

Case Report

A Case Report of Ludwig's Angina: Extensive Spread from Tooth to Breast

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Abstract

Ludwig's angina is a rapidly spreading and often fatal progressive cellulitis of soft tissues of the neck and floor of the mouth. It was first described in 1836, and before the widespread use of antibiotics, more than 50% of the cases were fatal. In most incidences, it is odontogenic (85%) in origin, while peritonsillar abscess, mandibular fractures, sialadenitis, oral ulcers, and mandibular osteomyelitis are other familiar sources. Most documented cases occur in adult males 20 to 60 years of age. We report a case of Ludwig's angina with necrotizing fasciitis spreading throughout the right breast, presented as an abscess.

Keywords: Ludwig's angina, Necrotizing fasciitis, Breast Abscess

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Introduction

Ludwig's angina is a rapidly spreading and often fatal progressive cellulitis of soft tissues of the neck and floor of the mouth. It was first described in 1836, and before the widespread use of antibiotics, more than 50% of the cases were fatal.^{1,2} In most incidences, it is odontogenic (85%) in origin, while peritonsillar abscess, mandibular fractures, sialadenitis, oral ulcers, and mandibular osteomyelitis are other familiar sources.^{3,4} Most documented cases occur in adult males 20 to 60 years of age.⁴⁻¹¹ While the Spread of these infections is reported to traverse through sublingual, pharyngeal maxillary, and even retropharyngeal spaces, breast abscesses are commonly breast parenchyma in origin from lactation, skin, and soft tissue infections or rarely breast cancers like inflammatory or advanced cases. Here, we present a 58-year-old female presenting with breast abscess eventually, which was discovered as a variation of Ludwig's angina. A systematic and multidisciplinary approach eventually managed the case.

Case Presentation

A 58-year-old diabetic, hypertensive female hailing from Barishal visited an odontologist who presented with a 5-days history of molar teeth agitation, jaw pain, trismus, and submandibular gland swelling. Oral antibiotics were prescribed for 14 days. One week later, gradually, she developed redness & tender swelling bilaterally at the neck. Eventually, she developed a pus point in the upper part of the right anterior triangle with a raised temperature (102°F) and deteriorating general condition. She went to a private dental medical from where she was further referred to the ENT department of a tertiary government medical college hospital in Dhaka. There, she was diagnosed as a case of Ludwig's Angina. After admission, following some investigations, incision and drainage at the neck abscess was done. However, the under-drained abscess was eventually extended to the right anterior chest wall and right breast. A second surgery, incision, and drainage, was performed in the surgery department. With a progressively deteriorating general condition as

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well as wound condition the patient was brought to Z.H. Sikder Women's Medical College & Hospital primarily presented with a large breast wound with extensive necrotizing fasciitis [Fig: 1], electrolytes imbalance, hypoalbuminemia and anemia (Marked leukocytosis and neutrophilia (89%) predominance, HB% 9.8 with RBS 33.7 mmol/L and creatinine 1.42 mg/dl. Chest X-ray showed mild left pleural effusion; Wound swab revealed growth of Klebsiella and Pseudomonas species). The patient was pale,

emaciated, and drowsy. There was a small wound with healing scar at the submental region, a large irregular pale non-healing ulcer at the right side of the neck extending to upper chest (8x16 cm) and another wound of similar characteristics at the right breast with several pockets. There were indurations and concealed abscesses over the chest wall. On examination of the oral cavity, there was a residual abscess at the loosened lower 7th molar tooth (right side).



Figure 1: Under drained abscess extended to neck and anterior chest wall followed by Ludwig's Angina



Figure 2: First postoperative view showing wound excision with partial mastectomy done.



