

An Unusual Presentation of Ludwig's Angina with Empyema Thoracis and External Carotid Artery Pseudoaneurysm

Abstract

Ludwig's angina is a rapidly spreading soft-tissue infection and commonly occurs following odontogenic infection. A 30-year-old male presented to the emergency department, 7 days after the extraction of molar teeth with a sudden onset of mandibular swelling. He was diagnosed with Ludwig's angina with empyema thoracis and external carotid artery (ECA) pseudoaneurysm. He was successfully managed with video-assisted thoracoscopic surgery-guided drainage and endovascular embolization of ECA pseudoaneurysm. We share our experience of challenges faced during the management of unusual presentation of complicated Ludwig's angina.

Keywords: Embolization, empyema thoracis, external carotid artery, Ludwig's angina, pseudoaneurysm

Introduction

Ludwig's angina is a rapidly spreading soft-tissue infection involving the connective tissue of the floor of the mouth and neck. It commonly occurs following odontogenic infection involving the second and third lower molars. The causative organisms are *streptococcus viridians*, *staphylococcus aureus*, *enterococcus*, *escherichia coli*, *bacteroides*, *actinomyces*, and *pseudomonas*. Airway management is the most important step in the treatment of Ludwig's angina.^[1] The mortality rate of patients with Ludwig's angina is reduced from 50% to 8% with good antibiotics.

Case Report

A 30-year-old male presented to the emergency department, 7 days after the extraction of two left-sided mandibular molar teeth, with a sudden onset of mandibular swelling. The patient had tachypnea, tachycardia, and fever. The clinical examination revealed a tense, tender swelling in the submandibular region [Figure 1]. The ultrasound revealed a large heterochoic area in the left submandibular region, extending along the left side of the neck with the presence of collection. Contrast-enhanced computed tomography with angiography

of the neck and chest revealed a lobular pseudoaneurysm arising approximately 1 cm distal to the origin of the external carotid artery (ECA) [Figure 2a, b and c]. The pseudoaneurysm was surrounded by peripherally enhancing soft tissue and collection [Figure 2a]. The collection in the left submandibular region was extending along the left side of the neck suggestive of Ludwig's angina. The loculated collection was also noted in the anterior mediastinum till the level of the diaphragm and right-sided empyema with the underlying collapse of the lung [Figure 2d].

Digital subtraction angiography revealed a large pseudoaneurysm arising from the proximal ECA [Figure 2c]. The patient was initially managed with endovascular embolization of the left ECA pseudoaneurysm. Postembolization angiogram from the left common carotid artery and the right ECA shows no antegrade or retrograde filling of the pseudoaneurysm [Figure 2f]. After angioembolization, the patient was operated for emergency video-assisted thoracoscopic surgery (VATS) drainage of the right empyema, followed by incision and drainage of the cervical abscess with tracheostomy. The patient was discharged in a stable condition after 1 month, with tracheostomy *in situ*.

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The patient was readmitted after 3 weeks with the complaints of an increased neck swelling and intermittent clot passage from the previous incision and drainage site for 1 week [Figure 3a]. He was taken for emergency neck

exploration, and the infected hematoma was evacuated surrounding the coiled ECA [Figure 3b]. The postoperative period was asymptomatic, and the tracheostomy tube was removed. The patient was discharged in a stable condition. In the follow-up period, the patient was stable, accepting oral diet, and has no complaints [Figure 3c].

Discussion

In 1836, Wilhelm Frederick von Ludwig first described Ludwig's angina as an infection of the submandibular space. This infection is rapidly progressive in nature and causes inflammation of the tongue, pharynx, and soft tissue of the neck. This untreated infection can lead to respiratory infection, airway obstruction, sepsis, empyema, and death. Odontogenic infection is the most common source of submandibular infection. The other causes include penetrating trauma, sialadenitis, foreign body, superficial skin infection, infection of congenital cyst in the neck, and underlying malignancy.^[2]

Our patient presented with the complications of mediastinitis with empyema and pseudoaneurysm of ECA



Figure 1: Clinical image showing a tense, tender swelling in the submandibular region and neck

