An Upper Airway Obstruction Emergency
Ludwig Angina

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Abstract: Ludwig angina remains a potentially lethal disease, rapidly spreading bilateral cellulitis of the submental, sublingual, and submandibular spaces, which bears the threat for rapid airway obstruction. Since the introduction of antibiotics in 1940s, the mortality was reduced significantly. This resulted in the rare occurrence of the disease, leaving many physicians with limited experience of Ludwig angina. Although the occurrence of Ludwig angina in adults is rare, its presence in the pediatric edentulous population is even more uncommon. Because the unfamiliarity with this disease is now increasing, unnecessary delaying diagnosis or inadequate management may occur and may result in serious complications. This presentation will consist of a historical review, discussion of pathophysiology, followed by clinical presentation, etiology, bacteriology, and management. With early diagnosis, airway observation and management, aggressive intravenous antibiotic therapy, and judicious surgical intervention, the disease should resolve without complications. In addition, the source of infection should be determined and eliminated if possible. A thoughtful, individualized management strategy seems to be the most reasonable approach to the disease.

Key Words: Ludwig angina, cellulitis, deep neck infection

Ludwig angina is a potentially life-threatening, rapidly spreading bilateral cellulitis of the submental, sublingual, and submandibular spaces, which bears the threat for rapid airway obstruction. "Angina" is from "anchone," the Greek word for strangulation, and is taken to connote throat pain and infection. Ludwig angina was initially described by German physician Wilhelm Frederick von Ludwig in 1836 as a rapidly progressive gangrenous cellulitis and edema of the soft tissues of the neck and floor of the mouth.[1] It originates in the submandibular gland region that extends by continuity, rather than lymphatic spread, and shows no special tendency to abscess.[2] Despite attempts at treatment, the disease was frequently fatal, giving rise to mortality rates exceeding 50% during the preantibiotic era.[3] Airway compromise has been recognized as the leading cause of death in the early 1900s.[4] Since the introduction of antibiotics in 1940s, improved oral and dental hygiene and aggressive surgical approach, the mortality was reduced significantly.[5,6] This resulted in the rare occurrence of the disease, leaving many physicians with limited experience of Ludwig angina. Because the unfamiliarity with this disease is now increasing, unnecessary delaying diagnosis or inadequate management may occur and may result in serious complications. Proper diagnosis, airway control, antibiotic therapy, and occasionally surgical management are essential to ensure the safety of the patient.

To our knowledge, the last and the only Ludwig angina incident in the pediatric population aged younger than 4 months was reported about 40 years ago.[7] Although the occurrence of Ludwig angina in adults is rare, its presence in the pediatric edentulous population is even more uncommon. We report a case of Ludwig angina in a 4-month-old male infant and review the presentation and management of this disease.

CASE

A 4-month-old male infant presented to the emergency department around 11:00 PM with fever of 39.5°C, irritability, and decrease in oral intake during the previous 24 hours. The patient had no significant trauma, or medical or surgical history. On examination, the infant was feverish (39.1°C) and had a pulse rate of 96 beats/min, respiratory rate of 36 breaths/min, and blood pressure of 80/60 mm Hg. Laboratory data revealed a white blood cell count of 16,320 cells/mL with a moderate left shift. The injected throat and severe oral thrush were noted as well. Then the patient was admitted to the general pediatric ward under the impression of acute pharyngitis.

Eight hours after the admission, the medical staff noticed swelling of his left submandibular area that rapidly progressed to the submental area. On physical examination, the infant had swelling, erythema, warmth, and tenderness in the submandibular area (Fig. 1). The patient was immediately transferred to the pediatric intensive care unit where the infant had the oxygen saturation of 92% to 95% in room air and 99% on 10 L/min oxygen. He was noted to have a body temperature of 38°C, pulse rate of 98 beats/min, respiratory rate of 38 breaths/min, and mild respiratory distress. The infant was uncomfortable and preferred to keep his mouth open. Bilateral generalized swelling over the submandibular and submental area were noted. Intraoral examination revealed a firm and tender swelling of the mouth floor, which elevated the patient's tongue upward and posteriorly (Fig. 2). The neck radiographs revealed prominence of the submandibular region's soft tissue (Fig. 3). The sonograph of the neck showed diffuse subcutaneous and soft tissue edematous change without any

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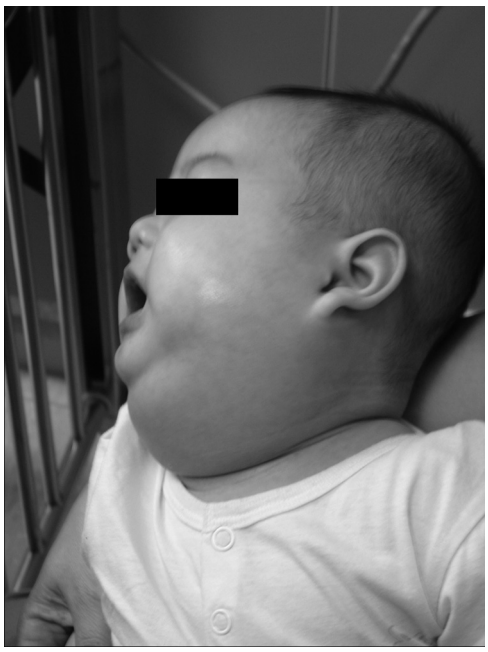

FIGURE 1. Marked swelling over submental and submandibular region.

