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Case Report

Ludwig's angina in children

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Abstract

Ludwig's angina is a potentially life-threatening, rapidly spreading, bilateral cellulitis of the submandibular spaces. It uncommonly occurs in adults and children and its early recognition is paramount. With early diagnosis, airway observation and management, aggressive intravenous antibiotic therapy, and judicious surgical intervention, this disease should resolve without any complications. Here, we report a case of Ludwig's angina in a 14-year-old boy. We also review the relevant anatomy and discuss the clinical presentation and current management of this disease.

Keywords: Abscess; Ludwig's angina; Pediatrics; Submandibular; Submental

1. Introduction

Ludwig's angina is a potentially life-threatening, rapidly spreading, bilateral cellulitis of the submandibular spaces. In the preantibiotic era, the mortality rate of the disease exceeded 50%.[1,2] Since the 1940s, the introduction of antibiotics has improved oral and dental hygiene, and aggressive surgical approaches have significantly reduced this rate.[3] Ludwig's angina is now uncommon in adults and children, therefore, many physicians have limited experience of it.

Ludwig's angina was initially described by the German physician Wilhelm Frederick von Ludwig[4] in 1836 as a rapidly progressive, gangrenous cellulitis and edema of the soft tissues of the neck and floor of the mouth. In 1939, Grodinsky[5] proposed four criteria to distinguish Ludwig's angina from other forms of deep neck abscesses in that the infection must: (1) occur bilaterally in more than one compartment of the submandibular space; (2) produce a gangrenous serosanguinous infiltrate with or without pus; (3) involve connective tissue fascia and muscle but not glandular structures; and (4) spread by continuity rather than by the lymphatics.

Here, we report a case of Ludwig's angina in a 14-year-old boy. We also review the relevant anatomy and discuss the clinical presentation and current management of this disease.

2. Case report

A 14-year-old Minnan Taiwanese boy presented to our pediatric emergency department with progressive submandibular neck swelling that began 6 days earlier (Fig. 1). There was initially a small nodule over the left submandibular area, and he experienced an intermittent high fever up to 39.0°C. The lesion was hard, immovable, and tender. He had toothache that affected his right first molar just 2 days before the nodule appeared. The patient had been seen 4 days earlier by a pediatrician. Upon presenting to the emergency department, he complained of odynophagia, dysphagia, and dysphonia. Trismus (his mouth could only open 2.5 cm) and tongue elevation were found, but no signs of breathing difficulty were noted during the physical examination. In the emergency department, his vital signs were as follows: temperature, 39°C; pulse, 85 beats/min; respiratory rate, 20 breaths/min; and blood pressure, 120/90 mmHg. A complete blood count

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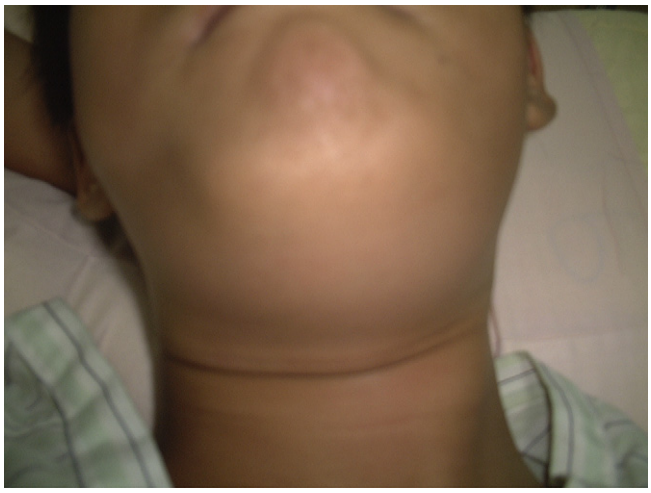

Fig. 1. Progressive submandibular neck swelling of a 14-year-old Minnan Taiwanese boy.

revealed a white cell count of 16,000/μL, consisting of 5.7% lymphocytes, 5.2% monocytes and 88.7% granulocytes, with no bands. An elevated C-reactive protein level of 9.29 mg/dL also was noted. Disseminated intravascular coagulation was not seen. Considering the clinical presentation and laboratory results, deep neck infection was suspected. Computed tomography (CT) revealed thickening of the fascial planes of the submandibular space, suggesting cellulitis, with a 2.6 × 1.5 cm abscess in the left sublingual space (Figs. 2–4). The patient was admitted with a preliminary diagnosis of deep neck infection with abscess formation over the submandibular space. After admission, carbapenem and palliative therapy