Figure 3 : Second postoperative view showing secondary closure and STSG with drain tube



Figure 4: Patient is under regular follow-up with a healing wound

A Medical Board was formed, constituting the Department of ENT, Dental, Medicine, and General Surgery along with Plastic Surgery decided-through wound excision following optimization of the patient's general condition. Total parenteral nutrition, blood transfusion, correction of albumin, and combinations of injectable broad-spectrum antibiotics and regular cleansing dressing in the surgery ICU ward. One week after admission, the patient underwent wound excision surgery under general anesthesia, where a partial mastectomy was done [Fig: 2], and the patient was shifted to the general ward. On the 8th POD of wound excision, the tooth extraction of the seventh molar tooth was performed to remove the potential source of recurrent infection. On the 17th POD, the chest wound became healthy, and the patient's general condition improved. After that, the wound was closed with secondary closure and split thickness skin graft, keeping the drain in situ. The Drains And stitches (and pins) were removed successively, assessing the wound condition. The patient was discharged on the 14th POD of her third surgery under our care [Fig: 3,4].

Discussion

Ludwig's angina is a potentially life-threatening cellulitis of the floor of the mouth and was first described by the German army physician Wilhelm Frederick von Ludwig in 1836.¹⁶ Although the incidence of the disease is relatively rare, approximately 0.1 to 0.5% of all odontogenic infections comprise 10-15% of all deep neck infections. It is usually associated with poor dental hygiene, pre-existing dental issues, and immunocompromised states like AIDS and uncontrolled DM.⁸⁻¹² Recent literature claims there is a notable increase in the incidence of Ludwig's angina after the COVID-19 pandemic. Moreover, the rate of spreading infections like necrotizing fasciitis has also increased substantially.¹⁵ In our case, although the lady had diabetes, her previous medical comorbidity or H/O Covid-19 positive could not be ruled out. The patient was seriously ill with systemic and electrolyte derangement associated with a severe chest wall infection involving the right breast. Even though the breast gland is separate from the chest's pre-pectoral fascia, it was partially involved and necrosed. Therefore, It may be assumed that deep neck infection might have spread through superficial skin and soft tissue rather than retropharyngeal spaces. The patient had

significant pleural effusion, so co-existing retro-sternal extension also might have taken place.^{10,11}

Most cases are polymicrobial, involving a combination of Gram-positive, Gram-negative, and anaerobic microorganisms. Streptococcus viridans and Staphylococcus aureus are the most common organisms isolated, as these are frequently associated with oral and skin flora. Nevertheless, anaerobic organisms can also grow in the oral cavity quickly.[10]. In the case of our patient, there was profuse growth of Klebsiella and Pseudomonas species resistant to most antibiotics except amikacin, colistin, Piperacillin, and tazobactam combination. The scenario might be a result of nosocomial infection and poor immunity. However, a multi-disciplinary approach and meticulous aseptic surgical dressing helped the patient recover quickly.

Conclusion

Our case is unusual in its extension and is an example of the rare presentation of Ludwig's angina. Ludwig's angina is a potentially life-threatening condition that can progress rapidly. The patient was presented to us with a late complication, and a Mastectomy was performed to save her from spreading necrotizing fasciitis. However, a multidisciplinary team approach made her disease recovery journey easy.

References

1. Rare Angina: A Case Report of Ludwig's Angina. Cureus. 2022 Jun; 14(6): e25873. Published online 2022 Jun 12. doi: 10.7759/cureus.25873, PMID: PMC9275530 PMID: 35836432 Anuradha Sakhuja,1 Dhan B Shrestha,1 Barun B Aryal,2 Wasey Ali Yadullahi Mir,1 and Larissa Verdal
2. Sonar PR, Panchbhai A, Lande AN. Potentially Fatal Ludwig's Angina: A Case Report. Cureus. 2023 Nov 16;15(11):e48885. doi: 10.7759/cureus.48885. PMID: 38106765; PMID: PMC10724867.
3. Romero J, Elkattawy S, Romero A, Latif A, Al-Fiky E, Al-Nasser A, Noori MA, Al-Alwani K. Ludwig's Angina. Eur J Case Rep Intern Med. 2022 Jun 1;9(6):003321. doi: 10.12890/2022_003321. PMID: 35821904; PMID: PMC9267709.