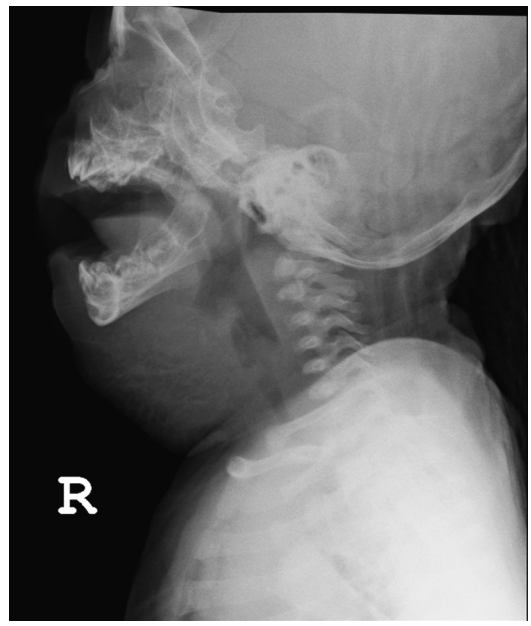

FIGURE 3. The prominence of the submandibular region soft tissue.

evidence of abscess or discrete fluid collection. The computed tomographic scan of the neck demonstrated marked soft tissue inflammatory process with edematous change of bilateral parotid gland region and extension to the parapharyngeal and carotid spaces (Fig. 4).

Our immediate management for the patient was to maintain his upright posture with continuous oxygen flow administration. The General Surgery and Ear-Nose-Throat services were consulted immediately. The patient was given intravenous vancomycin, ceftriaxone, and hydration. Ludwig angina was diagnosed clinically. The emergent airway management and surgical intervention were reserved with close observation. The General Surgery and Ear-Nose-Throat teams were standing by in case of airway compromise. The illness resolved after initial treatment with intravenous empiric antibiotic followed by oral sodium fusidate. Furthermore, the blood culture was sterile. The further laboratory workups for the underlying systemic illness such as immune globulins and complement levels revealed negative findings. The definite etiology in this patient is unknown.

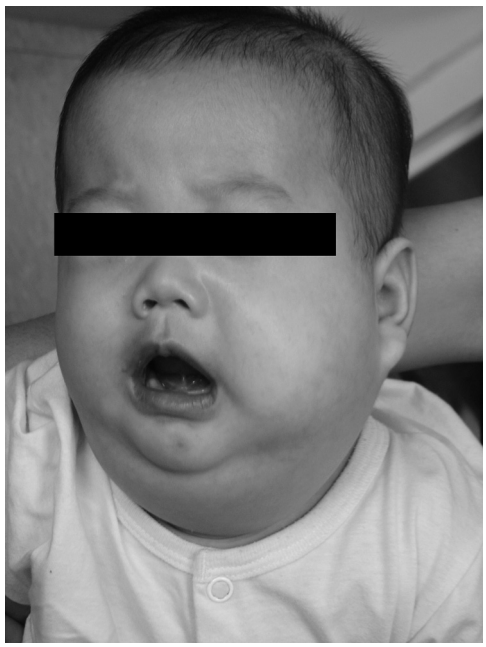

FIGURE 2. Elevated tongue caused by the swelling of the mouth floor.

DISCUSSION

To understand the pathophysiology of Ludwig angina, it is necessary to know the anatomy of the submandibular space. This space has a superior border formed by the mucosa of the floor of mouth and an inferior border formed by the superficial layer of the deep cervical fascia as it extends from the hyoid bone to the mandible. This space is subdivided by the mylohyoid muscle into 2 spaces: the submaxillary space below the mylohyoid and the sublingual space above the mylohyoid. The submaxillary and sublingual spaces are effectively in continuity with one another as infection can spread around the free posterior edge of the mylohyoid. In addition, extension of infection posteriorly along the intrinsic tongue musculature can lead to involvement of the parapharyngeal space and the retropharyngeal space. If there is posterior extension of infection, then there is potential for descent of the infection into the superior mediastinum.[8,9]

