Case Report

Ludwig's Angina with Evolution to Necrotizing Mediastinitis and Aorta Rupture Following to Death in a 23 Years-Old Woman

ABSTRACT

Ludwig's Angina is a rapidly progressing acute infectious process affecting the submandibular, sublingual, and submental spaces. If not promptly treated, it can be life-threatening and lead to sepsis and airway obstruction. Early detection, immediate and precise intervention including drainage, elimination of the causative factor, airway management, and antibiotic therapy are crucial for successful treatment. This manuscript presents a clinical case of Ludwig's Angina that evolved to mediastinitis and aorta rupture, resulting in a decease of a 23 years-old woman, despite treatment efforts.

Keywords: Ludwig's Angina, Mediastinitis, Aorta Rupture, Odontogenic Infection.

1. INTRODUCTION

Ludwig's Angina is a diffuse polymicrobial gangrenous cellulitis affecting the submandibular, sublingual, and submental spaces. It poses a potential threat to adjacent tissues [1, 2, 3]. This condition can arise from odontogenic or multifactorial causes, including foreign body presence in the oral cavity, oral mucosal laceration, mandible fractures, contaminated surgeries, penetrating traumas to the floor of the mouth, tonsillitis, and idiopathic factors [4].

The term "Ludwig's Angina" originates from the Latin word "angere", meaning to strangle, as patients often experience a sensation of suffocation accompanied by intense sialorrhea, dysphagia, odynophagia, hyperthermia, anorexia, dyspnea, tongue elevation with voice alteration, moderate leukocytosis, tachycardia, serosanguineous secretion, fetid odor, and trismus. Bilateral and diffuse swelling is also observed, along with systemic involvement. Various predisposing factors can contribute to the development of Ludwig's Angina, such as acquired immunodeficiency syndrome, chronic alcoholism, diabetes mellitus, glomerulonephritis, malnutrition, use of anti-inflammatory drugs, immunosuppressants, aplastic anemia, jaw osteomyelitis, salivary gland infection, infected oral neoplasms, peritonsillar abscesses, otitis media, tongue piercings, and the use of injectable drugs in cervical vessels [5, 6, 7].

If untreated, Ludwig's Angina can progress to involve cervical areas, the glottis, lateral pharyngeal space, and retropharyngeal space, ultimately leading to mediastinitis. The development of mediastinitis and acute airway obstruction are the most severe complications associated with increased mortality. While the overall mortality rate is reported to be up to 8% of cases, the presence of mediastinitis can escalate the mortality rate to as high as 50% [8, 9, 10].

This case report presents a rare instance of Ludwig's Angina progressing to mediastinitis due to extension of a dental infection.

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2. CASE PRESENTATION

The monitored patient, a 23-year-old woman, arrived to the emergency unit and was subsequently referred to the otorhinolaryngology service at Hospital São José do Avaí. She complained of hardened edema in the submandibular

region and trismus persisting for approximately 5 days. The patient reported a previous dental infection and had received outpatient treatment with oral administration of Amoxicillin, plus Clavulanate 1 g, every 12 hours for seven days, but her condition did not improve and later progressed to edema. During the physical examination, the patient exhibited hardened edema with fluctuation in the submandibular region, trismus, and oroscopy revealed the presence of caries and poorly maintained teeth. Due to suspicion of Ludwig's Angina, the initial approach on the first day of hospitalization included parenteral antibiotic therapy with intravenous administration of Ceftriaxone 1 g every 12 hours, as well as scheduled drainage of the submandibular abscess.

The drainage procedure was successfully performed the following day, with the placement of a penrose drain. On the third day, intravenous administration of Clindamycin 600 mg every 6 hours was added, leading to clinical improvement. However, on the subsequent day, the patient developed dyspnea, abdominal distension, and edema in the lower limbs.

A computerized tomography scan was performed (Fig. 1; Fig. 2), and laboratory examinations revealed a leukogram value of 34,400/mm^3, with metamyelocytes accounting for 3%, rods for 20%, and lymphocytes for 7%. Bacterioscopy of the pericardial fluid did not reveal any Gram-stained bacteria, and automated culture yielded a negative result.