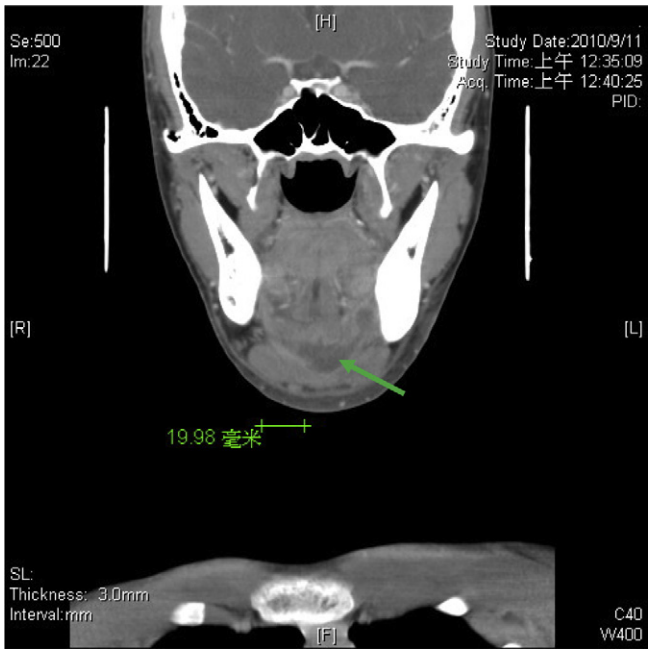

Fig. 3. Coronal section computed tomography scan showing the left sublingual abscess (arrow).

were administered. Dental caries over the first right and left molars were checked by the dentist. Echosonography of the neck showed a submandibular abscess with liquefaction over the lateral portion. No bacterial culture growth was observed in the blood samples.

Upon admission, the patient's airway was closely monitored and the course of treatment proceeded smoothly without

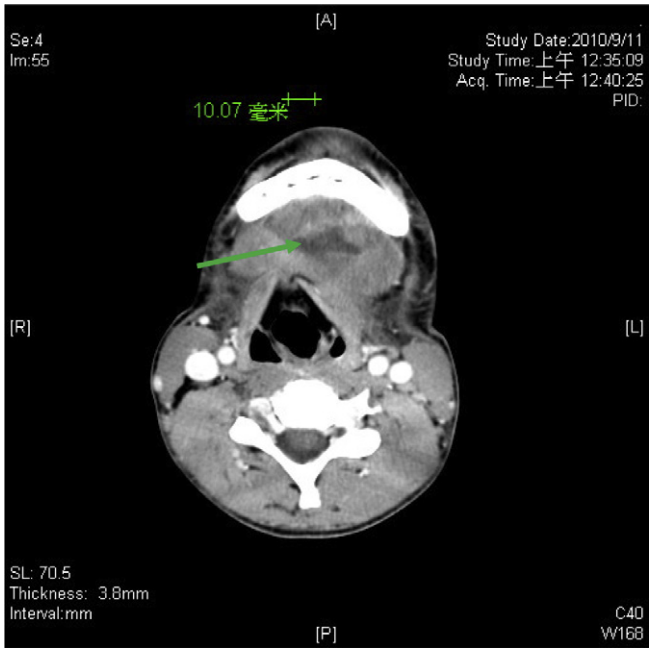

Fig. 2. Cross-sectional computed tomography image revealing submandibular cellulitis and a 2.6 × 1.5 cm abscess in the left sublingual space (arrow).

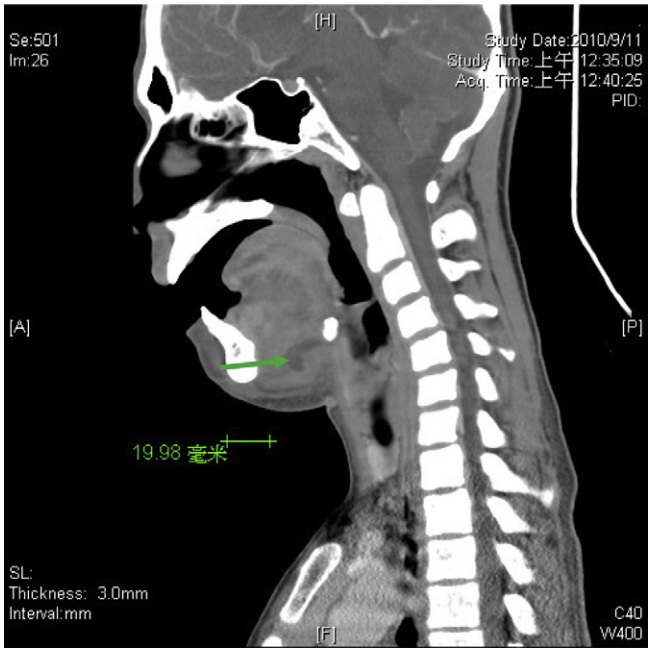

Fig. 4. Sagittal view computed tomography image of the left sublingual abscess (arrow).

complications. The submandibular mass gradually resolved without surgical drainage. The patient was discharged 10 days later.