Grodinsky[10] in 1939 proposed 4 criteria to distinguish Ludwig angina from other forms of deep neck abscesses. The infection must (1) occur bilaterally in more than 1 space, (2) produce gangrenous serosanguinous infiltration with or without pus, (3) involve connective tissue fascia and

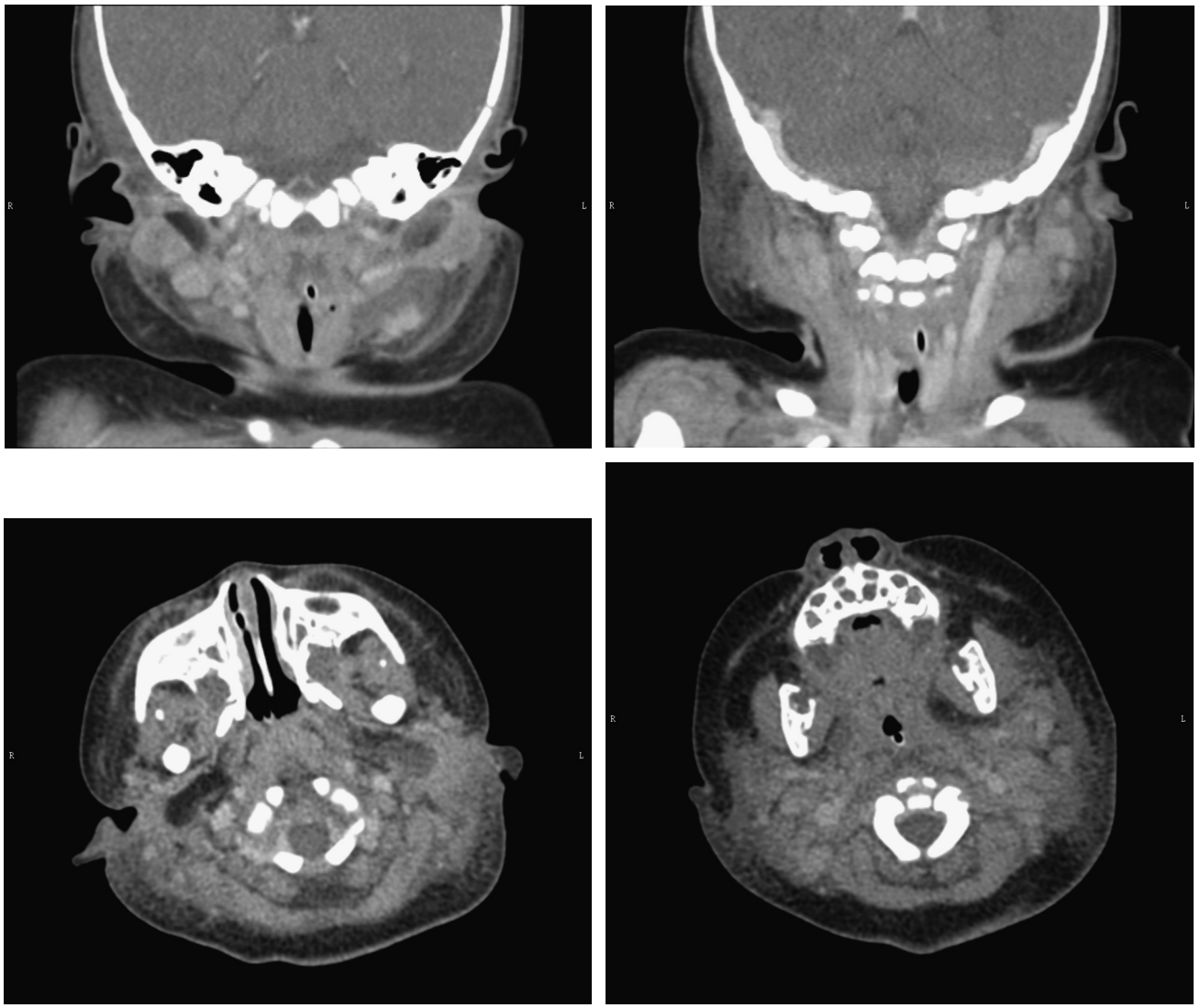

FIGURE 4. Marked soft tissue inflammatory process with edematous change of bilateral parotid gland region and extension to the parapharyngeal and carotid spaces.

muscle but not glandular structures, and (4) spread by continuity, not by lymphatics.

