies over 11 years (2006 - January 2017) who were referred for management to tertiary care hospital in Jeddah, KSA; all patients had removal of the foreign body followed by surgical repair of the fistula. This report describes the detailed steps of our surgical repair, including the approach to the fistula and the type of repair pointing out the importance of removal of the scar tissue and use of interposition vascular grafts with suture lines separation. This study presents our experience in management and surgical repair of the acquired tracheoesophageal fistula.

Keywords

Tracheoesophageal fistula; Acquired; Esophageal foreign body; Disc battery

Introduction

Acquired tracheoesophageal fistula (TEF) in infancy and childhood is a very rare condition which is not well documented in the literature and mainly described in the adult age group[1,2]. There is a huge similarity between the condition and the congenital TEF type. Etiologies for acquired TEF include ingestion with impaction of foreign body material in the esophagus (pressure necrosis), prolonged use of tracheostomy with high pressure inflated cuff and blunt traumatic injuries in the upper chest[3-5]. We present here 14 different cases as acquired TEF with different etiologies that have been managed successfully. The etiology, diagnosis, and management approach for acquired TEF are discussed.

Patients and Methods

Fourteen patients who had acquired TEF, complicated with recurrent chest infection and aspiration were seen in the King Faisal Specialist Hospital and Research Center, Jeddah over 11 years (May 2006 –January 2017) and were included in this study. The youngest was 9 months old and the eldest was 45 months old. Ten patients presented with recent history of foreign body ingestion (disk battery) and acute respiratory symptoms. The remaining four were presented with

recurrent aspiration and chronic cough, one of them was battery ingestion which was easily diagnosed by simple chest radiography but the other three cases were plastic foreign body which was radiolucent on X-ray and late diagnosis were made by an upper gastrointestinal (GI) contrast study that showed TEF and confirm by endoscopy (Fig. 1). They were treated with the appropriate antibiotics and diagnosis was confirm with contrast swallow or endoscopy. All babies were operated through right cervical incision except two cases requiring right lateral thoracotomy due to the location and size of the TEF.

After a full pre-operative assessment and an early anesthetic consultation, patient was taken to the operative room and an upper endoscopy with a bronchoscopy was performed. The fistula was confirmed and the foreign body was removed (Fig. 2). During intubation, the anesthetist tried to insert the endotracheal tube (ET) below the fistula to avoid respiratory problems and confirmed the site of the ET by flexible endoscopy. The surgical team repaired the acquired TEF at that time with a right transfer cervical incision. The trachea and the esophagus were identified and separated by a vessel loop. Then, with an upper traction of both the trachea and the esophagus the fistula was identified. Careful dissection around the dens and inflammatory tissues was performed until the fistula was opened and the two structures were completely separated. Dissection was continued until clean healthy vascularized edges were obtained. Trachea site was closed with Proline 4.0 suture in a horizontal fashion to prevent stricture formation. The esophagus was mobilized freely, rotated and fixed to the para-vertebral fascia. Finally, well vascularized strap-muscles were mobilized and used as interposition flap between trachea and esophagus.

For the two patients who required right thoracotomy because of the low level of the fistula (within 2 cm from the Carina) a multidisciplinary approach between the surgical team and the anaesthetists was established due to the lower position of the fistula that could lead to anaesthetic and respiratory complications. The patient was then shifted to the operating room (OR) after full pre-operative assessment, and the anaesthetist inserted the endotracheal tube below the level of the fistula (at the level of the crania) under flexible bronchoscopy guides to avoid wrong positioning of the tube through the fistula. Right thoracotomy was performed and the trachea and the oesophagus were completely

