



Case report

Native oesophageal mucocoele: A rare complication of double exclusion of oesophagus

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ABSTRACT

Introduction and importance: Native oesophageal mucocoele usually follows bipolar exclusion of oesophagus for various reasons and is very rare in literature. Though mostly asymptomatic, its symptoms can be divided into 3 groups – Compressive, Infective and fistulizing symptoms. The management options described in the literature are percutaneous drainage, chemical ablation, esophagectomy and internal drainage using Roux-en-Y reconstruction.

Case report: A 40 year old female, presented with complaints of dysphagia, weight loss and chest pain for 6 month. She had history of retrosternal gastric pull-up for oesophageal stricture following corrosive injury. On evaluation with CT chest, there was a well-defined fluid attenuated tubular elongated lesion in the mediastinum in the region of oesophagus which was non-opacified with oral contrast and a diagnosis of giant oesophageal mucocoele was made. She underwent internal drainage of mucocoele by roux-en-Y esophagojejunostomy with placement of transanastomotic drain and discharged with an uneventful recovery with the trans-anastomotic drain in situ, which was removed on outpatient basis. Now she is asymptomatic in the subsequent follow up.

Clinical discussion and conclusion: Though rare, mucocoele of oesophagus can lead to life threatening complication like respiratory distress, sepsis. Its diagnosis requires high index of suspicion and CT chest is helpful. Management options depend upon nutritional status of the patient and associated co-morbidities. Esophagectomy is the definitive form of treatment but not always possible and other options can be internal or percutaneous drainage.

1. Introduction

A mucocoele of the native oesophagus is rare with few cases reported in the literature [1]. It usually follows oesophageal exclusion or bypass procedures for cancers or benign strictures [2]. Although it is usually asymptomatic, it can have three types of presentation which are compressive, infective and fistulizing. When sufficiently large, it can cause compressive symptoms like chest or abdominal pain, vomiting and difficulty in breathing which includes life threatening respiratory distress due to compression of the tracheobronchial tree, septic complications and fistulization [3,4]. Various treatment options are available, like percutaneous aspiration with chemical ablation, resection of the mucocoele or rarely, internal drainage using a Roux en Y reconstruction.

We report the case of a female who had a symptomatic oesophageal mucocoele which developed as a late complication after surgery for a

corrosive oesophageal stricture. She was managed surgically with internal drainage using a Roux-en-Y reconstruction.

2. Case report

A 40-year-old woman who had no co-morbidities, accidentally ingested toilet cleaner in 2016 and was managed conservatively at that time without any ICU admission. Gradually she started having difficulty in swallowing which was initially to solids and gradually progressed to liquids. She was diagnosed to have a stricture involving the lower 8 cm of the oesophagus about 30 cm from the incisor teeth. There was a hold up of contrast in the mid oesophagus on gastrografin swallow. She was initially managed with stricture dilatation (3 times) and stenting (once) but due to recurrent symptoms she underwent an exploratory laparotomy with a retrosternal gastric pull up in 2018. She was initially planned for esophagectomy but abandoned due to dense adhesions in the

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mediastinum and the native oesophagus was left in situ after closing both the ends (Fig. 1). She remained asymptomatic for 6 months after surgery following which she again developed dysphagia and chest pain which was diagnosed to be a passable anastomotic stricture and managed by endoscopic dilatation. She then remained asymptomatic for a further year following which she started to develop fever along with weakness and presented to us in an emaciated condition with a body mass index of 14.58 kg/m². She was evaluated with gastrograffin follow through which showed a normal anastomotic site subsequently confirmed by repeat endoscopy. CT scan of the chest showed a well-defined fluid filled tubular elongated lesion in the mediastinum in the region of oesophagus which was not opacified with oral contrast. A diagnosis of a giant oesophageal mucocoele was made (Fig. 2). She was initially planned for percutaneous drainage due to her general condition under CT guidance but a safe window couldn't be found. Excision of mucocoele was also considered to be too hazardous because of the general condition of the patient and the presence of dense adhesions seen on CT.

