CASE REPORT Open Access



Ludwig's angina in a child: a case report and literature review

Othmane Benhoummad¹, Kaoutar Cherrabi^{1*}, Najib El Orfi¹, Zineb Mortaji¹ and Mehdi El Fakiri¹

Abstract

Background Ludwig's angina is a diffuse cellulitis in the sub-mandibular space, which extends to the sublingual space. It is an emergency that often occurs in adults as a complication of oral infections. It is rare in children and is particularly life-threatening due to the smaller size, as well as the characteristics in these spaces in a child. This is the case of Ludwig's angina in a 2-year-old boy, with rapid onset of signs of respiratory discomfort, no dental or systemic etiology, and great evolution.

Case presentation A little boy was brought by his mom to the emergency room for the onset a firm swelling in the sub-mental region along with pain and fever, which appeared 3 days prior to the consultation. He was first examined by a pediatrician who prescribed oral broad-spectrum antibiotics (amoxicillin). The symptoms worsened over 48 h, as the little boy presented respiratory discomfort in supine position.

He was admitted in the emergency department. Without dysphagia or respiratory distress.

The clinical examination showed swelling in the sub-mental and sub-mandibular region with/without trismus or signs of oral infection. The laboratory investigations showed hyper-leukocytosis with a microcytic hypochromic anemia. CRP = 300; HIV test was negative.

The computed tomography (CT scan) showed a diffuse abscess in the sub-mental and sub-mandibular and sub-lingual regions. No mediastinal collection was found.

The diagnosis of Ludwig's angina was established.

The patient underwent percutaneous surgical drainage of 15 ml of pus, which alleviated his symptoms, the treatment was carried out through broad-spectrum antibiotics, analgesics, and daily cleaning of the wound and change of surgical dressing. Bacteriological exam found gram-positive cocci in chains. The culture showed a *Staphylococcus aureus*. The patient presented clinical and biological improvement and was discharged after 7 days.

Six months follow-up showed a healthy child, without signs of infection or any other complication.

Conclusion Ludwig's angina in children -however rare- is a potentially life-threatening, rapidly spreading, bilateral swelling of the sub-mandibular. Its management is based on airway control, drainage of the collection and broad-spectrum intravenous antibiotics, as well as surveillance of the biological parameters.

Early diagnosis and appropriate management enhances outcome and decreases mortality significantly.

Keywords Ludwig's angina, Cellulitis, Child, Treatment, Management

*Correspondence: Kaoutar Cherrabi Cherrabi.kaoutar@gmail.com

Background

Ludwig's angina is a rare diffuse cellulitis bilateral in the sublingual and sub-mandibular spaces. It is an emergency that often occurs in adults as a complication of oral infections. It is rare in children and is particularly



¹ ENT and Cervico-Facial Surgery Department, Agadir, Morocco

life-threatening due to the smaller size, as well as the characteristics in these spaces in a child [1-7].

Ludwig's Angina was first described in the fourth century BC by Hippocrates, it was described as an illness that hardens the tongue, which could lead to the patient suffocating rapidly if not relieved [1]. It is a cellulitis that starts in the soft tissue of the sub-mandibular space [1, 2, 7]. Willhelm Frederick Ludwig 1836 described it as a progressive swelling of soft tissues of the floor of the mouth and the neck [4, 7].

Case presentation

A little boy was brought by his mom to the emergency room for the onset a firm swelling in the sub-mental region along with pain and fever, which appeared 3 days prior to the consultation. He was first examined by a pediatrician who prescribed oral broad-spectrum antibiotics (amoxicillin). The symptoms worsened over 48 h, as the little boy presented respiratory discomfort in supine position.

He was admitted in the emergency department. Without dysphagia or respiratory distress.

The clinical examination showed swelling in the submental and sub-mandibular region with/without trismus or signs of oral infection (Fig. 1) The biological investigations showed hyper-leukocytosis with a microcytic hypochromic anemia.CRP = 300; HIV test was negative.

The computed tomography (CT scan) showed a diffuse abscess in the sub-mental and sub-mandibular and sub-lingual regions. No mediastinal collection was found. (Fig. 2).

The diagnosis of Ludwig's angina was established.

The patient underwent percutaneous surgical drainage of 15 ml of pus, which alleviated his symptoms, the treatment was carried out through broad-spectrum antibiotics, analgesics, and daily cleaning of the wound and change of surgical dressing. Bacteriological exam found gram-positive cocci in chains. The culture showed a *Staphylococcus aureus*. The patient presented clinical and biological improvement and was discharged after 7 days.

