

Case Report

Intraoperative Finding of Potential Ludwig's Angina Resulting from the Spread of Middle Ear Cholesteatoma Following Head Trauma: A Case Report

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Cite this article as: Song X, Fei J, Li L. Intraoperative finding of potential Ludwig's angina resulting from the spread of middle ear cholesteatoma following head trauma: A case report. *J Int Adv Otol.* 2025, 21(4), 1804, doi: 10.5152/iao.2025.241804.

Ludwig's angina is a cellulitis involving multiple spaces in the floor of the mouth. It is mainly associated with odontogenic infections and head or facial trauma, which may result in death by asphyxiation due to sudden airway obstruction. By presenting a case of Ludwig's pharyngitis secondary to middle ear cholesteatoma, it was aimed to alert otolaryngologists to the potential risks and management of this disorder. The patient was a 49-year-old woman with a 40-year history of middle ear cholesteatoma. Ten days prior to her admission, she sustained an ipsilateral punch injury to the tempo-lateral region, which was followed by postauricular erythematous swelling, pain, and postauricular subperiosteal abscess formation. Sudden mandibular swelling was found during postoperative extubation from middle ear cholesteatoma surgery. In order to prevent the occurrence of sudden acute asphyxia in the postoperative period, an immediate prophylactic tracheotomy was carried out to establish an artificial airway. The patient was subsequently treated with antibiotics and had an uneventful recovery. Prompt diagnosis and aggressive therapeutic interventions are essential for successfully managing Ludwig's angina.

KEYWORDS: Bezold's abscess, cholesteatoma of middle ear, head trauma, Ludwig's angina, Mouret abscess, postauricular subperiosteal abscess

INTRODUCTION

Ludwig's angina is a soft-tissue cellulitis involving multiple spaces in the floor of the mouth, which can result in sudden death due to asphyxiation.¹ As opposed to odontogenic infections, traumatic maxillofacial lesions or otitis media rarely lead to diffuse sup-puration of the floor of the mouth causing necrotizing fasciitis. When this occurs, it allows rapid spread of the infection from the sublingual and mandibular spaces to the posterior pharynx and superior mediastinum, resulting in emergent airway obstruction. However, regardless of the etiology of its onset, rapid surgical intervention is required, and early establishment of an artificial airway is life-saving. A case was reported involving a patient with a history of middle ear cholesteatoma and its associated complications, as well as recent trauma to the temporal region. During middle ear cholesteatoma surgery, the patient experienced sudden onset of cellulitis in the floor of the mouth. The relationship among Ludwig's angina, middle ear cholesteatoma, and head trauma was assessed, including the possible routes of infection, the clinical course, and the emergency management. Verbal informed consent was obtained from the patient for anonymized information to be published in this article.

CASE PRESENTATION

A 49-year-old woman presented with painful swelling of the face and behind the ear, and tenderness and erythema in the neck lasting for 10 days after trauma to the right lateral temporal region. The patient had a 40-year history of middle ear cholesteatoma accompanied by ipsilateral peripheral facial paralysis. On examination, the patient's respiration was 18 breaths/min, with a heart rate of 83 beats/min, body temperature of 37.2°C. There was no congestion or swelling in her throat or the floor of her mouth, her mouth opening was unrestricted, and she did not experience difficulty breathing or trouble swallowing. She experienced redness, swelling, localized tenderness and a fluctuating sensation behind the right ear, elevated skin temperature, ulceration of the surface skin with purulent scabs adhering to it (Figure 1), purulent discharge out of the external auditory canal, and visible growth of inflammatory granulomatous neoplastic organisms. The pure tone average threshold was 85 dB. She exhibited right-sided peripheral facial



Figure 1. Image of the patient exhibited right-sided postauricular subperiosteal abscess.