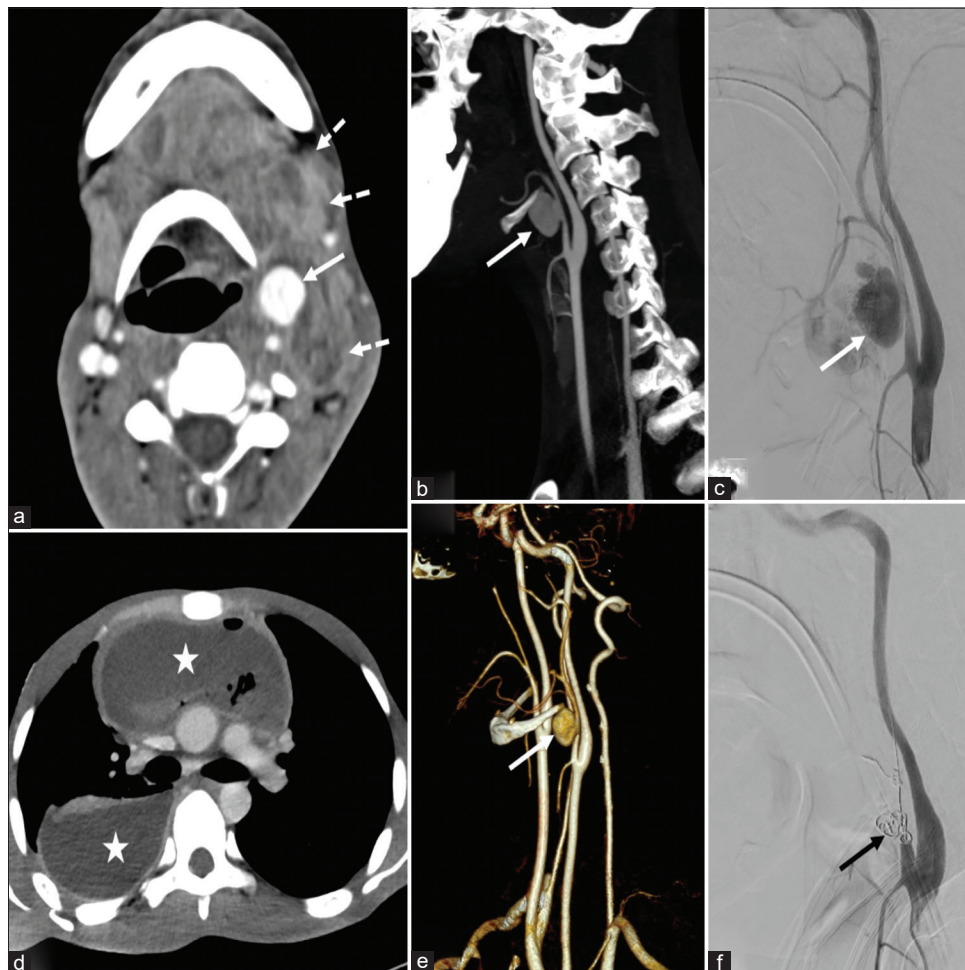


Figure 2: CT angiography (a-c and e) showing lobular pseudoaneurysm arising from the proximal part of ECA. (f) Postcoiling (black arrow) CCA angiogram showing no filling of the pseudoaneurysm (d) Axial CT image showing the loculated collection in the anterior mediastinum and right-sided pleural cavity. CT: Computed tomography, ECA: External carotid artery, CCA: Common carotid artery

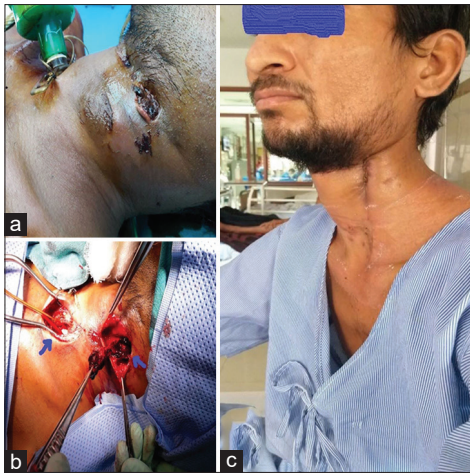


Figure 3: Image showing (a) Rupture pseudoaneurysm showing clots (b) Intraoperative image showing evacuation of clots from ECA pseudoaneurysm and control of common carotid artery to prevent bleeding (c) Follow-up image of the patient with a healed scar over the neck. ECA: External carotid artery

due to Ludwig's angina following dental extraction. The first step in the management of Ludwig's angina is airway management by tracheostomy.^[3] Empyema thoracis are a collection of pus within the pleural cavity. Over 3–6 weeks, empyema evolves through three phases which are exudative, fibrinopurulent, and organized phase. The initial step in the treatment of empyema is chest tube drainage.^[4] Early intervention by the thoracoscopic approach in empyema for drainage of pus has proven to be superior to delayed approach. Delay in surgical intervention has higher chance of conversion to open thoracotomy. The advantage of early intervention for the treatment of empyema is to stop the progression of the disease to the fibrotic stage by allowing drainage of pus and removal of necrotic tissue.^[5] The benefits of the thoracoscopic approach over thoracotomy include improved postoperative pain, blood loss is less, shorter hospital stay, lesser postoperative complications, and earlier return to routine work.^[6]

A pseudoaneurysm of the ECA is one of the complications of Ludwig's angina. A pseudoaneurysm is an outpouching of a blood vessel, which occurs due to a defect in the tunica intima and media. It communicates with the lumen of the vessel and is confined in the space by the surrounding tissue and fibrous reaction.^[7] Pseudoaneurysm can be managed by endovascular techniques or by open surgery. The available endovascular options are coil embolization, covered stent placement, and parent artery occlusion. The open technique is by resection of the pseudoaneurysm followed by end-to-end anastomosis.^[8]

This is the first report of Ludwig's angina associated with a pseudoaneurysm in the ECA, a complication that has never been observed before in any previous literature. However, only two cases were reported as isolated nontraumatic pseudoaneurysms occurring due to nontuberculous mycobacterium and mycotic infections.

Conclusion

Ludwig's angina is an infection of the submandibular space, most commonly following odontogenic infection. Ludwig's angina can be complicated by empyema thoracis and pseudoaneurysm of the ECA. Early and definitive multidisciplinary management of Ludwig's angina may prevent morbidity and mortality. Minimal invasive technique in the form of VATS-guided drainage and endovascular embolization is a better alternative to open surgical management.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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