Mediterranean BioMedical Journals International Journal of Medicine and Surgery

2018, Volume 5, Regular Issue, ID 245

DOI: 10.15342/ijms.v5ir.245

CASE REPORT

ACQUIRED TRACHEO-ESOPHAGEAL FISTULA: A RARE CASE REPORT OF THE NEGLECTED DENTURE

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ABSTRACT

Introduction: Dentures are common accidental ingested foreign body (FB) especially among elderly. It is frequent to have foreign body impacted at esophagus in adults however it is very unsual to have Tracheo-esophageal fistula (TEF) caused by denture. The diagnosis of TEF is challenging due to two reasons. Firstly, most of the dental prosthesis is radiolucent and not visible in routine radiological investigation. Secondly, patient with history of swallowed dentures prosthesis may be asymptomatic initially and develops symptoms over time. In contrary, prolonged history of FB in esophagus with TEF has higher risk to develop serious complication such as pneumonia and lung abscess.

Case Presentation: We report a case of 62 year old gentleman with background history of hypertension and temporal lobe epilepsy presented with history of choking on taking solid and liquid associated with significant weight loss past 2 months. He had lost his denture for almost 1 year during sleep. Endoscopic examination of the larynx showed normal anatomy but pooling of saliva. CT thorax showed foreign body within a tracheoesophgeal fistula. OGDS showed denture within a well formed tracheoesophageal fistula. He had acquired TEF secondary to the dentures.

Conclusion: Symptomatic elderly who lose their denture during sleep should not be neglected. They need immediate medical assessment thus will reduce further debilitating complication. Failing to identify and treat this condition urgently, patient will suffer acquired trachea-oesophageal fistula on which the treatment is challenging and the morbidity and mortality is high.

KEY WORDS: Acquired Tracheo-Esophageal Fistula, Denture, TEF, Endoscopic Stenting.

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INTRODUCTION

Foreign body (FB) ingestion is common in otorhinolaryngology practice. It is more common in pediatric, elderly, drug addicts, psychiatric patients and prisoners. [1] It is estimated that 80% of the non-food or true FB ingestion (mostly coins, buttons, small toys and marbles) occur in pediatric population due to natural oral curiosity between the ages of 6 months to 3 years. [2] The FB most commonly swallowed by adults are fish bones, bones and dentures. [3] Eighty percent of the foreign

body passed the esophagus spontaneously. [4] Only 10-20% require endoscopic removal and about less than 1% require surgical intervention. [5,6] Impacted foreign body in esophagus must be removed without delay, especially if the object is sharp because it leads to severe complications. These complications include esophageal perforation, mediastinitis, tracheoesophageal fistula and erosion to major blood vessels that results in mediastinal hemorrhage.

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This is a case report of a tracheoesophageal fistula (TEF) secondary to an impacted denture. While majority of TEF are caused by malignancy, prolonged tracheal intubation is the commonest cause of non-malignancy acquired TEF. [7] Immediate surgical intervention is crucial upon detecting TEF for any delay can lead to severe pulmonary complication due to the aspiration through TEF. To the best of our knowledge that only few papers in the English literature on acquired TEF caused by a denture. It give rise to a great diagnostic and therapeutic challenges as being demonstrated in this case.

CASE REPORT

A 62 year Chinese gentleman with background history of hypertension and temporal lobe epilepsy presented with history of choking on taking solid and liquid associated with significant weight loss past 2 months. There was no painful swallowing. He had loss his dentures during sleep for almost 1 year prior to the onset of the presentation. He had no dysphonia, chest pain or fever.

Examination of his neck, oral cavity and larynx did not show any significant findings. Flexible laryngoscopy showed normal supraglottic and laryngeal inlet. Fibreoptic Endoscopic Evaluation of Swallowing (FEES) reveled pooling of clear fluid, nectar and porridge over the valleculae and pyriform fossa despite multiple attempts of swallowing. A flexible nasoendoscopy passed until esophagus which exhibit normal nasopharynx and oropharynx. Both Vocal folds are normal and mobile bilaterally. Oesophagogastroduodenoscopy (OGDS) found there is foreign body seen in oesophagus, 29.5cm from upper incisor. Chest x-ray shows no pneumonitis and no FB was seen.

Computed Tomography (CT) scan showed a fistulous communication between the esophagus and the trachea at T5/T6 level. A slice like hyperdense lesion measuring 3.0cm in length is lodged within this fistula, partly within the trachea and partly within the oesophagus. The impression was acquired trachea-oesophageal fistula caused by foreign body.

A multidisplinary discussion between the otorhinolaryngology, cardiothoracic, general surgery and radiology was held.

Flexible bronchoscopy demonstrated a foreign body embedded at 1cm above the carina at the posterior wall. The foreign body was covered with granulation tissue and slough. Esophagogastroduodenoscopy (OGDS) performed later showed denture penetrating the oesophagus into the trachea at 20cm from incisura with a well-developed fistula (fig. 1 & fig. 2). Foreign body 'denture' was retrieved (fig. 3 & fig. 4). Repeated OGDS showed no further trauma to oesophagus (fig. 5). Patient was admitted for observation and administration of broad spectrum antibiotic. No elevated total white cell count and other biomarkers was within normal range. On the second day of admission patient had another bronchoscopy for the deployment of Y-stent. It was uneventful and patient was discharge on day 3 hospitalization. Patient had oesophageal stenting at done

2 weeks after the removal of the denture. Procedure was done at a different centre due to lack of facility.