We therefore performed, via a left thoracoabdominal incision, an internal drainage using a Roux-en-Y jejunal loop and placed a tube through the anastomosis into the mucocoele. Intra-operatively, we found a distended oesophagus measuring 6 cm in diameter which on aspiration revealed pus (Fig. 3) (on analysis – Gram stain showed polymorphonuclear cells but the culture was sterile). The Roux-en-Y esophagojejunostomy was done using a 40 cm jejunal loop and the anastomosis performed in a single layer using a mersilk interrupted sutures. She was discharged on post-operative day (POD) 9 with the trans-anastomotic drain in situ. A CT done after injecting contrast via the trans-anastomotic drain showed a collapsed oesophagus with no anastomotic leak (Fig. 4). The trans-anastomotic drain was removed on POD

25. She is now asymptomatic with a follow up CT showing a collapsed oesophagus (Fig. 4).

3. Discussion

Oesophageal exclusion or bypass procedures have excellent long term outcomes in patients with benign diseases like strictures, perforations, congenital tracheo-oesophageal fistulae. They are also an effective form of palliation in patients with advanced oesophageal carcinoma [5–7]. The native oesophagus can be left in situ in most disease except in patients with severe oesophagitis and malignancy [5,8,9]. It is always desirable to excise the diseased oesophagus if possible but this may be technically unsafe in patients with previous perforation, corrosive injury causing mediastinitis, dense adhesions or in cases of tracheo-oesophageal fistulae [2,4,9–11]. Oesophageal exclusion usually has a satisfactory outcome in patients with corrosive injuries, but the debate still remains whether to excise or bypass the native oesophagus. The complications associated with bipolar oesophageal exclusion can be either related to a mucocoele or, very rarely, malignant transformation, the actual incidence of which is not known and is limited to individual case reports [12].

A mucocoele of native remnant bypassed oesophagus is a rare condition with few individual case reports and a single case series reported in the literature (Table 1).

Viable oesophageal mucosa is needed for the formation of a mucocoele, so it is uncommon after corrosive injury which causes its destruction. After oesophageal exclusion, a mucocoele develops within weeks as suggested by Deaton et al. [13]. Usually the size of the mucocoele is limited by the pressure-induced atrophy of the secretory glands and rarely does it become large enough to cause symptoms. The

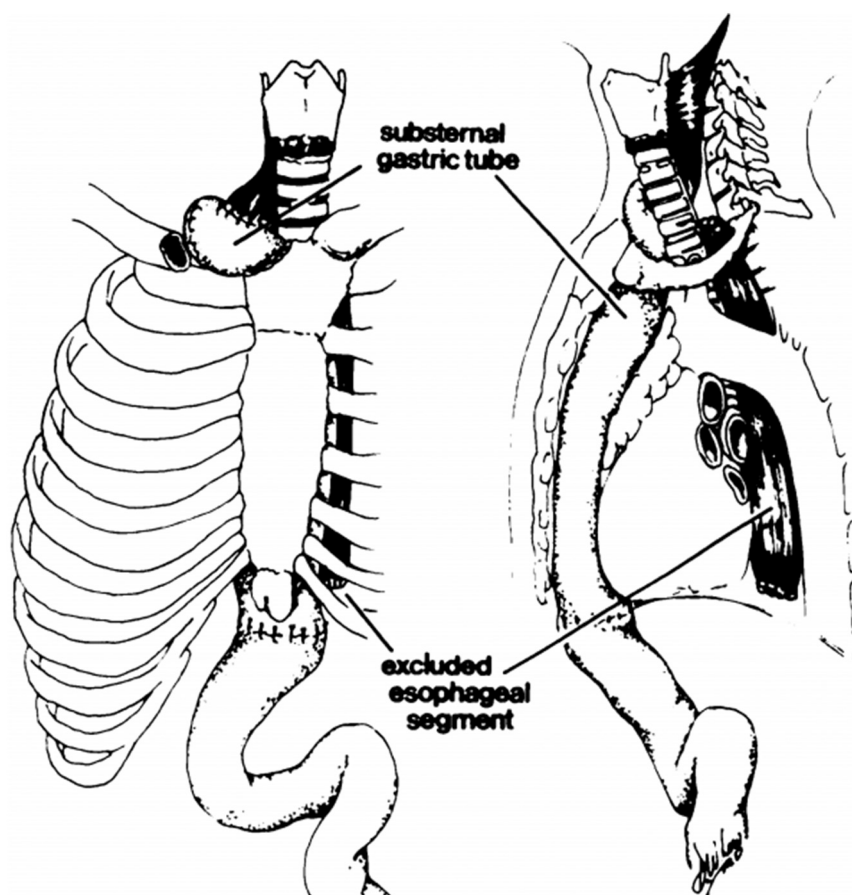


Fig. 1. Diagram of surgical procedure (frontal and lateral views) showing isolated oesophagus posteriorly with substernal gastric pull-through anteriorly. Taken from Glickstein et al. [27].