On the seventh day of hospitalization, due to the worsening of the clinical condition and based on computed tomography images (Fig. 3), patient was transferred to the intensive care unit with a suspected diagnosis of necrotizing mediastinitis. Subsequently, she underwent video thoracoscopy, which identified a right pleural empyema. The medical team performed lung decortication, partial transpleural pericardiectomy, with the presence of purulent content, as well as exploration of the superior and posterior mediastinum, thorough cavity cleaning, and appropriate drainage. Additionally, the patient underwent cervicotomy with dissection of the upper mediastinum and proper drainage of purulent mediastinitis. A thoracostomy with closed pleural drainage was performed on the left side. Intravenous antibiotic therapy was initiated with Meronem 2 g every 8 hours and Vancomycin 2 g loading dose followed by 1500 mg every 12 hours.

For three consecutive days, the cardiovascular surgery team evaluated the patient as the pericardial drain exhibited a flow of 1000 ml, with a bloody appearance, within a three-hour period. However, this flow ceased spontaneously, leading to a decision for conservative management. On the subsequent day, the pericardial drain output resumed, with 1200 ml drained in less than 1 hour, and the fluid appeared bloody. In light of these circumstances, emergency surgical intervention was deemed necessary. A sternotomy was performed, providing access to the pericardial cavity, where a laceration in the posterior region of the ascending aorta and origin of the brachiocephalic trunk was identified. Unfortunately, the bleeding could not be controlled, and the patient succumbed to class IV hemorrhagic shock approximately three hours later.

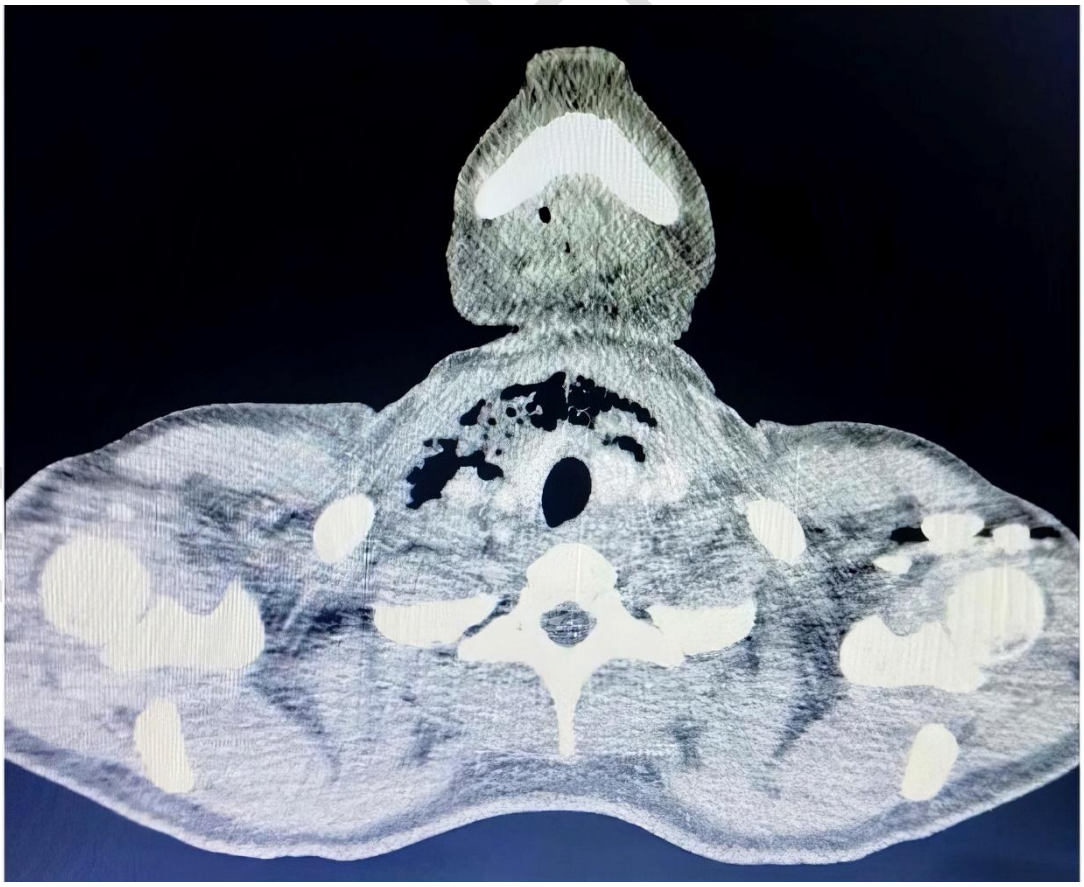

FIG. 1. COMPUTERIZED TOMOGRAPHY BEFORE THE PROCEDURE. AXIAL CUT; PRESENCE OF EMPHYSEMA FROM THE MANDIBULAR REGION TO THE UPPER 1/3 OF THE MEDIASTINUM.