paralysis, classified as grade VI on the House–Brackmann facial nerve grading scale. Additionally, elevated skin temperature and tenderness were observed on the right side of her neck. Blood test results indicated that her elevated leukocyte count was 12.89 (normal, $3.5-9.5$) $\times 10^9/L$ and neutrophil rate was 87.90% (normal, 40%-75%). Computed tomography (CT) scan of the temporal bone revealed enlargement of the right tympanic sinus, soft tissue filling, and complete disruption and resorption of chain of ossicles. Bone destruction

in the external auditory canal was evident, accompanied by swelling of the soft tissues behind and beneath the ear (Figure 2). Magnetic resonance imaging displayed a right middle ear mass with a high mixed signal, filling the lesion and spreading to the external auditory canal and periauricular skin, along with the presence of abscess formation (Figure 3). Magnetic resonance venography (MRV) of the brain revealed a lack of visualization of the right transverse sinus proximal to the heart, as well as the right sigmoid sinus (Figure 4). The patient's primary diagnoses included middle ear cholesteatoma with postauricular subperiosteal abscess, Bezold's abscess, grade VI peripheral facial palsy according to the House–Brackmann facial nerve grading system, and occlusive thrombus formation in the sigmoid sinus. Immediately after admission, a low incision was made to drain the abscess abundance behind the right ear, and the lower edge of the incision was made near the plane of the mandibular angle. This is to relieve the swelling and pain behind the patient's ear while the infection is controlled. She was also given ceftazidime (1 g Q8h IV) for empirical anti-infective therapy. However, in order to be able to completely remove the cholesteatoma from the right ear, the patient was scheduled for middle ear surgery. After 3 days of her admission, a modified radical right middle ear mastoidectomy, type III tympanoplasty, and meatoplasty were carried out under general anesthesia with intubation. Before tracheal intubation on the operating table, anesthesiologist, in collaboration with oral and maxillofacial surgeons, conducted a thorough assessment of the floor of the mouth, submandibular region, neck, vocal folds, and tracheopulmonary conditions to confirm that the infection did not involve the floor of the mouth and submandibular space preoperatively. The surgery lasted for 3 hours and 10 minutes, during which bone destruction of the mastoid tip was observed, and a small hole of about 5×5 mm² size was visible on its deviated medial side leading to the neck. A purulent secretion leakage of approximately 2 mL was found during the cleaning of the lesion, and the inflammatory reaction of the surrounding tissues of the mastoid process was marked and hemorrhagic, which was consistent with the preoperative diagnosis of the Bezold's abscess. Considering the bony destruction of the mastoid tip, the area of rupture is located at the digastric groove, and there is a possibility that the infection spreads towards the digastric groove into the parapharyngeal space, forming a Mouret abscess.² Therefore, the patient's floor of the mouth and neck was examined

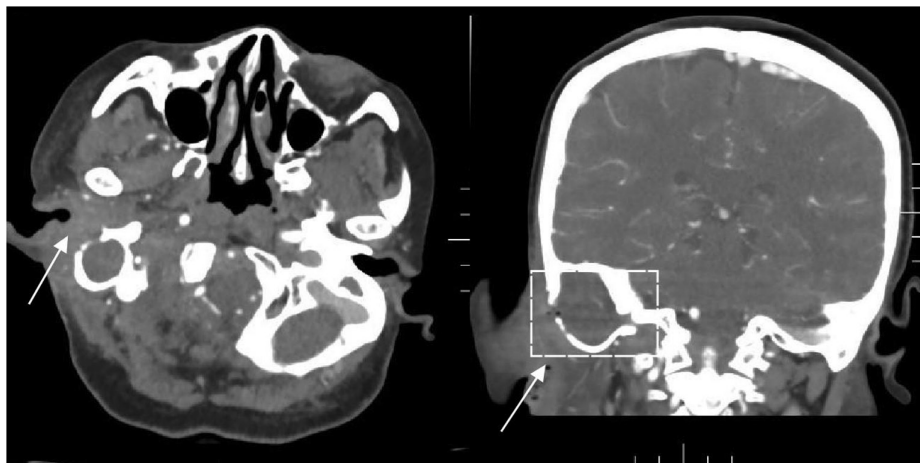


Figure 2. Imaging reveals a lesion filling the right middle ear with destruction of the mastoid bone and communication with the ethmoid sinus. The lesion extends into the external auditory canal and periauricular skin, accompanied by abscess formation.

