

An interesting and rare case of traumatic oesophagocutaneous fistula

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Abstract

Oesophagocutaneous fistula is a very rare disorder. No case of oesophagocutaneous fistula after blunt trauma has been reported. In this report we present a case of 25-year-old nursing student seen at the Lahore General Hospital, Lahore, with a history of blunt trauma to the neck. She was initially diagnosed with supra-sternal abscess. A few days after the incision and drainage of this abscess, she developed discharge of water and food particles from the wound site. It was investigated and diagnosed as oesophagocutaneous fistula. Her neck exploration was done and fistulous tract was found communicating with the upper mid-esophagus. The tract was excised and sent for biopsy. Histopathology revealed non-caseating granulomas with no evidence of malignancy. A gene X-pert was done to rule out tuberculosis and it came out to be negative. Post-operatively, the patient is living a normal life. This is the first of its kind case of oesophagocutaneous fistula reported from our part of the world.

Keywords: Blunt trauma, Esophagocutaneous Fistula.
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Introduction

Oesophagocutaneous fistula is an extremely rare condition with only a few cases reported in the literature. The cases that have been published are mainly the result of complications of anterior cervical spine surgery, tuberculosis, thyroidectomy, iatrogenic injuries during upper GI endoscopies, instrumentation or dilatation, carcinoma oesophagus and after penetrating neck trauma.^{1,2} Morbidity and mortality associated with oesophageal perforations is high and surgery (primary repair) is recommended in most cases.³ However, as the reported cases of oesophagocutaneous fistula are rare, standard management options are not available. There is no consensus regarding treatment of this condition in the best possible way for achieving maximum cure rates. Various strategies have been employed such as

conservative management through daily wound wash, nutritional management, use of intravenous antibiotics and fibrin sealants, etc.^{4,5} Surgery remains the mainstay of treatment for cases in which there is failure of conservative management.

We present a rare case of oesophagocutaneous fistula due to blunt trauma to the neck. This is the first reported case of formation of oesophagocutaneous fistula after blunt neck trauma in a young girl.

Case Report

A 25-year-old female, unmarried, third year nursing student was admitted on August 11, 2017 through outpatient department of the Lahore General Hospital, Lahore, with the complaints of chronic discharging sinus above supra sternal notch for more than three months. She had history of trauma to her neck eight months back when a box fell on her neck from a height of approximately eight feet. According to the patient, a few days later she developed suprasternal swelling which was diagnosed to be an abscess. It was incised and drained (I-&-D) in a local hospital. Her abscess wound did not heal post-operatively and there was persistent discharge for around three weeks. Later, the amount of discharge increased which was linked to her dietary intake. Whatever, she took orally came out through her wound on the neck. An ultrasound of the neck revealed "a small sinus tract which was communicating with the abscess in the subcutaneous tissue". A fistulogram was ordered which showed a "fistulous tract extending medially, posteriorly and inferiorly communicating with the upper dorsal oesophagus and opening outside onto the skin on the left side of lower neck." All these investigations were done before she was admitted with the diagnoses of oesophagocutaneous fistula. We ordered an upper GI endoscopy which showed a fistulous opening in the upper oesophagus. Biopsy revealed "severe acute or chronic inflammation and granulation tissue formation." CECT of the chest didn't show any communication with the trachea. After evaluation and discussion in the departmental multidisciplinary meeting, it was decided to explore the patient and excise her

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fistula. Hence, the patient underwent neck exploration. The tract was excised and the oesophageal opening was closed primarily with prolene sutures. Her post-operative course was unremarkable and she was discharged on fourth post-operative day. Her tract biopsy report revealed 'Fibrocollagenous and fibrofatty tissue with non-caseating granuloma formation.' Her gene X-pert was done in the OPD - which came out to be negative for mycobacterium tuberculosis. She was followed in the outpatient department after every two weeks and had a total of three visits. No further follow-ups were done.

Discussion

An oesophageal perforation is a life threatening condition and if not managed earlier it is associated with a mortality rate of up to 20-50%. Late presenting oesophageal perforations are preferably managed through surgical approach i.e. oesophagectomies. Oesophagocutaneous fistula is a complication of late presenting oesophageal perforations. It is an extremely rare condition and has been rarely discussed in literature. Hence, its standard management options are equally difficult to find. However, different strategies are being employed by surgeons all over the world to treat this rare condition. Some still prefer the conservative approach while others are using the surgical option of excising the fistulous tract and primarily repairing the oesophagus. Results have been promising with both these approaches. Surgical management depends on the feasibility of approaching the track, its location and the expertise of the primary surgeon.⁶

The diagnosis of oesophagocutaneous fistula can be made through chest radiograph, Contrast oesophagogram / Barium swallow, oesophagoscopy and CECT of the chest. CT scan helps to see the exact location of fistulous tract and delineates it. However, in our case barium swallow was not performed as upper gastroendoscopy was done which augmented the findings of the previously done fistula-gram outside our hospital. As trachea-oesophageal fistula is more common than oesophagocutaneous fistula and tuberculosis is endemic in our region, we had her CT of the chest done to see whether there was any communication with the trachea or not. However, no communication was found.

Cervical oesophageal perforations are usually managed conservatively. Among the conservative or non-operative approaches one is to keep the patient nil per oral, start

parenteral nutrition, intravenous antibiotics and do daily debridement of the wound. According to a case report published by Biplab Mishra et al, an 11-year-old child who presented with a late oesophageal perforation and had developed oesophagocutaneous fistula was successfully managed through this aggressive non-operative approach.⁷ However, in this case the patient had trauma to thoracic part of the oesophagus and being young had better tissue healing ability. Another way to conservatively manage oesophagocutaneous fistula is the use of Bio-absorbable material (Bio A) buffered with tissue sealants, i.e. platelet rich fibrin glue. There are case reports in which the patients were successfully managed with platelet rich fibrin glues alone. Marjin Joudi et al reported a case of a three-year-old child in which traumatic oesophagocutaneous fistula was initially managed conservatively but didn't resolve and was later successfully managed using platelet rich fibrin glues.⁵

As limited data on oesophagocutaneous fistula surgeries are available, the literature providing information on surgical management of late presenting oesophageal perforation can be extrapolated to their management. Generally those fistulas which cannot be managed conservatively, are large or have already failed the conservative approaches; these are managed through available surgical options.

There are several operative approaches -- from simple closure to oesophagectomies to augmentation with muscle flaps. Surgical approach depends upon the location, size and viability of oesophageal mucosa. Simple primary closure of oesophagus depends upon whether the tissue after refreshing margins is suitable enough for closure or not, and whether the size of the wound can allow approximation without any tension in the tissues. Some surgeons prefer augmentation after primary closure with an omental flap, pectoralis muscle flap, radial forearm flap or the most commonly used, sternocleidomastoid flap.

In general, small oesophageal perforations are treated by primary closure and large ones have to undergo oesophageal resections. In our case, fistulous tract was excised and the size of the oesophageal perforation allowed primary closure successfully. However, we didn't do any augmentation with muscle flap. The patient was followed one week after discharge and later every two weeks for three months. Her recovery was remarkable with complete cure of the disease. We didn't go for

conservative approaches in our case due to the patient's concerns for early recovery as she wanted to resume her studies as soon as possible.

Conclusion

Oesophagealcutaneous fistula secondary to blunt neck trauma is an extremely rare case with hardly any case previously reported in literature. Our case report highlights that cervical oesophagocutaneous fistula can be successfully managed surgically to achieve early and best possible cure.

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Consent From: A written informed consent has been taken from the patient for the publication of this case and any accompanying images. Approval was provided by the dean of surgery.

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