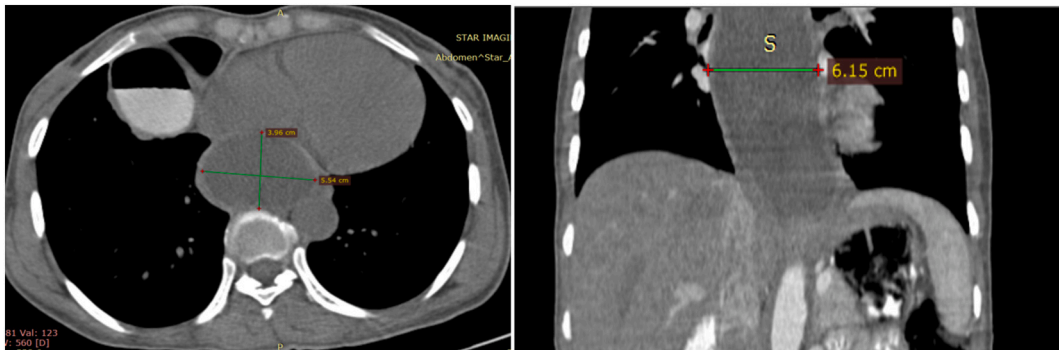


Fig. 2. Sagittal and Coronal section of CT chest showing giant oesophageal mucocoele (Green Ruler)

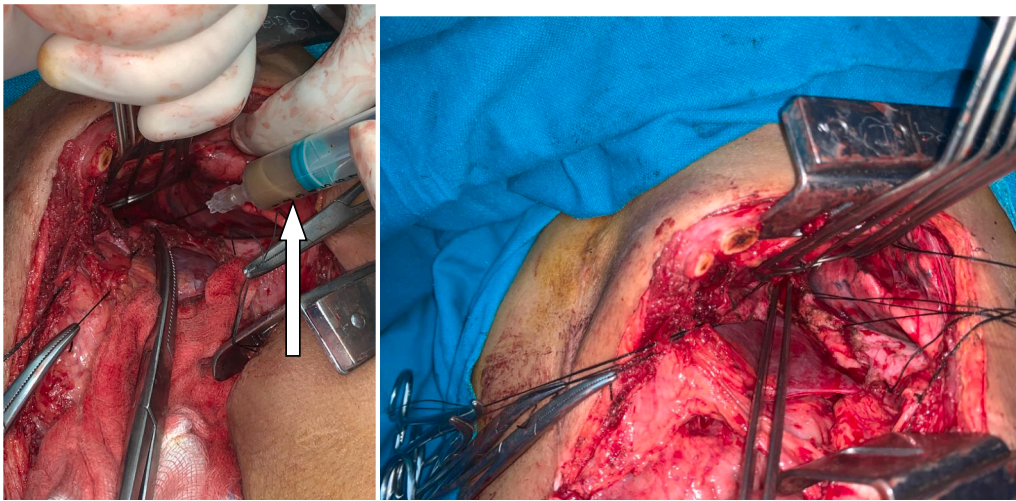


Fig. 3. Intra-operative image showing pus on aspiration (Block White Arrow) and mucocoele opening for anastomosis (Metal Forcep).

