

An Adolescent with Sore Throat and Odynophagia: A Case Report of Ludwig's Angina

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Abstract

We report a case of Ludwig's angina in an immunosuppressed adolescent presenting with sore throat and odynophagia. Ludwig's angina is a rare but potentially life-threatening infection of the submandibular space. We present an overview of the presentation, risk factors, microbiology, diagnosis, and management of this condition in addition to our case report. We present this case to highlight the importance of considering this condition, especially in an immunosuppressed pediatric patient.

Case Presentation

A 16-year-old female presented to a community pediatric clinic with a three-to-four-day history of sore throat, low grade fever, and worsening odynophagia beginning after a water park trip where the patient came in contact with a friend with upper respiratory tract infection symptoms. Following the trip, she reported feeling unwell and having a sore throat, in addition to being mildly febrile. A throat swab at a walk-in clinic two days prior to presentation was negative for Group A *Streptococcus*.

The patient's history was significant for Crohn's disease, polyarticular juvenile idiopathic arthritis, and a distant tonsillectomy and adenoidectomy for recurrent Group A streptococcal infections. She had not had any recent dental manipulations or infections, and her last dental cleaning occurred approximately two months prior to presentation. Her medications included methotrexate; approximately one month prior to presentation, she had also started infliximab and discontinued sulfasalazine. Her immunizations were up to date.

Upon presentation to her pediatrician, the patient endorsed odynophagia and sore throat, which was impairing her ability to eat both solids and liquids. On examination, the

patient appeared unwell: she was drooling with her tongue protruding and her voice was hoarse, though she was now afebrile. On auscultation, the chest was clear, including absence of stridor. She had submandibular tenderness but no palpable lymph nodes. The patient was sent to the local community hospital for assessment by the pediatrician on call.

Diagnosis and Management

At the hospital, the patient was further noted to have trismus, mild posterior oropharyngeal erythema, and submandibular tenderness with visible edema; there was no airway compromise. Neck x-ray and computed tomography (CT) were unremarkable except for mild swelling of the tonsils, and her white blood cell count (WBC) was $12.9 \times 10^9/L$. The patient was then seen by an experienced otolaryngologist, who diagnosed her clinically with Ludwig's angina based on presentation and a bedside flexible nasopharyngoscopy.

Pending microbiology results, the patient was treated with broad-spectrum antibiotics, with attention to her immunocompromised status and exposure to water and sick contacts. She was administered intravenous piperacillin-tazobactam, vancomycin, and azithromycin. Furthermore, her methotrexate was held, she was admitted to the intensive care unit for airway monitoring, and intravenous dexamethasone was administered.

Outcome

Following treatment overnight, the patient reported significant improvement, with greatly reduced edema of the floor of the mouth and only residual throat discomfort. She was transferred to the general pediatric floor. By the following day, her WBC had returned to $6.6 \times 10^9/L$, and she was stepped down to oral azithromycin and amoxicillin/clavulanic acid. She was determined to be well enough to be discharged and was asked to finish her course of antibiotics following discharge. Blood cultures and nasopharyngeal swabs were ultimately negative for all tested bacterial and viral agents. On follow-up in the hospital's pediatric outpatient clinic three days later, the patient had fully recovered. However, she returned to her pediatrician three weeks later, once again complaining of odynophagia and throat pain. This was assessed as a possible return of the Ludwig's angina, and she was successfully treated with a further course of azithromycin and amoxicillin/clavulanic acid.

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Discussion

Ludwig's angina is a severe manifestation of submandibular space infection involving both the sublingual and submylohyoid spaces that comprise the submandibular space. The two compartments communicate, allowing infection to spread between them.¹ The infection can further spread into the parapharyngeal space via the buccopharyngeal gap and, from there, to the retropharyngeal space and superior mediastinum, resulting in the rare complication of mediastinitis.²

Patients with Ludwig's angina usually present with fever, chills, mouth pain, neck stiffness, drooling, and dysphagia. They may have a muffled voice or be unable to speak.² There is also submandibular and submental neck swelling, induration of the floor of the mouth, and swelling of the tongue.^{3,4} Prior case series show that children comprise 24-35% of cases of Ludwig's angina, and the condition has been diagnosed in a neonate as young as 12 days in one case report.^{4,5}