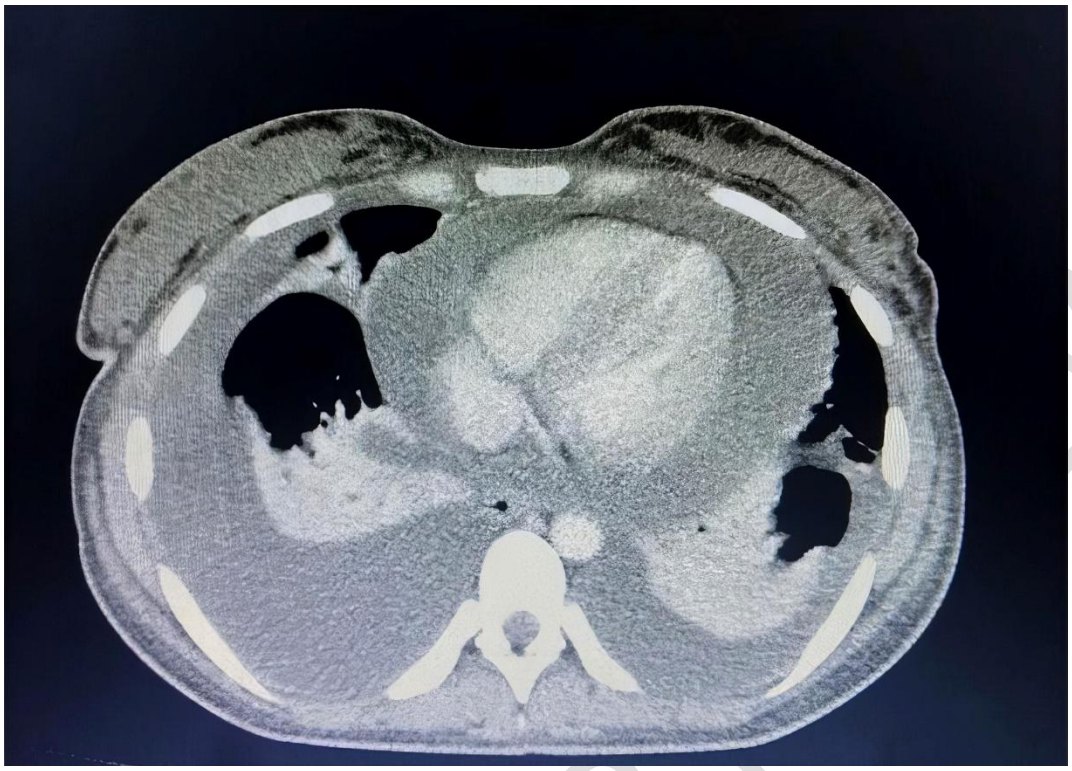

FIG. 2. COMPUTERIZED TOMOGRAPHY BEFORE THE PROCEDURE. AXIAL CUT; PRESENCE OF EMPHYSEMA FROM THE MANDIBULAR REGION TO THE UPPER 1/3 OF THE MEDIASTINUM.