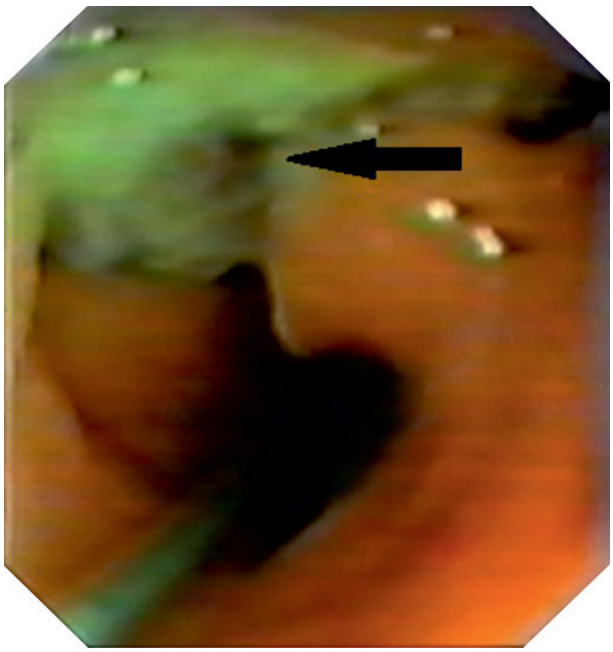

Figure 1A. Endoscopic view demonstrate of fibrotic tissue around the fistula (black arrow).

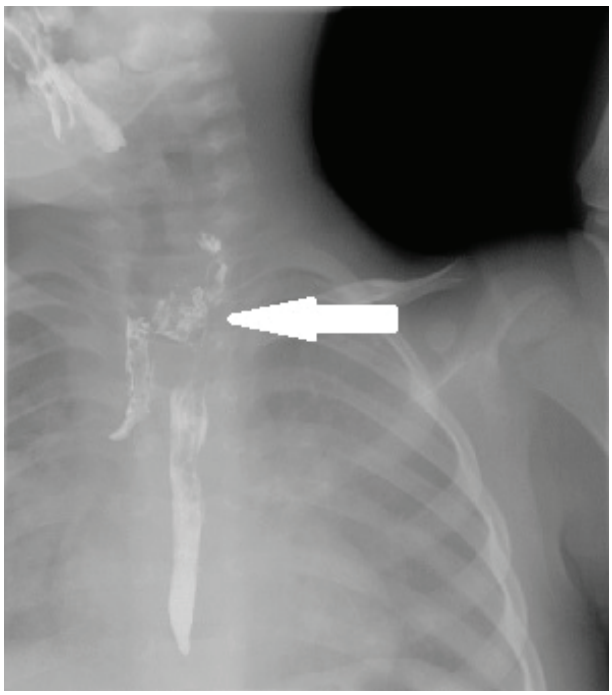

Figure 1B. An upper GI contrast study, the contrast reach the trachea through the irregular shape acquired TEF (white arrow).

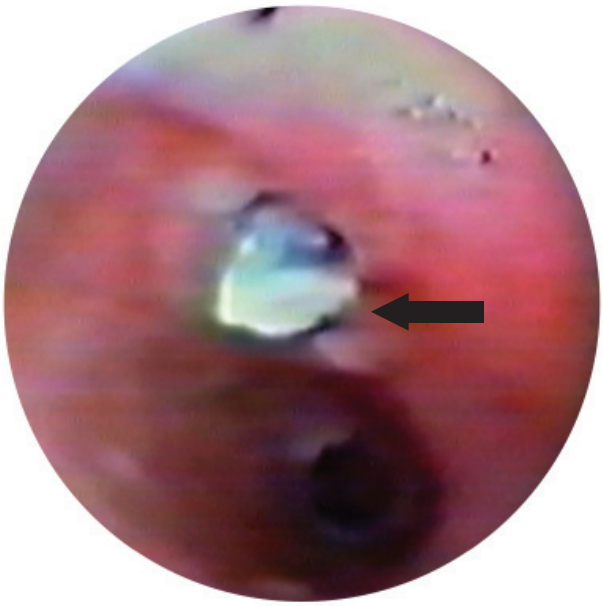

Figure 2A. Bronchoscopy view demonstrate plastic piece protruding from the fistula (black arrow).

