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CASE REPORT/CASO CLINICO

Ludwig's angina: a case report with a 5-year follow-up

Angina di Ludwig: un caso clinico con 5 anni di follow-up

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KEYWORDS
Ludwig's angina;
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Abstract
Aim: Ludwig's angina is a rare aggressive infection, often of dental origin, characterized by a rapid spread of cellulitis in the submandibular and sublingual spaces. Ludwig's angina is potentially fatal, if it obstructs the airways and if it is not treated with appropriate antibiotic therapy.
Summary: The case report describes the diagnosis and the management of a Ludwig's angina caused by an endodontic infection in a 16 years-old female patient. The infection has been caused by a decay of the second lower right molar. After hospitalization and systemic antibiotic therapy, in accordance with the patient and the parents endodontic and restorative treatments of the tooth were performed. After 3 and 5 years, the radiological examination revealed no periapical lesions around right lower second molar and the presence of lamina dura.
Key learning points: This aggressive infection may often be undervalued and this may cause dangerous consequences to the patient's life. The infection can be prevented by periodic dental care and interventions, which can avoid odontogenic infections. In the case of Ludwig's angina, early diagnosis is fundamental to save the patient's life. After the initial antibiotic therapy and once the life of the patient is no longer at risk, an appropriate endodontic therapy can be considered a valid therapy for this disease.

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PAROLE CHIAVE
Angina di Ludwig;
Terapia endodontica;
Infezione endodontica;
CBCT;
Gestione dell'infezione.

Riassunto
Obiettivi: L'angina di Ludwig è un'infezione aggressiva, spesso di origine odontogena, caratterizzata da una raccolta cellulitica a rapida evoluzione che coinvolge gli spazi sotto-mandibolare e sotto-linguale. Essa può ostruire le vie respiratorie ed essere potenzialmente fatale se non tempestivamente trattata con cure antibiotiche appropriate.
Riassunto: In questo articolo viene descritto un caso di diagnosi e trattamento di un'angina di Ludwig causata da un'infezione odontogena in una ragazza di 16 anni. L'infezione é stata causata da una carie a carico del secondo molare inferiore di destra. Dopo il ricovero ospedaliero ed una terapia antibiotica sistemica parenterale, con il consenso dei genitori abbiamo è stata eseguita una terapia endodontica e la successiva ricostruzione con materiali adesivi dell'elemento dentario in questione. Dopo 3 e 5 anni gli esami radiografici mostrano assenza di lesioni periapicali e la presenza della lamina dura.
Punti chiave di apprendimento: Questa infezione aggressiva è spesso sottovalutata e ciò può causare conseguenze gravi per la vita del paziente. Tali infezioni possono essere prevenute con controlli periodici del cavo orale. In caso di angina di Ludwig una diagnosi tempestiva è fondamentale per salvare la vita del paziente. Dopo l'iniziale terapia antibiotica ed aver messo in sicurezza la salute generale del paziente, la terapia endodontica può essere considerata una valida alternativa per questa patologia.

Introduction

Ludwig's angina is a rare aggressive infection, often of dental origin, characterised by a rapid spread of cellulitis in the submandibular and sublingual spaces.[1,2]

The first case was described by Karl Friedrich Wilhelm von Ludwig in 1836.[3] The most common victim of Ludwig's angina is a male, aged between 20 and 40.[4] The origin of Ludwig's angina is odontogenic in 90% of cases.[5] It is usually the consequence of tooth extraction or infection.[2] The origin is generally the second and third inferior molar (70—80%).[6] In fact, the roots of these teeth penetrate the mylohyoid ridge, such that any abscess or dental infection has direct access to the submaxillary space. The propagation of the cellulitis depends on the anatomy of the neck.[7] Once infection develops, it spreads contiguously to the sublingual space. Infection can also spread contiguously to involve the pharyngomaxillary and retropharyngeal spaces.[8] Other causes of infection have also been reported, such as pharyngeal infection or tonsillitis, infections due to foreign bodies, or infections that are secondary to squamous cell carcinoma, located at the base of the tongue and at the floor of mouth.[8] Other predisposing conditions include poor dental hygiene, dental caries, intravenous drug abuse, malnutrition, diabetes mellitus, AIDS, immunosuppression, and systemic lupus erythematosus. In children, Ludwig's angina can occur without any predisposing condition.[8]