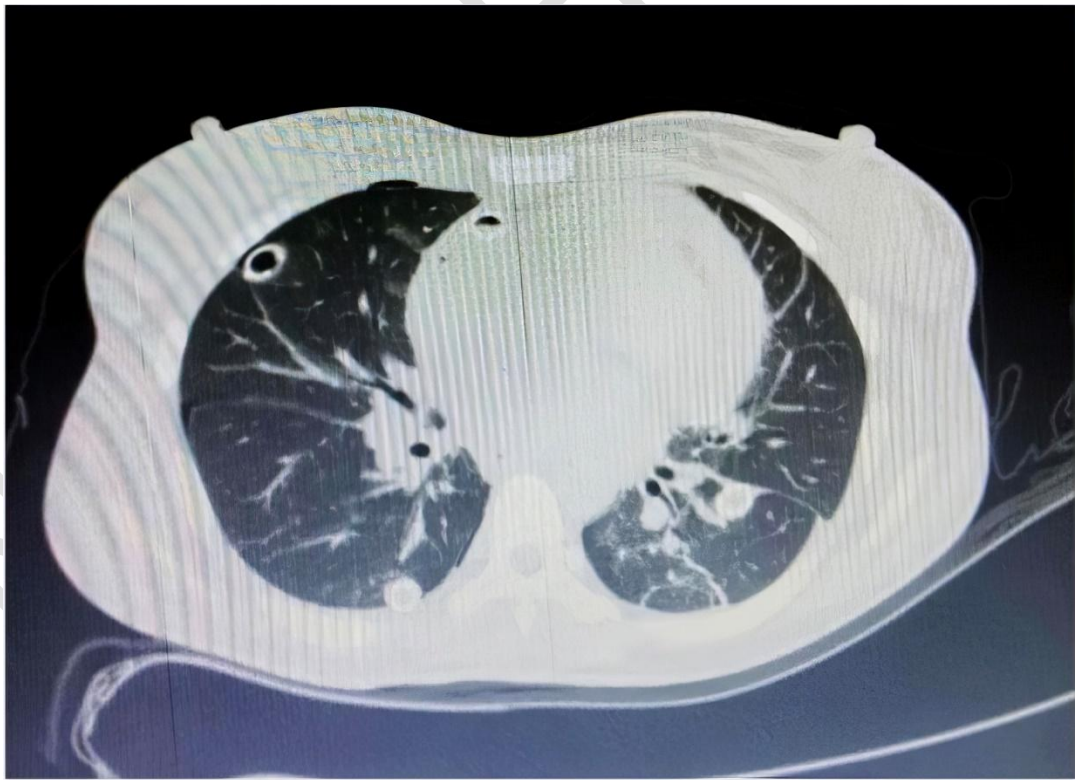

FIG. 3. COMPUTERIZED TOMOGRAPHY AFTER THE PROCEDURE. AXIAL CUT; BILATERAL PLEURAL DRAIN. MEDIASTINAL EMPHYSEMA. PERCUTANEOUS PROBE IN THE SUPERIOR MEDIASTINUM.

3. DISCUSSION

Ludwig's Angina was firstly described by Wilhelm Frederick von Ludwig in 1836, and is characterized as a rapidly progressing cellulitis primarily affecting the submandibular gland region, spreading contiguously without a propensity for abscess formation [4]. Presently, it is recognized as a gangrenous cellulitis that involves the submandibular, sublingual, and submental spaces, displaying a remarkable ability to spread rapidly through adjacent tissues [1]. This condition is of polymicrobial origin and various factors such as alcoholism, malnutrition, diabetes, drug use, and immunosuppression can exacerbate its severity [11].

The etiology of Ludwig's Angina is multifactorial; however, most cases initiate from odontogenic sources. Typically, dental infections are located in the lower second and third molars, where the tooth apices lie below the insertion of the mylohyoid muscle, establishing an intimate anatomical relationship with the submandibular space. Patients may have a history of recent tooth extraction, periodontal status, trauma, or poor oral hygiene, with the latter serving as a potential triggering factor in the presented clinical case [1-3, 5-9]. As the infection progresses into the submandibular space, it can extend further through the styloglossus muscle into the pharyngomaxillary space and subsequently the retropharyngeal space, ultimately affecting the mediastinum. Late diagnosis predominantly contributes to this progression. The most frequently encountered pathogens associated with these infections include *Streptococcus viridans*, *Staphylococcus aureus*, *Peptococcus* spp., and *Bacteroides* spp., which release endotoxins leading to tissue necrosis, local thrombophlebitis, fetid odor, and gas production [11]. Furthermore, it is noteworthy that Ludwig's Angina can also arise from other causes such as buccal floor laceration, lymphadenitis, tonsillitis, and pharyngitis [12].

The primary differential diagnoses to be ruled out in this case include angioneurotic edema, cervical cellulitis, lingual carcinoma, lymphadenitis, peritonsillar abscess, salivary gland abscess, and sublingual hematoma [1]. However, these conditions have been excluded based on the findings from diagnostic procedures such as computed tomography, laboratory tests, and culture. The diagnosis of Ludwig's Angina and its complications relies mainly on clinical manifestations, which encompass symptoms such as pain, cervical region enlargement, dysphagia, odynophagia, trismus, neck stiffness, tongue protrusion, fever, lymphadenopathy, dyspnea, mental confusion, and morbidity [13, 14]. In the reported case, the patient initially presented with indurated edema in the submandibular region and trismus, subsequently progressing to mild dyspnea, lower limb edema, and abdominal distension.

