



### CASE REPORT

## Ludwig's Angina Resulting in Mortality: an Autopsy Case

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### Abstract:

Ludwig's angina (LA) is a form of cellulitis that bilaterally affects the sublingual and submandibular soft tissue above the mylohyoid diaphragm at the base of the mouth. Due to its complications, the mortality rate of up to 50% has declined up to 8% in recent years with the contribution of modern treatment options. In this article, a case of LA resulting with death is presented with autopsy findings. In cases where phlegmon is opened with an intraoral incision, the risk of aspiration pneumonia should be taken into consideration. In order to detect LA and its complications, organs and soft tissues of the oral cavity, neck region and thoracic cavity should be carefully examined in autopsy cases.

**Keywords:** Ludwig's angina, Mediastinitis, Aspiration Pneumonia, Autopsy

### Öz:

Ludwig anjini (LA) ağız tabanında bilateral olarak mylohyoid diaframın üzerindeki sublingual ve submandibuler yumuşak dokuyu tutan bir selülit şeklidir. Komplikasyonları nedeniyle %50'ye varan mortalite oranı çağdaş tedavi seçeneklerinin katkısı ile son yıllarda %8'e kadar gerilemiştir. Bu yazıda ölümle sonuçlanan bir LA olgusu otopsi bulguları eşliğinde sunulmuştur. Ağıziçi insizyonla flegmonun açıldığı olgularda aspirasyon pnömonisi riski göz önünde bulundurulmalıdır. LA ve komplikasyonlarının saptanabilmesi için otopsi olgularında oral kavite, boyun bölgesi, mediasten ve göğüs boşluğundaki organ ve yumuşak dokular dikkatlice incelenmelidir.

**Anahtar Kelimeler:** Ludwig anjini, Mediastinit, Aspirasyon Pnömonisi, Otopsi

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### Conflict of Interest

The authors declare that they have no conflict of interests regarding content of this article.

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The Authors report no financial support regarding content of this article.

### Ethical Declaration

Informed consent was obtained from the participant and Helsinki Declaration rules were followed to conduct this study.

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## 1. Introduction

Ludwig's angina (LA) is an aggressively spreading form of cellulitis that bilaterally involves the sublingual and submandibular soft tissue above the mylohyoid diaphragm at the floor of the mouth (1). It is a rare life-threatening disease which often develops due to odontogenic infections (2, 3). The mortality rate up to 50% due to frequent complications has decreased to 8% in recent years with the widespread use of antibiotics and surgical treatment options (1, 4). LA cases presented with autopsy findings are rare in the literature. In this report, an LA case complicated by mediastinitis, septicemia and aspiration pneumonia and resulting in death is presented with autopsy findings.

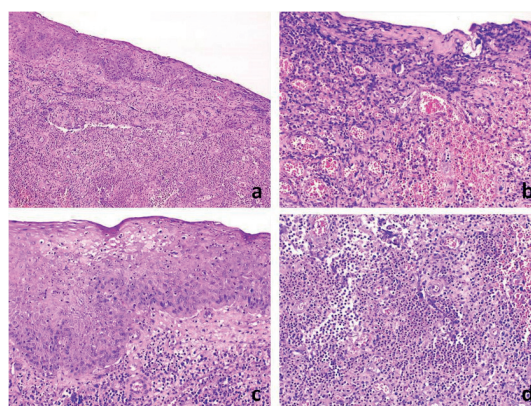
## 2. Case

A 37-year-old male patient admitting to the hospital with complaints of swelling of the floor of the mouth, difficulty of speech, restriction to mouth opening and pain had swelling that mainly involved the submental space of the neck in the physical examination, with fever determined to be 39°C, pulse 110/min, arterial blood pressure 120/80 mmHg. The patient's 34th tooth had been extracted 10 days before. In his history, he had controlled type 1 diabetes. On the first day, the patient was operated with diagnosis of phlegmon of the mouth floor and a large amount of bloody-purulent exudate was aspirated by intraoral incision performed to the mouth floor, and antibiotic treatment was initiated simultaneously. In exudate and blood culture anaerobic streptococcus growth was detected. Despite the treatment in the following days, the patient's fever did not come down and his general condition was evaluated as moderate. On the X-ray, signs of inflammation were observed in the lungs and mediastinum. On the 14th day in the hospital, the patient's condition suddenly worsened and respiratory distress developed. On the seventeenth day, the patient had a sudden cough, followed by a large amount of foul-smelling, green-gray purulent sputum. Meanwhile, the cardiac arrest has been developed in the patient, and despite interventions, biological death occurred.

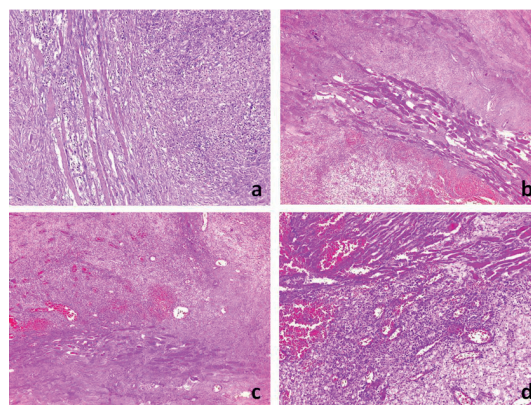
At autopsy, external examination revealed swelling in the submandibular space and cervical region, and crepitation during palpation. In the oral cavity, a dry socket was observed at the level of the 34th tooth, with hyperemia and swelling in the tissues around it, and an incision section with hyperemic and edematous margins at the floor of the mouth. There was abundant accumulation of purulent exudate at the floor of the mouth and partly inside the mouth. The corpse was opened via a single vertical incision. Abundant fibrinopurulent exudate was observed between

the soft tissue and skeletal muscles in the mouth floor and neck area, as well as in the mediastinum. In the sections made in both lungs, it was observed that fibrinopurulent exudate flowed from the sectional surface of the tissue.