At an initial examination, the general health of the patient is often already clearly compromised but, nevertheless, the local condition is not severe. Bilateral suprahyoid swelling is observed, with a hard, cardboard-like consistency. It is non-fluctuating and painful on palpation. The mouth hangs somewhat open and the tongue is in contact with the palate, with clear oedema of the floor of the mouth. There is difficulty in swallowing and breathing, which are the most salient presenting clinical features of the illness, and this is due to cellulitis, aided by the awkwardness resulting from the position of the tongue.[7,9] It can also highlight erythema and redness of the skin area in front of the neck.

The diagnosis in patients with Ludwig's angina is based on clinical findings. Panoramic radiography can help to discover the origin of the dental infection, while a cervico-thoracic CT scan can help to determine its extent, especially when there is abscess formation.[10,6]

Microbiological investigations are useful to assess the most effective antibiotic therapy. A common cause of Ludwig's angina is a mixture of aerobic and anaerobic bacteria, including, predominately, normal oral flora.[3]

The management of Ludwig's angina involves antibiotics and maintenance of a secure airway to prevent asphyxia, and surgical drainage if necessary.[2,11,12] Intravenous penicillin G, clindamycin, and metronidazole are the antibiotics recommended for use prior to obtaining culture and antibiogram results. Some authors also recommend the use of gentamicin.[13,14,6] Other studies are based on the fact that infections caused by aerobic Gram-negative organisms are uncommon in deep neck abscesses. Thus, the use of gentamicin is not recommended as a first choice in the initial treatment by some authors.[7,15]

Ludwig's angina is potentially fatal, if it obstructs the airways[2,16] and if it is not treated with appropriate antibiotic therapy.[7] Its mortality rate can reach up to 50%.[8] The odontogenic infections that may cause Ludwig's angina can largely be prevented by timely interventions and periodic dental care.[17]

Case report

A 16-year-old female was referred to the author's practice with pain, swelling and difficult swallowing for 1 day. The patient reported that another dentist had performed a pulpotomy and administered a medication with non-specified materials. The patient was taking 1 g amoxicillin per day.

A clinical examination revealed extra-oral bilateral swelling, especially on the right side of the face (Fig. 1). The skin of the perioral tissue and of the neck was red and hot. The temperature of the patient was about 38 °C. The patient had difficulties in opening her mouth. The lateral cervical lymph nodes were positive on palpation. During the visit, there was an increase in breathing frequency (26 breaths per minute) and subsequent increasing difficulty in breathing. Given the severe clinical situation, the patient was referred to the emergency department of Oral and Maxillofacial Surgery where she stayed for 7 days and treated with intravenous antibiotic therapy. During the hospitalization, an orthopantomography and CT scans with and without contrast liquid were performed to visualize the size of the lesion and its relationship with the surrounding anatomical structures (Figs. 2 and 3).

After the acute phase, initially the extraction of the right lower second molar was suggested because the poor endodontic therapy caused the severe clinical situation. A more accurate clinical examination revealed that the tooth had a temporary restoration, the clinical and radiography analysis showed a good residual structure and that extraction was not necessary.

The patient and her parents were advised of the technical difficulties and potential risks of endodontic treatment, of potential new swelling and of the uncertainty recovery. The parents gave written consent for the proposed treatment.

The treatment plan included cleaning of the canal space, endodontic obturation with gutta-percha and direct reconstruction with composite. A preoperative periapical radiographic examination (Soredx, Digora, MI, Italy) (Fig. 4A) showed an abnormal previous access to the endodontic space.