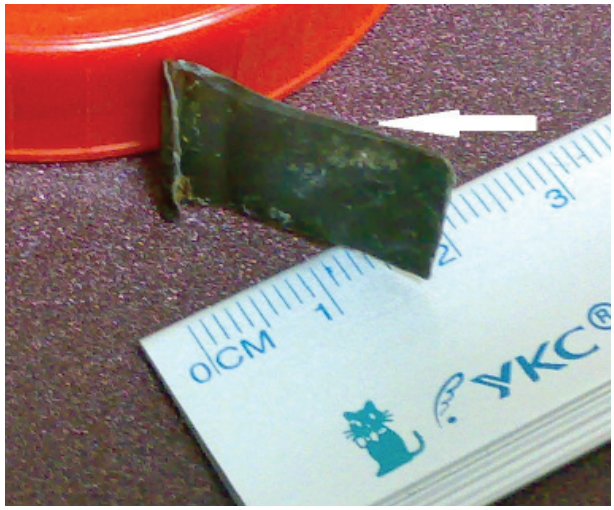

Figure 2B. A plastic piece removed from the fistula by endoscopy (white arrow).

dissected and isolated with a vessel loop. The huge fistula (2×1 cm) was removed and clean healthy edges were obtained, bovine pericardium patch was used to patch the tracheal defect and the oesophageal was closed using a 4.0 vicryl suture in a horizontal fashion and rotated, then a vascularized intercostal muscles flap was used as an interposition flap.

Post-operative an upper GI contrast study was done after eight days. If minor leaking was seen it was followed with conservative management using nasogastric (NG) tube feeding and antibiotics for a few weeks. When the upper GI contrast study showed no leak, the child was shifted gradually into oral feeding and then discharged from the hospital. All patients were seen in the outpatient clinic eight weeks after discharge and upper GI contrast study was performed again.

Results

All 14 patients had repair of the acquired TEF. Nine boys and five girls with average age of 23.4 months. In eleven cases, the cause was related to disk battery ingestion and in three cases were due to plastic pieces which cause delay and difficulty of the diagnosis.

Thirteen patients had primary repair of the trachea and one case required biological patch for large defect (1 by 2 cm) to prevent tracheal stenosis. All the esophageal sites were closed primarily in horizontal fashion and rotated and fixed to para-vertebral fascia.

Two patients had post anastomotic leak (Fig. 3) which were managed conservatively with NG feeding for 4 to 6 weeks until the leak disappeared completely.

One patient required a bovine pericardium patch for the tracheal defect, had no dysphagia or hoarseness in the follow up, with upper endoscopy and bronchoscopy showing no stricture formation and the patch was completely epithelised.

All 14 patients were seen in the outpatient department with median follow-up time of 5.6 years (range 1.1 to 9.3) with overall satisfactory results with no dysphagia or hoarseness.

Discussion

Congenital TEF is a well-known clinical condition to the pediatric surgeon with an incidence of approximately 1 in 3500 live birth[1], while acquired TEF in childhood and infancy is a very rare condition with no epidemiological numbers in the literature, it is mainly described in the adult age group[2]. The etiology of the acquired TEF includes ingestion with impaction of foreign