In microscopic examination, edema, hyperemia, granulation tissue formation, abundant lymphocyte and neutrophil infiltration, microabscess formations were observed in the soft tissues around the 34th tooth socket, and findings of erosion and ulceration in the surface epithelium (Figure 1). Widespread areas of necrosis and edema were observed in the soft tissue and skeletal muscles of the neck region, as well as abundant lymphocyte and neutrophil infiltration on this base (Figure 2). Considering the autopsy findings, the case was reported as "Ludwig's angina complicated by mediastinitis and aspiration pneumonia due to 34th tooth extraction" and it was concluded that death was natural.



**Figure 1.** In the soft tissue samples taken from around the tooth socket, erosion, ulceration and granulation tissue in the oral mucosa (a, b); in the surface epithelium (c) and subepithelial tissue, widespread lymphocyte/neutrophil infiltration and microabscess formation (a, b, d). (stain: Hematoxylin-Eosin).



**Figure 2.** In the soft tissue samples of the neck region, widespread necrosis and edema (a, b), granulation tissue and bleeding areas (c, d), lymphocyte/neutrophil infiltration (a-d) including skeletal muscles (stain: Hematoxylin-Eosin).

### Ethical Declaration

Informed consent was obtained from the participant and Helsinki Declaration rules were followed to conduct this study.

### 3. Discussion

LA was first described by the German physician Wilhelm Friedrich von Ludwig in 1836 (5). In large-scale studies, the disease was seen to be relatively more common in men than in women, and the average age was reported to be around 44. (3)

The most common cause is dental diseases, particularly lower second and third molar teeth in 90% of the cases (5). In general, any damage and infection in the oral cavity and mandibular region can cause LA. Traumatic injuries to the floor of the mouth, tongue and jawbones, intraoral piercings, peritonsillar abscesses, sialadenitis of the submandibular gland, and infected thyroglossal duct cyst are other causes (5, 6). Cases that develop due to rare causes such as snake bites have been reported in the literature (7). Infections are usually polymicrobial but often include organisms found in the oral flora. Organisms showing the most frequent reproduction in cultures taken from LA cases are *Staphylococcus*, *Streptococcus*, *Peptostreptococcus*, *Fusobacterium*, *Bacteroides* and *Actinomyces* (8). Predisposing factors, along with etiological factors, have an important role in the development of LA. Diabetes, oral malignancies, dental cavities, alcoholism, malnutrition, and immune deficiency are among these factors (5).

The most common findings are fever, chills, neck swelling, odynophagia and dysphagia. Less frequently, complaints such as regional pain, hoarseness, and choking may be encountered (5). On physical examination, swelling, stiffness, tenderness, and crepitation during palpation are observed in the submandibular space, and sometimes on the tongue and floor of the mouth. In LA cases, infection primarily involves the sublingual space and then progresses to the submandibular space, neck and mediastinum. Regional lymphadenopathy is not observed in the patient since LA does not show lymphatic spread (5). In cases with severe airway obstruction, findings such as respiratory movements <25/min and saturation below 95% can be observed (7). Laboratory tests make minimal contribution to specific diagnosis during the diagnostic process. Blood culture is important in terms of showing the hematogenous spread of infection. Radiological methods, such as computed tomography and ultrasonography, are often required to confirm abscess formation and thus to determine the indication for surgical intervention (5, 9).

Early diagnosis and treatment are very important for prognosis in LA cases, as the risk of complications and death is naturally higher in cases diagnosed late (10). Mediastinitis, septic shock, respiratory distress, venous thromboembolism, aspiration pneumonia, aortopulmonary fistula, necrotizing fasciitis are expected complications in LA cases (3, 4, 11). While it has a mortality rate of up to 50% due to its complications, the widespread use of antibiotics and surgical treatment options have decreased this rate to 8% in recent years (1, 4). In a study covering the years 2006-2014 in the USA, the mortality rate was determined as 0.3% (3). Respiratory failure is the most common cause of death in cases resulting in mortality (7).

Autopsy findings of only 4 cases were presented in the English literature (12-15). However, the authors could not reach a publication with a similar content in Turkish. In the autopsy cases reported, swelling of the neck, submandibular space and oral cavity organs, and oral mucosa covered with hyperemic and purulent exudate were prominent findings. Due to the cause of LA, attention should be paid to the presence of local lesions such as infected tooth socket, peritonsillar abscess, bone fracture during autopsy. In cases that are kept for post-mortem, green-brown colored dark spots may be observed due to rapid corpse decay in areas where the purulent exudate accumulates under the skin. In internal examination, specific findings for LA are generally observed in the organs and tissues of the oral cavity and neck region, and in complicated cases, additionally in the mediastinum and thoracic organs. In complications such as septicemia and septicopyemia, widespread inflammatory lesions may be encountered in all internal organs. Neutrophil-weighted inflammatory cell infiltration, colonies of various microorganisms, and additionally, in chronic cases, granulation tissue and fibrosis are expected findings on widespread necrosis base in the specified organs and tissues in microscopic examination (12-15).

### 4. Conclusion

In addition to usual complications such as mediastinitis and septicemia with high risk of complications and mortality, rare complications such as aspiration pneumonia can be seen in LA. In cases where phlegmon is opened with intraoral incision, the risk of aspiration pneumonia should be taken into consideration. In order to detect LA and its complications, oral cavity, neck region, organs and soft tissues in the mediastinum and thorax should be carefully examined in autopsy cases.

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