4. Bridwell R, Gottlieb M, Koyfman A, Long B. Diagnosis and management of Ludwig's angina: An evidence-based review. *Am J Emerg Med.* 2021 Mar;41:1-5. doi: 10.1016/j.ajem.2020.12.030. Epub 2020 Dec 23. PMID: 33383265.
5. Chaabouni H , Bechraoui R , Kriaa M , Zainin R , Besbes G, Ludwig's Angina, *LA TUNISIE MEDICALE-2023*; Vol 101 (08-09): 718-720
6. Kawasaki M, Yoshida H, Araki M. Ludwig's Angina. *Intern Med.* 2021 Aug 15;60(16):2707. doi: 10.2169/internalmedicine.5477-20. Epub 2021 Feb 15. PMID: 33583883; PMCID: PMC8429285.
7. Pak S, Cha D, Meyer C, Dee C, Fershko A. Ludwig's Angina. *Cureus.* 2017 Aug 21;9(8):e1588. doi: 10.7759/cureus.1588. PMID: PMC5650252.
8. Saifeldeen K, Evans R. Ludwig's angina. *Emerg Med J.* 2004 Mar;21(2):242-3. doi: 10.1136/emj.2003.012336. PMID: 14988363; PMCID: PMC1726306.
9. Osaghae IP, Adebola AR, Amole IO, Olaitan AA, Salami YA, Kuye O, et al. The disease of the poor and health inequality; Vol 20 (4) 2022 (Aug): Pages e129-e133; <https://doi.org/10.1016/j.surge.2021.05.007>
10. Balakrishnan Thenmozhi Priya. Balakrishnan Thenmozhi Priya's scientific contributions. Available from: <https://www.researchgate.net/scientific-contributions/Balakrishnan-Thenmozhi-Priya-2128289015> (accessed 28 Mar 2022).
11. Kelly Jj, Hodge Ge, Grossman A. Ludwig's angina. *Can Med Assoc J.* 1957 Dec 15;77(12):1089-93. PMID: 13489603; PMCID: PMC1824297.
12. Kobayashi M, Watanabe K. Ludwig angina. *CMAJ.* 2017 Feb 13;189(6):E246. doi: 10.1503/cmaj.160279. Epub 2016 Dec 12. PMID: 27956390; PMCID: PMC5305406.
13. Vallée M, Gaborit B, Meyer J, Malard O, Boutoille D, Raffi F, Espitalier F, Asseray N. Ludwig's angina: A diagnostic and surgical priority. *Int J Infect Dis.* 2020 Apr;93:160-162. doi: 10.1016/j.ijid.2020.01.028. Epub 2020 Jan 23. PMID: 31981767.
14. Kovalev V. A Severe Case of Ludwig's Angina with a Complicated Clinical Course. *Cureus.* 2020 Apr 16;12(4):e7695. doi: 10.7759/cureus.7695. PMID: 32431974; PMCID: PMC7233510.
15. Canas M, Fonseca R, De Filippis A, Diaz L, Afzal H, Day A, Leonard J, Bochicchio K, Bochicchio GV, Hoofnagle M. Ludwig's Angina: Higher Incidence and Worse Outcomes Associated With the Onset of the Coronavirus Disease 2019 Pandemic. *Surg Infect (Larchmt).* 2023 Nov;24(9):782-787. doi: 10.1089/sur.2023.163. Epub 2023 Nov 10. PMID: 37944093; PMCID: PMC11075171.
16. Shemesh A., Yitzhak A., Ben Itzhak J., Azizi H., Solomonov M: Ludwig angina after first aid treatment: possible etiologies and prevention-case report. *J Endodon.* 2019, 45:79-82.10.1016/j.joen.2018.10.001