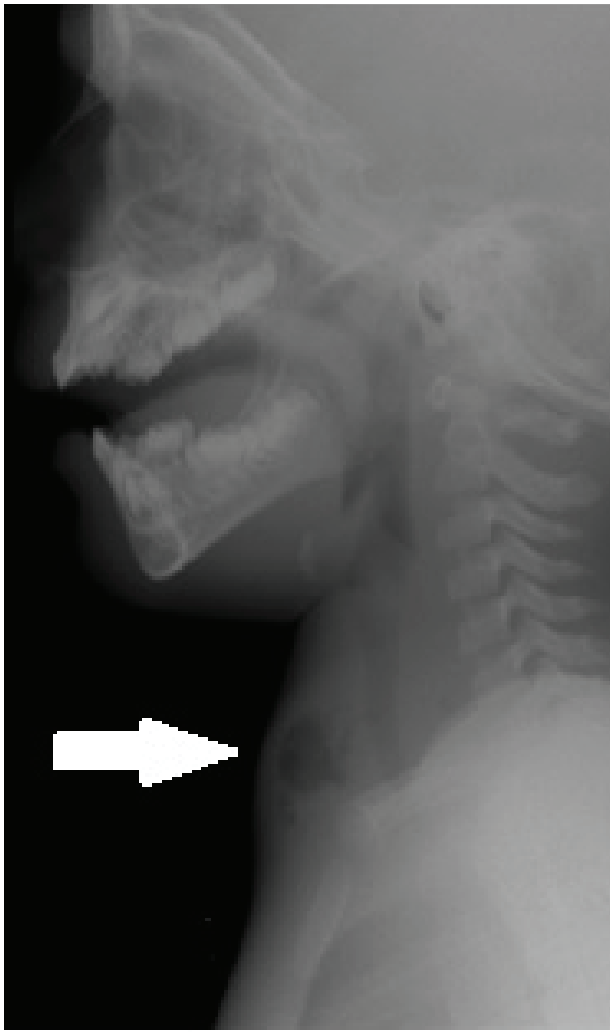

Figure 3A. Post –OP (10 days) Neck X-ray show large subcutaneous air collection indicting fistula and abscess formation (white arrow).

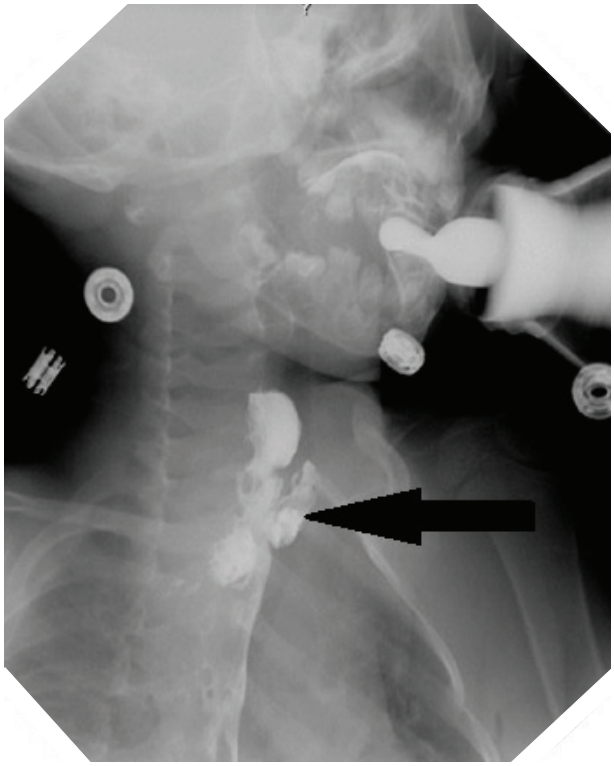

Figure 3B. Post –OP (10 days) Contrast study show leak in lower neck (black arrow).

body material in the esophagus (pressure necrosis), prolonged use of tracheostomy with high pressure inflated cuff and blunt traumatic injuries in the upper chest[3-5].

The foreign body materials which get impacted in the esophagus and lead to acquired TEF are disc (bottom) battery, small plastic objects, crown cork and pistachio shell[5]. Disc battery is the most common cause among them all, it could be due to combined physical pressure and electrochemical effect of the battery on the mucosa that leads to greater risk of penetration that can be as early as six hours after ingestion[6]. Up to our knowledge there are ten cases reported in the English language literature about disc battery. While plastic objects impaction take a longer duration with sub-acute symptoms to achieve penetration and acquired TEF, up to our knowledge there is three cases reported in the English language literature[2-7]. The annual report of the American Association of Poison Control Centres in 2011 mentioned that there were 6925 cases of toy ingestion among children 12 years and below with only one major complication in the outcome and zero deaths[8].

Diagnosis can easily be made if the child presents with acute symptoms like dysphagia and choking attack, but usually in these cases it can be confusing if the picture is sub-acute with a variety of symptoms including digestive and respiratory with unwitnessed foreign body ingestion[9]. Usually in this scenario, false initial diagnosis of croup, respiratory infection or asthma would be made by primary health care centre, especially if the foreign body is radiolucent, and would be treated with an antibiotics and supportive management that will make the patient improves for a while with a delay in the actual diagnosis[5], so careful history taking and high index of suspicion is crucial for diagnosis.

