Case Report

Neonatal Ludwig's angina with a whirlwind progression and catastrophic outcome: A case report

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ABSTRACT

Ludwig's angina is a rapidly progressive life-threatening disease, characterized by gangrenous cellulitis and edema of the soft tissues of the neck and the floor of the mouth. The incidence is very rare in children and even rarer in neonates, with sparse case reports. Early diagnosis is crucial in anticipating airway compromise and early management. We are reporting a case history of an 11-day old neonate who presented with an excess cry of 1-day duration and suspected sepsis. Gradually, he developed stridor and swelling was noted in the submental area, which got progressed to a state of airway compromise and multiorgan dysfunction over a few hours. Drainage of the submental area yielded 10 ml pus which grew methicillin-resistant *Staphylococcus aureus* later. The baby succumbed to death within 12 h of the onset of symptoms.

Key words: Airway, Ludwig's angina, Neonatal stridor

Ludwig's angina is a rapidly progressive and potentially life-threatening disease, characterized by gangrenous cellulitis and edema of the soft tissues of the neck and the floor of the mouth. The soft-tissue elevation causes the posterior displacement of the tongue, resulting in airway obstruction. In adults, it is mostly odontogenic in origin, primarily resulting from infections of the second and third molars. However, the incidence is less in the post-antibiotic era due to timely use of appropriate antibiotics in the at-risk cases and also due to improved dental care. The roots of these teeth penetrate the mylohyoid ridge in such a way the submaxillary space becomes vulnerable to an abscess or dental infection [1].

The incidence is very less in children and even rarer in neonates. We could not find any reported case of Ludwig's angina in neonates, though a reference to the same described in a 12-day-old neonate came across in our reading of related literature [2]. Here, we describe a case of Ludwig's angina in a term neonate on the 11th day of life to highlight the need for a high index of suspicion in a neonate with stridor and the possibility of the existence of this entity even in the neonatal period. Because of the unfamiliarity with this disease, unnecessary delay in the diagnosis and inadequate management may occur, resulting in serious complications. Early anticipation, precautions for airway control, antibiotic therapy, and occasionally surgical management are essential to ensure the safety of the patient [3].

Access this article online	
Received - 27 March 2022 Initial Review - 18 April 2022 Accepted - 22 April 2022	Quick Response code
DOI: 10.32677/ijch.v9i4.3411	

CASE REPORT

An 11-day-old baby was brought to our out born unit with complaints of poor feeding and an irritable cry of 1-day duration. Although the baby was feeding well after admission, he had episodes of inconsolable crying in between. Intravenous antibiotics (piperacillin-tazobactam and amikacin) were started for suspected sepsis. Among the sepsis markers, only C-reactive protein was elevated. Lumbar puncture showed no evidence of meningitis. Blood culture and cerebrospinal fluid culture were negative. The baby was hemodynamically stable. By the 3rd day of admission, the baby was feeding well and had no significant cry. Discharge was planned after 5 days of antibiotics.

On the morning of the day when the baby was to be discharged, we noticed mild stridor. Stridor was attributed to the baby's mild micrognathia and suspected congenital laryngeal stridor. Opinion

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was sought from otolaryngology for the same, but nothing peculiar was found. By evening, the baby had a small swelling in the submental area. There was no redness or elevated temperature over the swelling. The otolaryngology and pediatric surgery departments were consulted again and expectant management was advised. By late night, the swelling progressed rapidly and the baby went into respiratory distress. The position of the swelling and rapid deterioration of the condition of the baby led us to suspect the rare possibility of Ludwig's angina. Ultrasound of the neck confirmed the findings. The baby was not clinically stable for a CT scan.

Antibiotics were upgraded to intravenous meropenem and vancomycin. Because of increasing stridor and respiratory distress, intubation was planned. However, intubation posed difficulty as the entire larynx appeared swollen and intubation was possible only with a 2.5 size endotracheal tube. 10 ml of pus was drained by the pediatric surgeon from the submental region. Although all supportive management was given, the baby went into septic shock and multiorgan failure within a couple of hours. Despite our best efforts, we lost the baby.