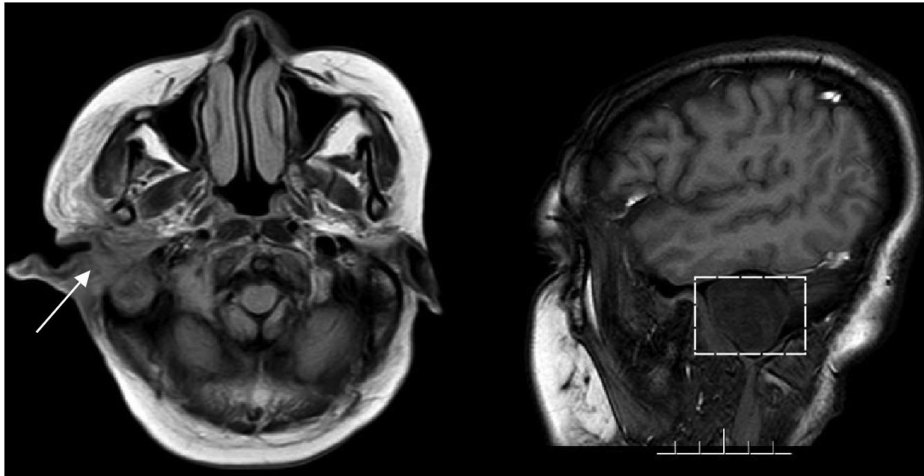


Figure 3. MRI displayed a right middle ear mass with a high mixed signal.

postoperatively, and it was found that the patient's submandibular space and sublingual caruncle were markedly swollen and distended (Figure 5), suggesting the sudden onset of cellulitis of the floor of the mouth (Ludwig's angina), and a prophylactic tracheotomy was immediately performed. Postoperative pathology diagnosed cholesteatoma of the right middle ear. Postoperatively, anti-infection was continued with ceftazidime. A follow-up CT (Figure 6) of the temporal bone on the first postoperative day suggested that there was a significant improvement in the swelling of the soft tissues around the ear, and on the third postoperative day, the leukocyte count was $9.68 \times 10^9/L$, with a neutrophil rate of 63.70%. Physical examination revealed that there was a significant reduction in the swelling and congestion of the submandibular space. The tracheal tube was removed on the tenth postoperative day, and the incision was

closed. The patient was discharged from the hospital on the 16th postoperative day, and followed up according to the postoperative process for middle ear cholesteatoma.

DISCUSSION

Ludwig's angina is a rapidly progressive gangrenous cellulitis that occurs in the floor of the mouth and has an etiology primarily related to odontogenic infections, accounting for approximately 75%-90% of cases.³ Other etiologies include a penetrating injury to the floor of the mouth, osteomyelitis of the jaws, sialadenitis, sialolithiasis of the submandibular glands, and piercings of the tongue.⁴ Infection typically involves the submandibular, sublingual, and submental spaces with little or no abscess formation,^{1,5,6} while may extend into other spaces of the face and neck, and even further inferiorly into the mediastinum. Common clinical manifestations include neck swelling and



Figure 4. MRV revealed a lack of visualization of the right sigmoid sinus.

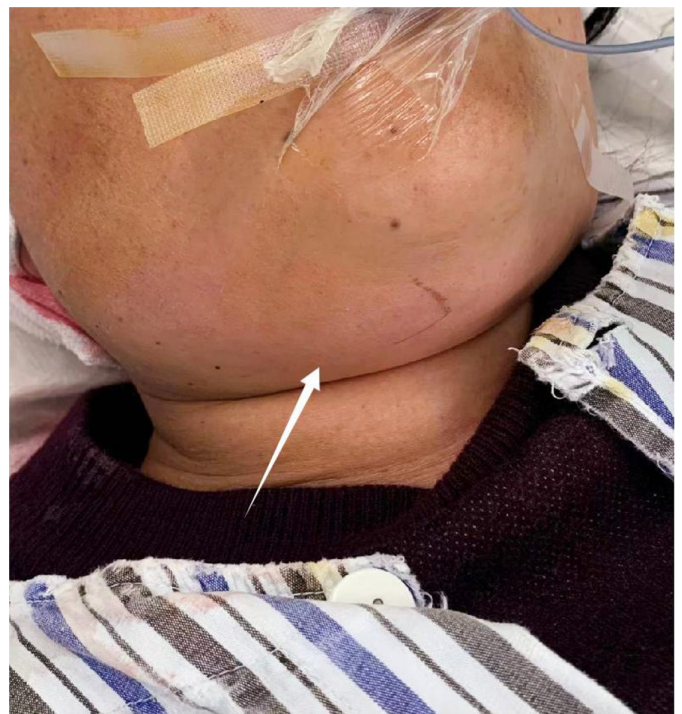


Figure 5. The above images display the patient's intraoperative picture.