Six months follow-up showed a healthy child, without signs of infection or any other complication.



Fig. 1 Clinical images

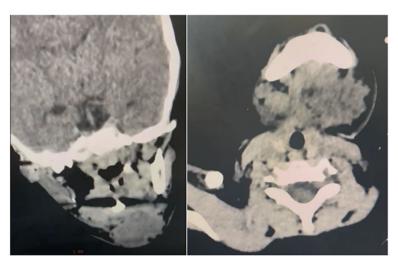


Fig. 2 Radiological images

Discussion

Ludwig's angina is a diffuse cellulitis in the sub-mandibular space, which extends to the sublingual space. It is an emergency that often occurs in adults as a complication of oral infections. It is rare in children and is particularly life-threatening due to the smaller size, as well as the characteristics in these spaces in a child. This is the case of Ludwig's angina in a 2-year-old boy, with rapid onset of signs of respiratory discomfort, no dental or systemic etiology, and great evolution [1–7].

The anatomical feature of the sub-mandibular space and its communication with adjacent spaces explain the disease's evolution and complications [1, 4]. The submandibular's superior limit is the mucosa of the floor of the mouth, it is divided by the mylohyoid muscle into the submandibular and the sublingual spaces [1, 3, 4, 6-8].

The infection of the submandibular space could extend to the sublingual space and cause the tongue to be elevated posteriorly and superiorly, which causes airway way obstruction [1, 4]. The involvement of deep neck spaces is rare but has been described [4, 8].

The frequency of this disease has decreased in the last six decades because of the enhancement of oral hygiene as well as the ubiquitous use of antibiotics [1, 8].

Predisposing factors associated with Ludwig's are poor dental hygiene, diabetes mellitus, obesity, malnutrition, alcoholism, self-medication with non-steroid anti-inflammatory drugs, organ transplantation, and trauma [7, 8].

Two cases in infants younger than four months of age, 50% of cases in children are related to odontogenic etiologies, as opposed to 90% of children, association with systemic diseases -such as diabetes and HIV – is observed in 1/3 of adult cases, versus 1/4 in children [3].

Generally, bacterial culture isolated from surgical drainage of Ludwig's abscesses contains aerobic and anaerobic (e.g., b-hemolytic streptococci, staphylococci) species. Also, gram-negative bacteria have been described in the literature (*Neisseria catarrhalis, Escherichia coli, Pseudomonas aeruginosa*, and *Haemophilus influenza*) [2–6].

Bacterial culture isolates from surgical drainage of Ludwig's abscesses usually have both aerobic (e.g., b-hemolytic streptococci, staphylococci) and anaerobic species [2–6].

Clinical presentation of Ludwig's angina includes focal symptoms, such as tongue pain as well as tooth or throat pain, trismus, dysphagia, dysphonia, and hypersialorrhea. Systemic extension of the infection has been described, as bacterial blood culture has been found positive in 35% of cases in the literature [3, 5, 6, 8].

The most important etiologies are oral infections that originate from the second or third mandibular molar and

spread beyond the mylohyoid muscle to the sublingual space. Also, periapical abscess, penetrating injury of the floor of the mouth, otitis media, and oral neoplasms have also been linked to Ludwig's angina [2–5, 7].

Other etiologies in children include lacerations of the mucosa, sialadenitis, gingivostomatitis, tongue piercing, and infection of vascular malformations [3, 5–7].

The presence of dental caries is associated with a higher risk of airway complications, more frequently encountered in adults [1]. Dental infections are not a prognostic factor in infants younger than 03 years old. A viral infection preceding Ludwig's angina is described especially in children in which oral infections aren't linked as etiologies [1, 8].

Streptococcus species are the most implicated, the second most common agent is Haemophilus influenzae which decreased upon the introduction of the hemophilus influenzae vaccine [1, 7, 8].

The prognosis of Ludwig's angina was fatal in the preantibiotic era with 80% of mortality and has gotten better since then dropping down to 5% [7–9].

In the early stages of the disease, conservative management was sufficient in 2/3 of patients, and only 1/3 of cases in the literature require airway management. Complicated cases present with signs of respiratory distress, and necessitate a team that involves the anesthesiologist [1, 2].

Airway management could resolve in tracheal intubation; flexible nasotracheal intubation requires skill and experience, cricotomy, or even tracheostomy; and the mortality rate is of 2% [2, 4, 8, 9].