The total event duration from the onset of stridor to death was hardly 18 h and from the appearance of swelling to death was 11 h. The pus culture grew methicillin-resistant *Staphylococcus aureus*. The source of infection could not be traced. After the event, we thought of the possibility of otitis media preceding the event because the baby had an inconsolable cry for 2 days.

DISCUSSION

Ludwig's angina was initially described by the 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 [4]. As many as one in three cases of Ludwig's angina occur in children [5]. It originates in the submandibular gland region and extends by continuity, rather than lymphatic spread, involving bilateral submental and sublingual spaces also with no special tendency to an abscess [3]. The submandibular space has a superior border formed by the mucosa of the floor of the 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 two 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, an extension of infection posteriorly along the intrinsic tongue musculature can lead to the involvement of the parapharyngeal space and the retropharyngeal space. If there is a posterior extension of infection, then there is potential for the descent of the infection into the superior mediastinum [6,7]. The criteria proposed by Grodinsky in 1939 to distinguish Ludwig angina from other forms of deep neck abscesses are as follows: 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 muscle but not glandular structures, and (4) spread by continuity, not by lymphatics [8].

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 [9-11]. Case reports also have implicated herpetic gingivostomatitis [12], tongue piercing [13], and lymphatic vascular malformation superinfections [14] as causes. Another review article revealed that about 25% of Ludwig angina cases in the pediatric population demonstrated no etiologic factors [15]. In children, Ludwig's angina develops concomitantly with systemic diseases, such as immune deficiency [16] and diabetes mellitus [5].

Bacterial culture isolates from surgical drainage of Ludwig's abscesses usually have both aerobic (e.g., β-hemolytic streptococci and staphylococci) and anaerobic species [9]. Gram-negative bacteria, such as *Neisseria catarrhalis*, Escherichia coli, *Pseudomonas aeruginosa*, and influenza, have also been reported in such isolates [17]. A foul breath odor usually indicates the presence of an anaerobe [18]. Blood cultures of Ludwig angina patients are usually negative [17].

Early recognition of the disease is very important. Painful swelling of the neck, dysphagia, dyspnea, fever, and malaise is the most common complaints. Neck swelling and a protruding or elevated tongue are seen in most cases [19]. 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.

Management involves securing the airway, antibiotic therapy, surgical drainage in selected cases, nutrition, and hydration. Airway management is difficult. If intubation is unsuccessful, cricothyrotomy and tracheostomy under local anesthesia are occasionally performed in those with advanced stages of the disease [20]. Elective awake tracheostomy is a safer and more logical method of airway management in patients with a fully developed Ludwig's angina [21]. Although these recommendations of management are taken from cases of Ludwig's angina in adults, the same may have to be followed in neonates as well.

Intravenous penicillin with metronidazole or clindamycin has been recommended as an early empirical therapy. Combination drugs such as ticarcillin/clavulanate, piperacillin/tazobactam, ampicillin/sulbactam, or amoxicillin/clavulanate are also considered highly effective [22]. Intravenous dexamethasone, given every 6 h for 48 h, has been beneficial in decreasing edema and cellulitis, which helps maintain airway integrity and improves the penetration of antibiotics in the area [17].

CONCLUSION

Ludwig's angina should be considered in a baby with recent onset stridor. Early aggressive treatment with empirical antibiotics along

with timely steroids and surgical drainage in airway obstruction is the cornerstone in management. In such cases, tracheostomy should be anticipated and performed early.

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Funding: None; Conflicts of Interest: None Stated.

How to cite this article: Wilson KN, Rahul A, Krishnan S, Sujatha R, Saraswathyamma SK. Neonatal Ludwig's angina with a whirlwind progression and catastrophic outcome: A case report. Indian J Child Health. 2022; 9(4):58-60.