From the initial pre-operative radiograph (Fig. 4A) in not evident a big periapical lesion but we remember that the patient had an abscess that required hospitalization for 7 days (Fig. 1).

At the first session, after a mouth rinse with 0.2% chlorhexidine gluconate (Curasept, Curaden Healthcare, Saronno, VA, Italy) the tooth was anaesthetised with inferior alveolar nerve block and buccal infiltration of 2% articaine containing 1:100,000 epinephrine (Ubistesin 3M ESPE, Neuss, Germany). The tooth was isolated with a rubber dam (Nictone Manufacturera Dental Continental, Zapopan, Jalisco, Mexico). The occlusal access cavity was modified with diamond bur D6C Intensiv (Intensiv, Lugan, Switzerland).

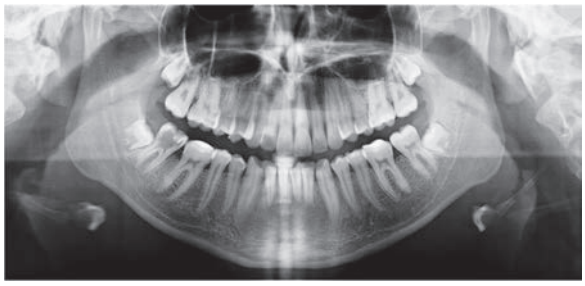

Figure 2 Orthopantomography showing an abnormal access to the endodontic space.

The tooth had three canals: two mesial and one distal. Root canals were cleaned and then the electronic working length was taken with an apex locator (Morita Denta Port ZX, Dietzenbach, Germany). The canals were instrumented using a crown-down technique with ProTaper Universal files (Dentsply Sirona Endodontics, Ballaigues, Switzerland) up to a size F3 (Fig. 4B) and abundant irrigation with 5.25% sodium hypochlorite at 50 °C (Niclor Ogna, Muggió, MB, Italy) was performed. Calcium hydroxide was placed as interappointment dressing (Stomidros Funo, BO, Italy) and the access cavity was temporarily sealed with Cavit G (3M ESPE, Neuss, Germany).

After 7 days, the root canals were irrigated with EDTA (17%, Ogna, Muggió) and 5.25% sodium hypochlorite. The root canals were dried with calibrated absorbent paper points and the canal was obturated with gutta percha and Pulp Canal Sealer (SybornEndo, Amersfoort, Netherland) using the warm vertical compaction with heated pluggers and condensers (Hu-Friedy, MI, Italy). At the next clinical session, the coronal access was adhesively restored with a fibre post (D.T. Light Post, Dentsply, Rome, Italy) resin cement and composite (All Bond 2, Bisco, Schaumburg, IL, USA; Clearfil SA Cement, Kuraray, Hattersheim am Main, Germany; Filtek, 3M ESPE, Neuss, Germany) and an immediate postoperative radiograph was taken (Fig. 4C). All the treatment was performed under magnification (4.3×; Zeiss, Oberkochen, Germany).

After the therapy, the patient moved to another city. After 3 years, the patient came back to the author's practice with a CBCT (Fig. 5A and B) and she gave her consent to take a periapical radiograph (Fig. 5C). The radiological examination revealed no periapical lesions around tooth 47, the presence of *lamina dura* (Fig. 6A—C) and showed the confluence of the mesial canals (Fig. 7).

After 5 years, another periapical control radiograph was taken, revealing a normal periapical status (Fig. 8).

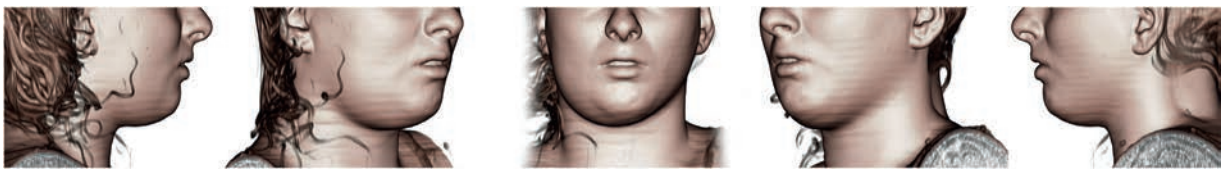

Figure 1 Three-dimensional reconstruction of the patient's face obtained by 3D reprocessing of the CT. Note the strong swelling of the right side.