First standard step for diagnosis of TEF is an upper GI study with a contrast that will demonstrate the communication, followed by an endoscopic evaluation both bronchoscopy and esophagoscopy for full evaluation about the fistula size, level and degree of tissue damage. Previous reports recommended the use of multislice CT scan with 3D images or virtual bronchoscopy, which will have accurate results, as the standard method for evaluation of the acquired TEF. Therefore, it will give an accurate preoperative evaluation, precise surgery planning, avoid the risk of child exposure to contrast media, and reduce the risk of pulmonary aspiration and the technical difficulties with child in the standard method[6].

Management and surgical approach depends on the degree of tissue damage, level and size of the fistula. Because there are inflammatory changes and mucosal damage with or without chest infection, surgical delay is recommended until the inflammation subsides. Meanwhile, during this waiting period optimization of the patient condition and gastric decompression by gastrostomy is useful to prevent further damage to oesophageal mucosa, reflux with aspiration and providing feeding entry, total parenteral feeding is also used in some cases[10]. Spontaneous closure of the fistula with supportive management only within 4-11 weeks is rare but reported in the literature, usually if the diameter of the fistula is 15 mm or more spontaneous closure is unlikely[11]. The simple recommended producer is one stage repair surgery with simple division of the fistula and closure with interposition flap (strap muscle, intercostal muscle, omentum) between trachea and oesophagus[2,12]. Surgical approach depends mainly on the level of the fistula, mostly through right lateral cervical approach just at the anterior border of sternomastoid muscle. Thoracotomy is performed when the level is low around the carina or lungs are involved and may need resection[13].

Acquired TEF is more difficult to treat compared to congenital TEF with high recurrence rate and leakage post-operatively, due to the inflammatory process, fibrous tissue formation and mucosal damage[2,6]. From our limited experience, surgical delay for optimization of the patient condition, allowing the inflammatory process to subside, good removal of all the fibrotic tissue intra-operatively, cleaning the edges, and separation with well vascularized muscle flap will increase the success rate over all. Recently, conservative management had been drawing attention in the literature. It includes oesophageal rest with providing feeding via nasogastric tube or gastrostomy tube plus reflux medication and antibiotics if needed. A period of at least 6-10 weeks have been recommended for conservative management[14]. However, there have been no controlled studies which compare differences between surgical and conservative management outcomes for this condition.

Lastly, there is some anaesthetic difficulties reported in the literature in some cases, good early communication and planning between the surgeon and the anaesthesia team (mainly airway management) with a multidisciplinary approach will lead to excellent outcomes[7].

Conclusion

Our experience in management of acquired TEF showed a satisfactory result in restoring the oesophageal continuity and regaining normal oral intake. Early diagnosis and team approach management may reduce the intra-operative complication. Separation of the suture line and bringing intra-positional vascular graft is a key factor in minimizing the post-operative complication.

Conflict of Interest

The author has no conflict of interest.

Disclosure

The author did not receive any type of commercial support either in forms of compensation or financial for this study. The author has no financial interest in any of the products or devices, or drugs mentioned in this article.

Ethical Approval

Obtained.

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الناسور الرغامي المريئي المكتسب عند الرضع والأطفال

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المستخلص. الناسور الرغامي المريئي المكتسب عند الرضع والأطفال حالة نادرة جدًا ولا توجد بيانات وبائية كافية عنه في المنشورات العلمية. هناك تشابه كبير بين هذه الحالات وحالات الناسور الرغامي المريئي الخلقي. علاج الأطفال المصابين قد يكون صعب مع احتمال عالي للمضاعفات. على مدى ١١ عام تم اختيار ١٤ طفل محول للعلاج بمسببات مختلفة. الجسم الغريب استخرج من كل المرضي واجريت لهم عملية الاصلاح بعد ذلك. وقد اوضحنا تفاصيل العمل الجراحي ونوعية الإصلاح مركزًا على أهمية قص الأنسجة الميتة والملتهبة وتغطية اماكن الاصلاح بأنسجة حية. وفي هذه الدراسة نقدم خبرتنا للعلاج والاصلاح الجراحي للناسور الرغامي المريئي المكتسب عند الأطفال.