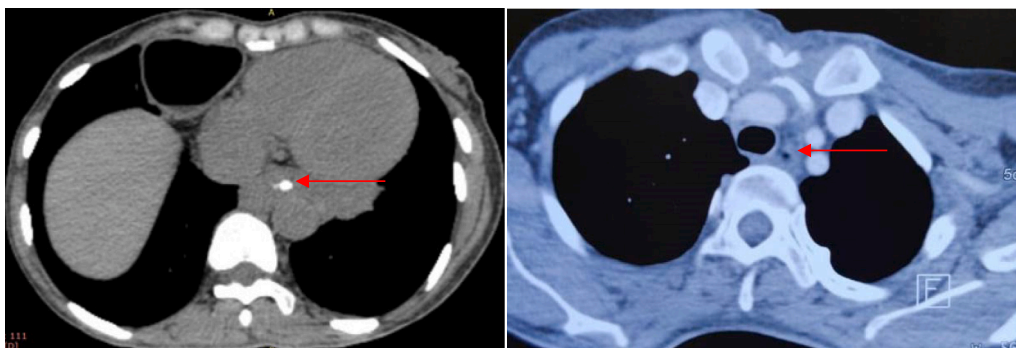


Fig. 4. CT done through trans-anastomotic stent (Red arrow) showing collapsed oesophageal mucocoele and follow up CT showing collapsed oesophagus (Red Arrow)

threshold diameter for a mucocoele to produce compressive symptoms is 5 cm according to the literature though sizes up to 14 cm have also been reported.

The presentation varies from incidental detection to abdominal or chest pain, vomiting and weight loss. Rarely some small mucocoeles become infected and cause symptoms like fever with an abscess in the abdomen, neck or thorax depending upon their situation. Rarely, they may cause tracheo-oesophageal fistula which may be due to pressure induced ischaemic necrosis or infection. They can also lead to secondary pulmonary infection or acute aspiration and death [14]. In long standing cases, there is a risk of malignant transformation of the mucocoele,

which may be due to chronic long standing infection or any of the inciting factors that were present prior to exclusion [12].

The usually asymptomatic nature of the disease makes it difficult to diagnose. A high index of suspicion is required as physical examination is not much of help. Chest X ray can be used as an initial tool which shows a widening of the mediastinum but it is not reliable as the conduit in patients with an oesophageal bypass may appear similar. Contrast studies are helpful to rule out any fistula or leak, as is endoscopy. CT of the thorax is the most accurate method to diagnose this condition. The mucocoele appears as a fluid filled structure in the posterior mediastinum with a high tomographic density of around (25–35) HU. CT also

Table 1
Native oesophageal mucocoele - a review of world literature.

Author	Year	Index surgery	Cause of index surgery	Mode of presentation	Interval	Treatment
Olsen et al. [24]	1984	Gastric pull up	Radiotherapy induced stricture for carcinoma of larynx	Fever, night sweat and weight loss	2 years	Internal drainage by roux-en-Y esophagojejunostomy
Kamath et al. [25]	1986	1. Gastric pull up - 5 patients 2. Colonic conduit	1. Achlasia cardia - 2, Tracheoesophagal fistula - 1, Oesophageal perforation - 1, Oesophageal atresia - 1. 2. Achlasia cardia	1. Respiratory distress, pain, nausea, sepsis 2. Pain nausea	1. 1 and 6 week, 1.5, 2.5, 3 year 2. 7 year	1. Esophagectomy 2. Esophagectomy
Genc et al. [12]	2001	Colonic conduit	Tracheo-oesophageal fistula	Tracheo-oesophageal fistula	23 years	Esophagectomy
Fong et al. [3]	2006	Colonic conduit	Oesophageal perforation	Cutaneous fistula in the neck	7 months	Esophagectomy
Lal et al. [26]	2014	Gastric pull up	Corrosive oesophageal stricture with oesophageal perforation during endoscopic dilatation	Respiratory distress due to tracheal compression	1 years	Esophagectomy
Hadad et al. [11]	2008	1. Gastric pull up 2. Gastric pull up	1. Caustic injury 2. Boerhaave Syndrome	1. Chest pain and cough 2. Respiratory distress	1. 8 years 2. 3 years	1. Esophagectomy 2. Esophagectomy
Rathinam et al. [1]	2016	Colonic conduit	Corrosive injury	Fever, respiratory distress	2 years	Esophagectomy
Sapkota et al. [23]	2019	Gastric pull up	Oesophageal perforation	Fever, dysphagia, neck swelling	1 ½ years	Thoracoscopic esophagectomy
Neethirajan et al. [16]	2019	Gastric pull up	Boerhaave syndrome	Chest pain, dysphagia	3 months	Esophagectomy
Manoharan et al. [14]	2019	Gastric pull up	Oesophageal perforation	Tracheo-oesophageal fistula	10 years	Conservative

helps in accurate delineation of anatomy and the relation of mucocoele with surrounding structures. MRI can also be used for diagnosis [11].