Figure 6. A follow-up maxillofacial CT on the second postoperative day revealed persistent submandibular swelling in the patient.

pain, dyspnea, dysphagia, and elevation of the tongue.⁷ Patients may experience asphyxia due to airway obstruction, leading to a mortality rate of up to 10%.⁸ The maxillofacial and cervical regions have a complex structure, featuring numerous bones, muscles, and fascia. The cervical fascia, composed of dense connective tissue, encases important organs, muscles, blood vessels, and nerves in the neck. This arrangement creates various potential spaces between the fascia, including the infratemporal, submandibular, sublingual, submental, supraglottic, and anterior cervical median spaces. These spaces contain a small amount of loose connective tissue, which communicates between the gaps and creates a pathway for easy spread of infection.

In this case, the patient's underlying disease was cholesteatoma of the middle ear with a long history of otorrhea, as well as extensive bone destruction of the temporal bone and intracranial and extracranial complications, such as disruption and resorption of the ossicles and external auditory canals, enlargement of the right tympanic sinus, and sigmoid sinus thrombosis, as detected by preoperative CT and MRV. Extensive destruction of the temporal bone, persistence of infection in the middle ear, and trauma to the lateral temporal region led to the spread of infection through the damaged bone crevices into the pterygomaxillary space in the maxillofacial region. Although only 2 mL of purulent secretion was drained from a small hole in the medial aspect of the mastoid tip intraoperatively, the patient's preoperative signs of elevated skin temperature and tenderness on the right side of the neck were indicative of the formation of submandibular abscess, known as Bezold's abscess, in which the infection had encroached upon the deep section of the sternocleidomastoid muscle by the bony destruction of the mastoid tip.⁹ The submandibular space extends upward through the posterior border of the mylohyoid muscle to the sublingual space and connects anteriorly to the submental spaces. Furthermore, middle ear surgeries, such as mastoidectomy and cholesteatoma removal, contributed to the infection's further spread to the floor of the mouth, causing swelling of the patient's neck and floor of the mouth. Another noteworthy observation in this case was the very rapid onset of Ludwig's angina,

which occurred 3 hours postoperatively, despite thorough preoperative assessment and preparation by the surgical team.

In this case of sudden intraoperative finding of Ludwig's angina, imaging examinations could not be performed to further clarify the diagnosis due to intraoperative conditions. Therefore, close postoperative clinical observation of the patient and timely symptomatic management must be performed. It has been reported that airway obstruction in Ludwig's angina isthmus can occur within 30-45 minutes of the first presentation.¹⁰ Moreover, no instances of middle ear cholesteatoma secondary to Ludwig's angina have ever been reported. Additionally, most otolaryngologists lack sufficient knowledge about the emergence of the disease. To avoid delays in disease management due to insufficient knowledge, management, and attention to the airway, vigilance should be maintained throughout the perioperative period. Tracheotomy is the gold standard for airway management in cases of deep neck infections and is an absolute indication in the treatment of Ludwig's angina. In the case of Ludwig's angina, thorough removal of the primary site of infection while controlling the airway is also important in controlling the spread of the infection.

CONCLUSION

Airway management in Ludwig's angina should be maintained continuously throughout the perioperative period. For otolaryngologists, it is crucial to increase awareness of this condition to ensure timely diagnosis and treatment while also fostering close collaboration with oral and maxillofacial surgeons and anesthesiologists.

Data Availability Statement: All data in the article is real and available.

Ethics Committee Approval: This study was approved by the Ethics Committee of the Affiliated Hospital of Southwest Medical University (approval no.: KY2023337; date: November 1, 2023).

Informed Consent: Verbal informed consent was obtained from the patient who agreed to take part in the study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – X.C.S., F.J., L.J.L.; Design – L.J.L.; Supervision – X.C.S., F.J., L.J.L.; Resources – X.C.S., F.J., L.J.L.; Materials – X.C.S., F.J.; Data Collection and/or Processing – X.C.S., F.J.; Analysis and/or Interpretation – L.J.L.; Literature Search – X.C.S.; Writing – X.C.S., F.J., L.J.L.; Critical Review – F.J., L.J.L.

Declaration of Interests: The authors declare that they have no conflicts of interest. Funding: The authors declared that this study received no financial support.

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