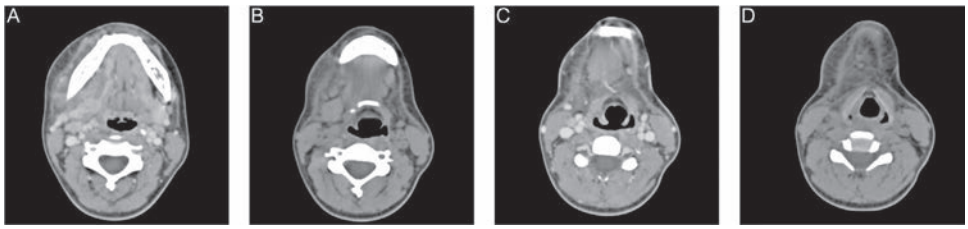

Figure 3 (A—D) Cervico-thoracic CT scan with and with-out contrast fluid showing the extension of cellulitis, the occlusion of esophagus and the deviation of trachea.

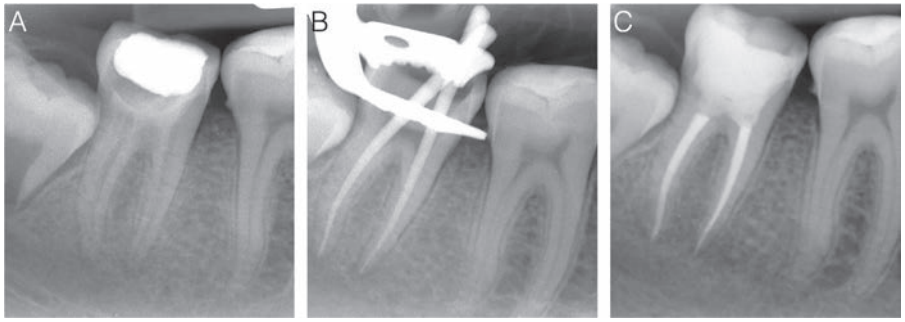

Figure 4 (A) Pre-operative periapical radiograph showing a good residual structure and small periapical lesions; (B) working length with gutta-percha points; (C) final periapical radiograph showing a good compaction of endodontic obturation.

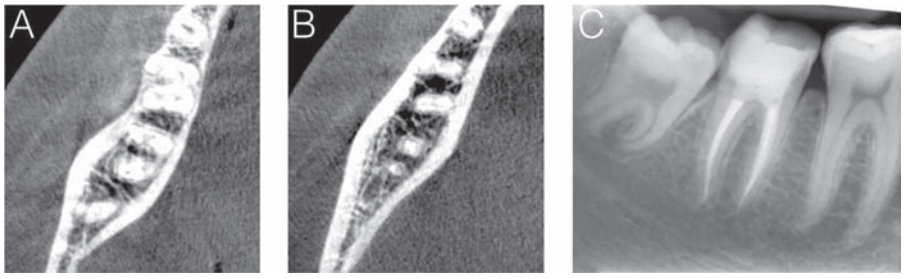

Figure 5 (A and B) Axial CBCT cross-sections showing good endodontic obturation and healing of the bone. (C) Periapical radiograph 3 years after the therapy showing absence of periapical lesions.