Besides evaluating the clinical presentation, complementary examinations are crucial for accurate assessment and management of the case [9, 11]. In this regard, a complete blood count was performed, serving as an important tool to evaluate the patient's general condition. The obtained results revealed significant leukocytosis with values of 34,400/mm^3, followed by left deviation. Imaging modalities, such as computed tomography, are essential to determine the severity of the infection. This imaging technique enables the identification of airway edema and the localization of any mediastinal fluid collection.

Infections affecting the maxillofacial complex can extend into the deep cervical regions through fascial spaces, leading to severe complications and potentially life-threatening conditions such as intracranial and parapharyngeal abscesses, as well as mediastinitis. These infections spread through various cervical planes and spaces, including the parapharyngeal, carotid, retropharyngeal, anterior neck, back of the neck, parotid, prevertebral, masticatory, and paratracheal regions, often resulting in the formation of abscesses or cellulitis, thereby posing serious risks to the patient's life [13].

When Ludwig's Angina extends into the mediastinal space, it evolves to acute mediastinitis, which should always be considered in patients presenting with fever and signs of head and neck infection, such as deep neck abscesses [14, 15]. This dissemination occurs through the aponeuroses of the vertically oriented muscles, creating a pathway that affects the lateral pharyngeal and retropharyngeal spaces. By traversing structures such as the carotid artery, vagus nerve, and internal jugular vein fascia, the infection reaches the mediastinum, progressing to acute mediastinitis. This condition can later develop into severe complications, including sepsis, pleural effusion, empyema, pericarditis, vessel compression, and even death [16]. It is important to highlight that Ludwig's Angina and mediastinitis can spread through contiguous routes, such as cervical necrotizing fasciitis, as well as through hematogenous and lymphatic routes [17]. In the described case, the patient developed descending necrotizing mediastinitis as a consequence of a submandibular abscess, which represents a rare and severe form of mediastinal infection with a high fatality rate if not promptly and appropriately treated. The treatment protocol consisted of antibiotic therapy, with the primary antibiotics used being ceftriaxone as the first-line agent, followed by clindamycin. Complementary measures employed in conjunction with antibiotic therapy included drainage, debridement, tooth extraction, tracheostomy, and surgical intervention. According to Edetanlen et al. [13], adequate early surgical intervention and intravenous antibiotics are crucial for controlling Ludwig's Angina, especially in resource-limited settings, as the risk of airway compromise outweighs the potential benefit of solely relying on vigilant antibiotic therapy in case of treatment failure. Intravenous antibiotics alone have been associated with a higher incidence of airway compromise compared to those treated with surgical decompression and intravenous antibiotics. Additionally, emergency cricothyroidotomy and tracheostomy are indicated for patients presenting in the later stages of the disease [18]. Surgery is recommended for patients who develop abscesses and do not respond to antibiotics and conservative management. Typically, decompression of the submental, submandibular, and sublingual spaces is achieved through external incision and drainage [19]

Authors referred in our literature review documented that pre-existing medical conditions including diabetes, oral malignancy, dental caries, alcoholism, malnutrition, and immunocompromised status are closely linked to odontogenic infections, although some studies report a low incidence of diabetes in infectious conditions [15,16]. However, McSpadden *et al*. assert that the presence of diabetes can expedite the progression of the lesion [20]. According to Holland et al., any recent infection or lesion in the relevant area can predispose the patient to the development of Ludwig's Angina [16]. Although the clinical evolution of the pathogenesis is widely recognized, Ludwig's Angina rapidly progressive submandibular cellulitis may still be fatal, despite clinical management efforts to mitigate the damage caused by this infection [9,21,22,23, 24].

4. CONCLUSION

Ludwig's Angina and mediastinitis are considered severe conditions characterized by fast evolving and potential fatality if not promptly recognized and appropriately treated. The primary complications involve airway obstruction and deterioration of the patient's overall health status. The diagnoses rely on clinical assessment and should be augmented by diagnostic procedures such as computed tomography, X-ray, and ultrasonography, focusing on the relevant regions of the head, neck, and thorax. Early intervention should entail the administration of empiric antibiotics and maintenance of airway patency to achieve a favorable prognosis. However, these measures cannot replace the importance of thorough drainage, extensive debridement, and excision of necrotic tissue. Intensive and readiness measures, nevertheless, can be unfortunately insufficient to lead to a better prognostic, as seen in the presented case report. Therefore, it is strongly recommended to investigate, manage, and closely monitor odontogenic infections and other infectious conditions affecting the cervical region to avoid the progression to Ludwig's Angina and all associated complications.

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