Ludwig's angina is most often caused by spread from dental infections, most frequently of the second, and in adults, the third, mandibular molars.⁶ Odontogenic causes are more common in adults (70-90%), but still make up 50% of the causes in children.⁷ Other sources include peritonsillar abscesses, mandibular fractures, oral lacerations, piercings, and oral malignancies.⁷ Patients can further be predisposed by recent dental procedures, systemic illness, malnutrition, and, as in the case of this patient, impaired immune function.^{3,5,8} Notably, in children, 25% of Ludwig's angina can occur without any precipitating cause.⁴ The most common causative organisms include viridans group streptococci and oral anaerobes, such as peptostreptococci and *Fusobacterium nucleatum*.^{5,8} Gram-negative bacteria, including *Neisseria catarrhalis*, *Escherichia coli*, *Pseudomonas aeruginosa*, and *Haemophilus influenzae*, may also be present.⁵

Ludwig's angina is usually diagnosed based on history suggestive of submandibular infection and clinical presentation, including swelling and erythema of all tissues and compartments of the floor of the mouth, superoposterior displacement of the tongue, firm induration of the submandibular and anterior neck area, trismus, voice change, and dyspnea.⁹ Imaging may support the diagnosis, in which case CT is usually the preferred modality¹⁰ to search for presence of abscesses.⁹ Blood culture may be indicated to check for bacteremia, though 65-83% of cultures may not show any growth.⁴

Ludwig's angina is particularly noted for the risk of airway obstruction and subsequent asphyxiation that may result from superoposterior displacement of the tongue and progressive swelling. Airway obstruction may be imminent particularly if the patient presents with stridor, cyanosis, or an inability to swallow.⁴ Although mortality was once greater than 50%, modern antibiotic therapy and improved imaging have reduced this to 0-8% in the general population, and 10-17% in the pediatric population.^{7,10}

Treatment includes airway management and empiric broad-spectrum antibiotics. Currently, no clinical trials have been performed on antibiotic regimens for Ludwig's angina, so antibiotic choice should be informed by the expected or known microbiology. For immunocompetent patients, antibiotics should cover beta-lactamase-producing aerobes and

anaerobes. Recommended antibiotics for initial treatment of Ludwig's angina prior to culture results include penicillin G with metronidazole, or clindamycin, which is the drug of choice in those with penicillin allergies. If methicillin-resistant *Staphylococcus aureus* (MRSA) is a concern, the patient should also be treated with vancomycin. Antibiotic treatment should continue for two to three weeks until there is clinical improvement and resolution of fever and leukocytosis.¹⁰ In patients found not to be at risk of imminent airway compromise, management may initially begin with close observation and antibiotics. In some cases, intravenous steroids have circumvented the need for invasive airway management.⁷ With progression of swelling, however, control of the airway should be gained immediately. Fibre-optic nasotracheal intubation is the recommended approach, but tracheostomy and cricothyrotomy are also options if this is impossible or unsuccessful. It has been recommended that blind nasal intubation should be avoided due to low success rate and risk of trauma and laryngospasm with repeated attempts. It is also recommended to avoid the use of direct laryngoscopy; distorted airway anatomy and tissue immobility lead to difficult access,¹¹ and mucosal friability and proneness to edema can make further attempts to gain airway control more difficult.¹² Furthermore, the requirement for general anesthesia to perform direct laryngoscopy can precipitate complete airway closure, making mask ventilation and tracheal intubation impossible.¹¹

In immunocompromised patients and particularly in children, gram-negative species may also be present. As such, empiric antibiotic treatment should also include coverage of gram-negative bacteria in addition to the coverage recommended for immunocompetent patients. A common regimen includes a cephalosporin with action against *Pseudomonas*, such as cefepime, combined with an agent targeting oral anaerobes, such as metronidazole. For more advanced infections, a carbapenem or piperacillin-tazobactam may be used. As with immunocompetent hosts, vancomycin should be included in the regimen if there is concern about MRSA.⁹

Conclusion

Ludwig's angina is a potentially life-threatening condition that can usually be resolved with early identification and treatment. Given that children make up a large portion of cases of Ludwig's angina, and that pediatric cases may present without any identifiable precipitating factor, it is especially important to keep Ludwig's angina on the differential diagnosis in clinical presentations such as these, especially in those who are immunosuppressed or immunocompromised.

Informed Consent

The authors verify that informed consent was obtained from the patient's caregiver, in addition to assent from the patient herself.

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