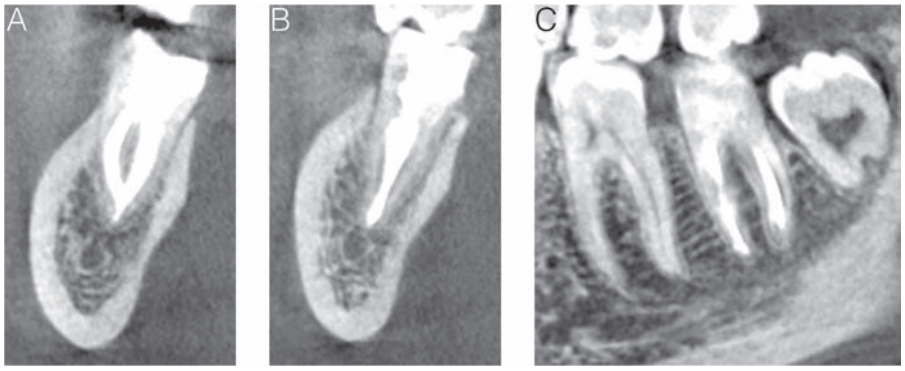

Figure 6 Axial CBCT cross-sections showing of the mesial root (A) and the distal root (B) the absence of periapical lesions around tooth 47 and the presence of *lamina dura*. (C) Sagittal cross-sections confirm the presence of *lamina dura*, the absence of material extrusion of root canal obturation and the proximity of the apical root to the mandibular canal.

Figure 7 Three-dimensional reconstruction obtained from the CBCT of the right lower second molar showing the root canal filling. Note the absence of gaps in the three-dimensional filling and the confluence of the mesial canals.

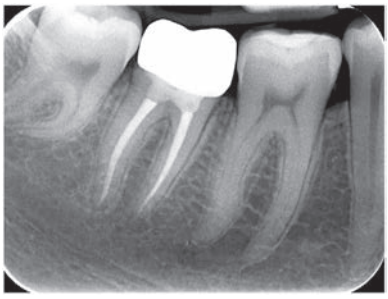

Figure 8 A periapical radiograph 5 years after the therapy showing absence of periapical lesions.

Discussion

Ludwig's angina usually presents in patients with poor dental hygiene, which accounts for approximately 75—95% of cases.[3] The clinical presentation consists of malaise, dysphagia, bilateral cervical swelling, neck tenderness, dysphonia, elevation and swelling of the tongue, pain in the floor of the mouth, sore throat, restricted neck movement and stridor, suggestive of impending airway obstruction.[3,8] The anatomy of the neck is crucial to the understanding of the pathophysiology of Ludwig's angina. Odontogenic infections are the most common cause of Ludwig's angina especially, when the second and third lower molars are involved.[7] Infection in the submandibular space may extend to the lateral pharyngeal and retropharyngeal spaces because these lie below the level of the deep superficial cervical fascia. Infections in the retropharyngeal space can travel downward to the mediastinum and cause acute mediastinitis, empyema, or pericarditis.[7,8]

A mixture of aerobic and anaerobic bacteria can cause the infection. Anaerobic bacteria are responsible for the gas formation in the soft tissues.[18] Dentists should be conscious of the signs of infections, especially those that extend into the deep planes. These signs can include fever, swelling of the floor of the mouth, swelling below the inferior border of the mandible, asymmetric bulging of the pharyngeal walls, and trismus or pain out of proportion to the amount of swelling.[3,7] A cervicothoracic CT scan, with or without contrast fluid, can help to determine the extent of the infection, especially when there is abscess formation.[3] Antimicrobial therapy for odontogenic infections has been described in many other references and is not the focus of this article.

Although many authors recognise the importance of surgical drainage,[8] in this case it was not considered necessary by the maxillofacial surgeons, who focused on the antibiotic therapy and on the evaluation of the vital signs of the patient.

In the literature, there is no previous case of Ludwig's angina with an endodontic origin that has been treated with endodontic therapy, with follow-up.

Conclusions

Ludwig's angina is a rare and not very well known disease that can rapidly progress and can be potentially fatal. This aggressive infection may often be undervalued and this may cause dangerous consequences to the patient's life. The infection can be prevented by timely interventions and periodic dental care, which can avoid odontogenic infections. In the case of Ludwig's angina, early diagnosis is fundamental to save the patient's life. After the initial antibiotic therapy and once the life of the patient is no longer at risk, appropriate endodontic therapy can be considered a valid therapy for this disease.