Due to the risk of rupture causing sepsis and tracheoesophageal fistulae, all symptomatic mucocoeles should be treated. Though excision of the native oesophagus is the most definitive form of management, it is not possible in all cases. Excision of the native oesophagus also depends upon various factors and the main one being the reason behind the initial oesophageal exclusion. In patients with a history of corrosive injury and perforation there is severe mediastinal fibrosis contraindicating a safe resection. The interval between the duration of the initial insult and second surgery also plays a vital role. The longer the duration after the first operation, the less the fibrosis and the easier is the resection. Haddad et al. reported two patients and found that resection was easier in the first who had a 10 year gap between two procedures while resection was very difficult in the other who had a gap of 3 years [11]. Leaving behind a part of the oesophageal wall in case of dense adhesions followed by argon plasma coagulation of the mucosa is another option but it is associated with recurrence as seen in a case report by Collins et al. [15]. Another factor is the general condition of the patient and the ability to withstand a long thoracotomy. There are reports of CT guided drainage of a mucocoele but this was associated with recurrence as described by Collins et al. [15]. Instillation of alcohol into the cavity has also been used with some success [15,16]. A traditional method is internal drainage of the mucocoele by a Roux-en-Y jejunal reconstruction which was initially described by Kirschner in 1920. This had gradually grown out of favor in view of its associated complications like anastomotic leakage [17,18]. It gained popularity gradually as the morbidity and mortality of the procedure improved [19,20,21]. Another modification is to leave behind a portion of the cardia attached with native oesophagus during the primary surgery to facilitate the anastomosis [22]. Thoracoscopic mucocoele excision can also be tried safely and is increasingly replacing the traditional thoracotomies, though it is technically demanding. The natural history of the disease and previous surgery poses a challenge to thoracoscopy due to dense adhesion, which can be managed with partial excision [23].

As with any other condition, prevention is better than cure. But, information regarding this is lacking in the literature. One of the methods is to avoid inclusion of columnar e.g. Barrett's epithelium in the excluded oesophagus but this is not always possible [24]. Our protocol in this regard is to place a tube or Foley catheter into the oesophageal

lumen from below during the index surgery for a long period of time which helps in obliterating the lumen by drying of the secretions and possibly by inducing fibrosis in the form of foreign body reaction. In our experience over 10 years, 30 patients presented with corrosive ingestion out of which 15 underwent oesophageal exclusion procedures. In 5 patients, the stomach was used as a conduit while the colon was used in 10. All of them are in regular follow up and only this concerned patient developed a mucocoele of the native oesophagus. None of our surviving patients have developed malignancy of the native oesophagus with the longest follow up being 9 years and 4 months.

As our patient presented with emaciation and a poor general condition, a complete resection of the mucocoele by a thoracotomy was not feasible and we were also unable to find a safe window for CT guided drainage. We therefore decided to drain the mucocoele internally and had a gratifying result.

4. Conclusion

An oesophageal mucocoele is a rare entity which usually develops after bipolar oesophageal exclusion. To prevent potential complications, all symptomatic mucocoeles should be treated. Various options are available from CT guided percutaneous drainage with ethanol ablation to mucocoele resection. Though resection of the native oesophagus is most desirable, it is not always possible due to safety concerns. Internal drainage of the mucocoele is a safer alternative where resection is not possible.

The work has been reported in line with the SCARE 2020 criteria [28].

Ethical approval

Ethical committee approval was not needed as per the regulations of the local committee.

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CRedit authorship contribution statement

SSJ- Data collection and analysis, writing full article. **RCRO** – Data collection and analysis, Proofing of article. **AY**, **SAPD** – Study conceptualization and designing, Article proofing. **SN**– Final proofing of article and final approval.

Guarantor

Suwendu Sekhar Jena

Provenance and peer review

Not commissioned, externally peer-reviewed.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Data availability statement

No datasets were generated or analyzed during the production of case report.

Declaration of competing interest

None.

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