Most Ludwig angina infections are odontogenic, accounting for approximately 75% to 90% of cases.[11–13] Tschiassny[14] described how the roots of the second and third lower molars penetrate the thin inner cortex of the mandible and extend inferiorly to the insertion of the mylohyoid muscle. A periapical abscess can result in an infection of the submandibular space. Mandibular trauma, penetrating injuries of the mouth floor, oral laceration, otitis media, oral neoplasms, and submandibular sialadenitis have all been reported as potential causes of Ludwig angina.[15–17] Systemic illness such as diabetes mellitus, malnutrition, alcoholism, compromised immune system such as acquired immune deficiency syndrome, and organ transplantation and trauma are also common precursors to today's cases of Ludwig angina. In fact, one third of the cases of Ludwig angina are associated with systemic illness. One review reporting the incidence of various illnesses associated with Ludwig angina found that 18% of cases involved diabetes mellitus, 9% involved acquired immune deficiency syndrome, and another 5% were human immunodeficiency virus positive.[18,19] Another review article revealed that about 25% of Ludwig angina cases in the pediatric population demonstrated no etiologic factors.[15] Early recognition of the disease is the most important. Painful neck swelling, tooth pain, dysphagia, dyspnea, fever, and malaise are the most common complaints. Neck swelling and a protruding or elevated tongue are seen in most cases.[20] Stridor, trismus, cyanosis, and tongue displacement suggest an impending airway crisis. Early signs and symptoms of obstruction may be subtle. Complications of Ludwig angina include sepsis, pneumonia, asphyxia, empyema, pericarditis, mediastinitis, and pneumothorax.[15]

The bacterial isolates vary and are often mixed, comprising both aerobes and anaerobes: α-hemolytic streptococci,

staphylococci, and *Bacteroides*. Other anaerobes such as peptostreptococci, peptococci,[21–23] *Fusobacterium nucleatum*, *Veillonella* species, and spirochetes are also seen.[13] Deep neck infections generally involve oral flora. Obligate anaerobes constitute approximately 75% or normal oral flora. A foul breath odor usually indicates the presence of an anaerobe. Two thirds of deep neck infections are polymicrobial. Gram-negative organisms such as *Neisseria catarrhalis*, *Escherichia coli*, *Pseudomonas aeruginosa*, and *Haemophilus influenzae* have also been reported.[21] Blood cultures of Ludwig angina patients are usually negative; in a recent review, 83% of cases reported no growth from blood samples.[23]

The treatment of Ludwig angina has 4 principles. First, sufficient airway management is essential. A review reported that 67% of patients with Ludwig angina required either anticipatory or emergent intubation.[23] Recommendations concerning management of the airway vary throughout the literature and are based upon each author's personal experience and resources available at the time of publication. The trend toward airway observation associated with Ludwig angina has emerged as antimicrobial therapy has become increasingly available and has resulted in the reduction of airway intervention to less than 50% since 1943.[13] Airway observation may play an important role in pediatric patients particularly. In a retrospective review of 41 patients longer than a 13-year-period, only 10% of children with Ludwig angina needed airway control, whereas 52% of patients older than 15 years had tracheostomy.[24] Articles in support of airway observation uniformly propose this as an active form of airway management, recommending airway intervention for those patients demonstrating objective findings of compromise. Stridor, difficulty managing secretions, and cyanosis are the late manifestations of impending airway obstruction. Airway management should be tailored to each patient and to the experience of the treating physician.[25,26]

Second, early and aggressive antibiotic therapy must be designed to cover both aerobes and anaerobes. Penicillin with metronidazole is the first-line empiric therapy. For those patients allergic to penicillin, clindamycin can be used instead. Cefoxitin or combination drugs such as ticarcillin/clavulanate, piperacillin/tazobactam, ampicillin/sulbactam, or amoxicillin/clavulanate are also considered highly effective.[22] Empiric antibiotic treatment in the immunocompromised host is considered safe; however, medical therapy must be guided by cultures, clinical progression of disease, and sensitivities in the host. Intravenous dexamethasone, given every 6 hours for 48 hours, has been beneficial in decreasing edema and cellulitis, which helps maintain airway integrity and improves penetration of antibiotics in the area.[21]

Third, in the past, incision and drainage of abscesses were routine. Now, surgical therapy is usually reserved for cases of medical treatment failure and infections forming localized abscess collection. If indicated, surgical decompression and drainage are performed with removal of all offending teeth in the first 24 to 48 hours.[21,27]

Finally, adequate nutrition and hydration support would be important in patients with significant oropharyngeal edema, especially the young children. Furthermore, the patient must be maintained in a sitting posture and should never be left unattended. The use of contrast-enhanced computed tomography may not be essential to confirm the diagnosis of Ludwig angina, but it is used to assess the extent of the abscess in all cases of retropharyngeal extension.[28] In addition, the source of infection should be determined and eliminated if possible.

CONCLUSIONS

Despite improved outcomes, Ludwig angina remains a potentially lethal disease. With early diagnosis, airway observation and management, aggressive intravenous antibiotic therapy, and judicious surgical intervention, the disease should resolve without complications. Cautious airway observation may be crucial in pediatric patients with Ludwig angina. A thoughtful, and individualized treatment plan that takes into consideration of the patient, setting, and physician experience seems to be the most reasonable approach to the management of the disease.

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