Surgical management with surgical drainage and surveillance of surgical wound and general hemodynamic state of the patient [2, 4, 6-9].

Before bacterial culture, intravenous antibacterial agents with wide spectrum are recommended. Penicillin, clindamycin and ciprofloxacin or metronidazole. Some other authors recommend the association of gentamycin. Some reports discussed that the use of steroids avoided the need for airway management [2, 4, 6, 8–10].

Conclusion

Ludwig's angina in children -however rare- is a potentially life-threatening, rapidly spreading, bilateral swelling of the sub-mental, sub-maxillary space, and sub-lingual. Its management is based on airway control, drainage of the collection, and broad-spectrum intravenous antibiotics, as well as surveillance of the biological parameters.

Early diagnosis and appropriate management enhances outcome and decreases mortality significantly.

Acknowledgements

Not applicable.

Authors' contributions

O.B conceived and designed analysis, collected data, performed analysis, wrote the paper, and performed surgery. K.C Performed article redaction. N.E collected radiological and clinical figures. Z.M collected medical history and follow-up information. M.F contributed to designing analysis and supervised redaction. All authors have read and agreed to its content.

Funding

Not applicable.

Availability of data and materials

Not applicable.

Declarations

Ethics approval and consent to participate

Not applicable

Consent for publication

Written informed consent for publication of the patient's clinical details and clinical images was obtained from the parent of the patient.

Competing interests

The authors declare they have no competing interests, financial or non-financial. All authors have read and agreed to its content.

Received: 1 February 2023 Accepted: 23 March 2023

Published online: 26 April 2023

References

- Brotherton H, Templeton K, Da R, Montague M (2014) Ludwig's Angina: Paediatric Case Report and Literature Review. Intern Med 4:1–4
- Kao J-K, Yang S-C (2011) Ludwig's angina in children. JACME 1(1):23–26. https://doi.org/10.1016/j.jacme.2011.07.002
- Candamourty R, Venkatachalam S, Babu MR, Kumar GS (2012) Ludwig's Angina - An emergency: A case report with literature review. J Nat Sci Biol Med 3(2):206–208. https://doi.org/10.4103/0976-9668.101932. (PMID:232 25990;PMCID:PMC3510922)
- Sakhuja A, Shrestha DB, Aryal BB et al (2022) Rare Angina: A Case Report of Ludwig's Angina. Cureus. 14(6):e25873. https://doi.org/10.7759/cureus. 25873.
- Balasubramanian S, Elavenil P, Shanmugasundaram S, Himarani J, Krishnakumar Raja VB (2014) Ludwig's angina: A case report and review of management. SRM J Res Dent Sci 5:211–214
- Pandey M, Kaur M, Sanwal M, Jain A, Sinha SK (2017) Ludwig's angina in children anesthesiologist's nightmare: Case series and review of literature. J Anaesthesiol Clin Pharmacol. 33(3):406–409. https://doi.org/10.4103/ 0970-9185.214318. (PMID: 29109646; PMCID: PMC5672514)
- Lin HW, O'Neill A, Cunningham MJ (2009) Ludwig's angina in the pediatric population. Clin Pediatr (Phila) 48(6):583–587. https://doi.org/10.1177/ 0009922809333095. (Epub 2009 Mar 12 PMID: 19286617)
- Vallée M, Gaborit B, Meyer J, Malard O, Boutoille D, Raffi F, Espitalier F, Asseray N (2020) Ludwig's angina: A diagnostic and surgical priority. Int J Infect Dis 93:160–162. https://doi.org/10.1016/j.ijid.2020.01.028. (Epub 2020 Jan 23 PMID: 31981767)
- Rajib SFA, Banerjee S, Uddin MM, Shameeussalam K, Rahman MM (2016)
 Distribution and Determinants of Ludwig's Angina: Experience of 30
 Cases in Bangladesh. Journal of Current and Advance Medical Research 3(1):16–21
- Lin Q-L, Du H-L, Xiong H-Y, Li B, Liu J, Xing X-H (2020) Characteristics and outcomes of Ludwig's angina in patients admitted to the intensive care unit: A 6-year retrospective study of 29 patients. Journal of Dental Sciences. https://doi.org/10.1016/j.jds.2019.10.004

Publisher's Note

Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

Submit your manuscript to a SpringerOpen journal and benefit from:

- ► Convenient online submission
- ► Rigorous peer review
- ▶ Open access: articles freely available online
- ► High visibility within the field
- Retaining the copyright to your article

Submit your next manuscript at ▶ springeropen.com