Conflict of interest

The authors decline any conflicts of interest.

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References

1. Fursta IM, Ersil P, Caminiti M. A rare complication of tooth abscess Ludwig's angina and mediastinitis. *J Can Dent Assoc* 2001;**67**(6):324—7.
2. Chueng K, Clinkard DJ, Enepekides D, Peerbaye Y, Lin VY. An unusual presentation of Ludwig's angina complicated by cervical necrotizing fasciitis: a case report and review of the literature. *Case Rep Otolaryngol* 2012;**2012**:931350.
3. Marcus BJ, Kaplan J, Collins KA. A case of Ludwig angina: a case report and review of the literature. *Am J Forensic Med Pathol* 2008;**29**(3):255—9.
4. Bross-Soriano D, Arrieta-Gomez JR, Prado-Calleros H, Schimelmitz-Idi J, Jorba-Basave S. Management of Ludwig's angina with small neck incisions: 18 years experience. *Otolaryngol Head Neck Surg* 2004;**130**(6):712—7.
5. Moreland LW, Corey J, McKenzie R. Ludwig's angina. Report of a case and review of the literature. *Arch Intern Med* 1988;**148**(2):461—6.

6. Jimenez Y, Bagan JV, Murillo J, Poveda R. Odontogenic infections. Complications. Systemic manifestations. *Med Oral Patol Oral Cir Bucal* 2004;**9**(Suppl. 143—147):139—43.
7. Saifeldeen K. Ludwig's angina. *Emerg Med J* 2004;**21**(2):242—3.
8. Candamourty R, Venkatachalam S, Babu MR, Kumar GS. Ludwig's angina — an emergency: a case report with literature review. *J Nat Sci Biol Med* 2012;**3**(2):206—8.
9. Scully C, Langdon J, Evans J. Marathon of eponyms: 12 Ludwig angina. *Oral Dis* 2010;**16**(5):496—7.
10. Har-El G, Aroesty JH, Shaha A, Lucente FE. Changing trends in deep neck abscess. A retrospective study of 110 patients. *Oral Surg Oral Med Oral Pathol* 1994;**77**(5):446—50.
11. Boscolo-Rizzo P, Da Mosto MC. Submandibular space infection: a potentially lethal infection. *Int J Infect Dis* 2009;**13**(3):327—33.
12. DeAngelis AF, Barrowman RA, Harrod R, Nastri AL. Review article: maxillofacial emergencies: oral pain and odontogenic infections. *Emerg Med Australas* 2014;**26**(4):336—42.
13. Dugan MJ, Lazow SK, Berger JR. Thoracic empyema resulting from direct extension of Ludwig's angina: a case report. *J Oral Maxillofac Surg* 1998;**56**(8):968—71.
14. Hart BT. Tracheotomy for Ludwig's angina. *Oral Surg Oral Med Oral Pathol* 1994;**78**(4):414—5.
15. Parhiscar A, Har-El G. Deep neck abscess: a retrospective review of 210 cases. *Ann Otol Rhinol Laryngol* 2001;**110**(11):1051—4.
16. Diaz Manzano JA, Cegarra Navarro MF, Medina Banegas A, Lopez Meseguer E. Diagnostic and treatment of necrotizing cervical fascitis. Clinical course after a Ludwig angina. *An Otorrinolaringol Ibero Am* 2006;**33**(3):317—22.
17. Allareddy V, Rampa S, Nalliah RP, Allareddy V. Longitudinal discharge trends and outcomes after hospitalization for mouth cellulitis and Ludwig angina. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2014;**118**(5):524—31.
18. Whitesides L, Cotto-Cumba C, Myers RA. Cervical necrotizing fasciitis of odontogenic origin: a case report and review of 12 cases. *J Oral Maxillofac Surg* 2000;**58**(2):144—51.