3. Discussion

The superior border of the submandibular space is formed by the mucosa of the floor of the mouth, whereas the inferior border is formed by the superficial layer of the deep cervical fascia. The submandibular space is divided by the mylohyoid muscle into two continuous compartments: the sublingual and submaxillary spaces. Although one report[6] has shown that the causative infectious pathogens most frequently originated at the second or third mandibular molars and extended below the mylohyoid line of the mandible into the submaxillary space, the dental pathological source of our patient originated at the left first mandibular molar.

A periapical abscess, penetrating injuries of the mouth floor, otitis media, and oral neoplasms have been reported as potential causes of Ludwig's angina.[7] Alternative etiologies in children include oral mucosal lacerations,[8] submandibular sialadenitis,[9] and mandibular fractures.[10] Case reports also have implicated herpetic gingivostomatis,[11] tongue piercing,[12,13] and lymphatic vascular malformation superinfections[14] as causes.

Cellulitis invaded the sublingual space in our case, causing superioposterior displacement of the tongue. Such an occurrence in serious cases might create a potentially life-threatening obstruction of the airway at the level of the oral cavity, oropharynx, and retropharyngeal spaces. If the infection extends posteriorly, it can descend into the superior mediastinum.[4,5]

As many as one in three cases of Ludwig's angina occur in children.[3,15] Two cases have been reported in infants <4 months of age.[16,17] In children, 50% of Ludwig's angina cases have an odontogenic etiology,[3,7] whereas 70–90% of adult cases are odontogenic.[18–20]

One-third of all adult cases of Ludwig's angina are associated with systemic illnesses.[21] However, 25% of pediatric cases of Ludwig's angina do not have predisposing or precipitating causes.[3,7,15] In children, Ludwig's angina develops concomitantly with systemic diseases, such as immune deficiency[22] and diabetes mellitus.[3]

Bacterial culture isolates from surgical drainage of Ludwig's abscesses usually have both aerobic (e.g., β-hemolytic streptococci, staphylococci) and anaerobic species.[7] Gram-negative bacteria, such as *Neisseria catarrhalis*, *Escherichia coli*, *Pseudomonas aeruginosa*, and *Haemophilus influenzae*, have also been reported in such isolates.[23] One study[7] has suggested that these patients need to be closely monitored for hemodynamic sequelae of sepsis, because 35% have positive blood culture results. However, as in our case, another study[24] has reported that 83% of blood samples from patients with Ludwig's angina did not show any bacterial growth.

Patients with Ludwig's angina typically present with focal and systemic signs and symptoms. Focal symptoms can include tongue and tooth pain, throat pain, dysphagia, trismus, dysphonia, and drooling, and are often accompanied by local physical examination findings such as progressive bilateral submandibular and submental neck swelling, firm induration of the floor of the mouth, and edematous posterior and superior displacement of the tongue (e.g., protrusion or elevation).[21]

Frequently encountered systemic signs and symptoms include fever, chills, malaise, dehydration, and an ill appearance. More serious findings such as dyspnea, cyanosis, stridor, and tongue displacement imply an impending airway crisis. The early signs and symptoms of obstruction might be subtle.

Early recognition of Ludwig's angina is crucial for initiation of proper medical therapy and timely consultation for emergency and surgical treatment. Airway compromise was the leading cause of death in the early 1900s, at which time, 67% of patients with Ludwig's angina required anticipatory or emergent intubation.[24] Since 1943, antimicrobial therapy has reduced the frequency of airway intervention to <50%.[19] Complications of Ludwig's angina include sepsis, pneumonia, asphyxia, empyema, pericarditis, mediastinitis, and pneumothorax. The mortality rate from Ludwig's angina is currently 10–17% in the pediatric population.[7,15]

A patient with Ludwig's angina should be primarily assessed for airway stability. Currently, airway observation is an important component in the care of pediatric patients due to the widespread reluctance for immediate airway intervention. In a retrospective review,[15] 10% of children with Ludwig's angina needed airway control, whereas 52% of patients >15 years of age underwent tracheostomy. Children with significant oropharyngeal edema should sit upright and be closely monitored.

Early and aggressive antibiotic therapy must be designed to eradicate both aerobes and anaerobes. Penicillin or a penicillin derivative, with or without additional anaerobic coverage with clindamycin or metronidazole, is frequently used.[3,7,15,18,23] Empiric antibiotic treatment in an immunocompromised host is considered safe. Intravenous steroids decrease edema and cellulitis, which helps maintain airway integrity, improves antibiotic penetration into the infected area, and reduces the length of hospital stay.[23]

There is no preferential formation of abscesses in Ludwig's angina.[1] Surgical decompression of the cellulitis is reserved for cases that are unresponsive to medical therapy or for patients who show clinical evidence of localized abscess formation upon initial physical examination.[7] Contrast-enhanced CT imaging should be considered to assess the extent of the abscess and detect possible odontogenic etiology.[25] If a dental infectious source or involvement of the parapharyngeal, retropharyngeal, or mediastinal space is noted, immediate consultation with the relevant surgical services is indicated.