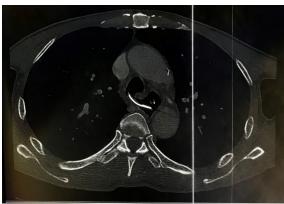
Figure 1: OGDS view of foreign body penetrating the esophagus



Figure 2: OGDS view of foreign body located within the fistula



Figure 3: Denture retrieved.



<u>Figure 4: Computer tomography scan upper thorax</u> showing foreign body within a tracheoesophageal fistula.



Figure 5: OGDS view of the well-formed tracheoesophageal fistula noted after retrieval of the foreign body.

An OGDS 2 months post stenting revealed stent migration to distal part of fistula and it was adjusted. A repeated OGDS 10 days later showed oesophageal stent migrated distally and not covering the fistula. The fistula still large and stent was adjusted upward to cover the fistula. There was mucosa ingrowth of the previous stent covering. Stomach and duodenum was normal. Another OGDS 2 month later found stenosis 15cm just proximal to stent and it was dilated with 9mm and 12mm dilator. Stent was still insitu.

On the other hand, patient had monthly surveillance flexible bronchoscope to review the placement of Y stent. While repeated bronchoscopy, we noted that there is secretion accumulated in between the trachea wall and Y-stent, a small granulation tissue at the distal end of both Y-stent division and present of the non-fully closed fistula. It was not progressive thus Y-stent was kept in situ

A series of Computed Tomography (CT) scan thorax was performed at 5th month post retrieval of dentures which showed a linear air pocket seen connecting the oesophagus and the trachea suggestive of residual patent fistula in between the oesophagus and the trachea. At 10th month post retrieval of dentures CT scan thorax

showed The tracheo-esophageal fistula is still present but the residual tract appears smaller in size. There was no pneumonitis reported.

Since the initial removal of the dentures patient given Ryle's tube feeding. Patient however was well throughout the 8 months follow-up and procedures. No further complication such as lung abscess, mediastinitis, pneumomediastinum or pneumonia detected. He was referred to another tertiary centre as the tracheaoesophageal fistula is not healed.

DISCUSSION

The diagnosis of foreign body is normally straight forward. Patient is generally aware of related symptoms. Given the history of swallowing any object that give abrupt symptoms such as odynophagia, dysphagia, coughing, neck discomfort and retrosternal fullness. [8] However, in our case patient had delayed presentation. History of missing his dentures earlier without any symptoms. Later on he develops choking on swallowing solid and liquid with dysphagia and significant weight lost in two months makes the diagnosis become difficult. The diagnosis of TEF is challenging due to two reasons. Firstly, most of the dental prosthesis is radiolucent and not visible in routine radiological investigation. Secondly, patient with history of swallowed dentures prosthesis may be asymptomatic initially and develops symptoms over time. [9]

A case reported by Mohajeri et al. [8] of a 57 year old man who presented with progressive dysphagia after he had swallowed dentures for 9 months. Samarasam et al. [10] reported a case of 51 year old man with history of swallowing his upper denture 10 years ago with any medical treatment, now presented with 2 months history coughing on taking solid or liquid with recurrent chest infection. N Razafmanjato et al. [11] reported a case of 46 years old gentlemen who presented with 3 years chronic dysphagia that toward the hospitalization it become more progressive.

A literature by Kwok SL et al. [12] showed that there is a significant morbidity and mortality following a prolonged history of foreign body in oesophagus. Specifically stated in his literature that after 24hours of impaction there is a significant risk of major complications. P. Rekha et al. [7] reported of non-resolving pneumonia and P.B Rajesh et al. [13] and Akira Mogi et al. [14] reported a lung abscess in a patient who developed serious complication following acquired TEF.

We observed a different scenario in our case by which patient only had TEF without other related complications. Probably due to the size and location of the fistula which are the huge factor to be consider for complication to arise.

Managing TEF is challenging. Our case was treated in a more conservative manner in view that the age is advanced, patient is physically fit without limited activity of daily living and isolated TEF without other serious complications. Patient was treated with temporary deployment of stent in the trachea as well as in the

oesophagus. We experienced adverse complication emerged from our intervention. To the best of our knowledge there is no reported literature on endoscopic stenting of the TEF in hope of the secondary intention of healing.

In contrary of our approach, few reported literatures described a good outcome following primary closure of the fistula in a cases who developed life threatening complications (non-resolving pneumonia and lung abscess). [12, 13, 14] PB Rajesh et al., R Rekha et al. and Viwar K et al. advocate primary closure of acquired TEF caused by denture reason being spontaneous closure is

AUTHORS' CONTRIBUTIONS

The participation of each author corresponds to the criteria of authorship and contributorship emphasized in the Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly work in Medical Journals of the International Committee of Medical Journal Editors. Indeed, all the authors have actively participated in the redaction, the revision of the manuscript and provided approval for this final revised version.

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rare and delay of repair is hazardous. [7, 13, 15] We will need more data to compare in these two different approaches.

CONCLUSION

Symptomatic elderly who lose their denture during sleep should not be neglected. They need immediate medical assessment thus will reduce further debilitating complication. Failing to identify and treat this condition urgently, patient will suffer acquired trachea-oesophageal fistula on which the treatment is challenging and the morbidity and mortality is high.

PATIENT CONSENT

Written informed consent was obtained from the patient for publication of this case report.

COMPETING INTERESTS

The authors declare no competing interests.

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