Finally, adequate nutrition and hydration should be provided to patients with Ludwig's angina, especially children.

4. Conclusion

Although modern medical and surgical interventions for Ludwig's angina have improved outcomes, it remains a potentially lethal disease in the pediatric population.[3,15,18] Early

recognition of the disease is paramount. Pediatricians and physicians should consider Ludwig's angina when patients present with symptoms such as recent oral cavity and neck swelling, even when no offending pathologies are immediately apparent. With early diagnosis, airway observation and management, aggressive intravenous antibiotic therapy, and judicious surgical intervention, this disease should resolve without any complications.

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References

1. Burke J. Angina ludovici: a translation, together with biography of Wilhelm F.V. Ludwig. *Bull Hist Med*. 1939;7:1115−1126.
2. Williams AC. Ludwig's angina. *Surg Gynecol Obstet*. 1940;70:140−149.
3. Patterson HC, Kelly JH, Strome M. Ludwig's angina: an update. *Laryngoscope*. 1982;92:370−377.
4. Muckleston HW. Angina Ludovici and kindred affections: historical and clinical study. *Ann Otol Rhinol Laryngol*. 1928;37:711−735.
5. Grodinsky M. Ludwig's angina: an anatomical and clinical study with review of the literature. *Surgery.* 1939;5:678−696.
6. Tschiassny K. Ludwig's angina: an anatomic study of the lower molar teeth in its pathogenesis. *Arch Otolaryngol*. 1943;38:485−496.
7. Britt JC, Josephson GD, Gross CW. Ludwig's angina in the pediatric population: report of a case and review of the literature. *Int J Pediatr Otorhinolaryngol*. 2000;52:79−87.
8. Gross SJ, Nieburg PI. Ludwig angina in childhood. *Am J Dis Child*. 1977; 131:291−292.
9. Lerner DN, Troost T. Submandibular sialadenitis presenting as Ludwig's angina. *Ear Nose Throat J*. 1991;70:807−809.
10. Rosen EA, Schulman RH, Shaw AS. Ludwig's angina: a complication of a bilateral mandibular fracture: report of case. *J Oral Surg*. 1972;30: 196−200.
11. Chen CJ, Huang YC, Lin TY. Ludwig's angina following herpetic gingivostomatitis in a toddler with tetralogy of Fallot. *J Formos Med Assoc*. 2004;103:311−313.
12. Perkins CS, Meisner J, Harrison JM. A complication of tongue piercing. *Br Dent J*. 1997;182:147−148.
13. Keogh IJ, O'Leary G. Serious complication of tongue piercing. *J Laryngol Otol*. 2001;115:233−234.
14. Tasca RA, Myatt HM, Beckenham EJ. Lymphangioma of the tongue presenting as Ludwig's angina. *Int J Pediatr Otorhinolaryngol*. 1999;51: 201−205.
15. Kurien M, Mathew J, Job A, Zachariah N. Ludwig's angina. *Clin Otolaryngol Allied Sci*. 1997;22:263−265.
16. Steinhauer PF. Ludwig's angina: report of case in a 12-day-old boy. *J Oral Surg*. 1967;25:251−254.
17. Chou Y, Lee C, Chao H, Chao Hai-Hsuan. An upper airway obstruction emergency. Ludwig angina. *Pediatr Emerg Care*. 2007;23:892−896.
18. Srirompotong S, Art-Smart T. Ludwig's angina: a clinical review. *Eur Arch Otorhinolaryngol*. 2003;260:401−403.
19. Moreland LW, Corey J, McKenzie R. Ludwig's angina. Report of a case and review of the literature. *Arch Intern Med*. 1988;148:461−466.
20. Quinn FB. Ludwig's angina. *Arch Otolaryngol Head Neck Surg*. 1999; 125:599.
21. Finch RG, Snider GE, Sprinkle PM. Ludwig's angina. *JAMA*. 1980;243: 1171−1173.
22. Barkin RM, Bonis SL, Elghammer RM, Todd JK. Ludwig angina in children. *J Pediatr.* 1975;87:563−565.
23. Busch RF, Shah D. Ludwig's angina: improved treatment. *Otolaryngol Head Neck Surg*. 1997;117:S172−S175.
24. Har-El G, Aroesty JH, Shaha A, et al. Changing trends in deep neck abscess. A retrospective study of 110 patients. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 1994;77:446−450.
25. Parhiscar A, Har-El G. Deep neck abscess: a retrospective review of 210 cases. *Ann Otol Rhinol Laryngol*. 2001;110:1051−1054.