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## Case Report

# Ludwig's angina complicating pregnancy: an unusual presentation

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### ABSTRACT

Pregnancy is a state of several physiological changes, which predisposes many to oral health problems. However, the majority of pregnant women do not receive oral health counseling. Pregnant women who receive inadequate dental care are at risk for odontogenic infections, which can have serious consequences. Here we are presenting a case of 32 weeks pregnancy with Ludwig's angina complicated by acute renal failure.

**Keywords:** Ludwig angina, Pregnancy, Odontogenic infection, Acute renal failure, Dental caries

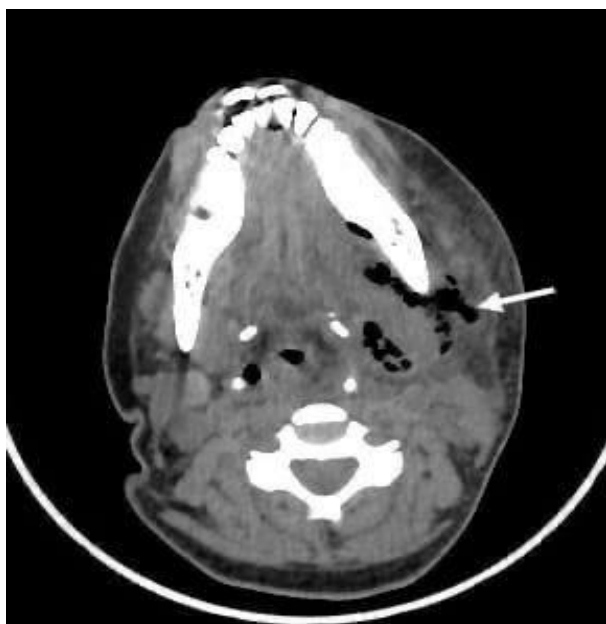
### INTRODUCTION

Ludwig's angina is a potentially fatal widespread cellulitis that affects the floor of the mouth and submandibular gland regions. It spreads quickly and progressively obstructs the airways. The most common cause is an odontogenic infection from one or more severely decaying teeth. However, other possible causes include tongue-base lymphangiomas, sialadenitis, sialadenitis of the submandibular gland, and tongue piercings.<sup>1</sup> Various complications that can arise from untreated infections are upper airway obstruction, septicemia, septic shock descending mediastinitis, jugular vein thrombosis, acute renal failure, disseminated intravascular coagulation, carotid artery pseudo aneurysm and pericardial effusion.<sup>2</sup> Following airway blockage, septicemia has been one of the main causes of death in Ludwig's angina. It spreads rapidly and causes significant morbidity and mortality, particularly when associated with immunocompromised states like pregnancy. Acute kidney injury has not been reported in any of the previous cases which makes it a unique case report. Here we present a case of Ludwig's angina in pregnancy that was complicated by acute renal failure (septic acute tubular necrosis) requiring emergency hemodialysis and caesarean delivery subsequently.

### CASE REPORT

A 28-years old previously healthy second gravida at 32 weeks of gestation, was referred to our centre with deranged renal function, thrombocytopenia and anemia. She had prior one delivery by cesarean section. Upon admission to our hospital, she gave a history of decreased urine output for 3 days. There was a history of rapidly increasing painful swelling on the face and neck, which was not associated with any fever, sore throat, or shortness of breath. She also revealed a history of toothache for the past 2 days, she had never undergone any dental examination or procedure. On admission, the patient was afebrile, pale, arousable, and responsive to commands. There was evidence of asymmetric facial swelling in submandibular gland region. As the pain was in pain and had limited mouth opening, her palate and dentition were not entirely visible. and her trachea was midline. Her heart rate (HR) was 108 beats per minute, blood pressure (BP) was 120/80 mmHg, oxygen saturation was 97% on ambient air and there was no evidence of any respiratory compromise. Bilateral fine basal crepts were audible on chest auscultation and her cardiac examination was unremarkable. Per abdominal examination revealed a soft abdomen with height of the uterus corresponding to the gestational age, uterus was relaxed and fetal heart sound

(FHS) was present and regular. Laboratory investigations showed a total white cell count (WBC) 42,200/mm<sup>3</sup>, platelet count 67,000/mm<sup>3</sup>, haemoglobin (Hb) 6.9 g/dl, and renal function tests were deranged with urea 119 mg/dl, creatinine 9.3 mg/dl. She had hypoglycaemia (RBS=48 mg/dl) and arterial blood gases (ABG) showed a pH of 7.3 and lactate of 3. Her liver function tests, coagulation profile, electrolytes and rest of the antenatal investigations were within normal limits. Obstetric ultrasound revealed evidence of absent amniotic fluid and her non-stress test (NST) was reassuring. Other parameters of her biophysical profile like fetal movements and breathing movements were normal. With this history and clinical examination, we made a clinical impression of Ludwig's Angina with sepsis and acute kidney injury. Other serological tests like HIV, HBsAg, HCV and VDRL were negative, as were blood cultures for bacteria. She had an elevated procalcitonin of 17.5. An ENT consultation was sought and the diagnosis of Ludwig's Angina was confirmed, initiated Parenteral intravenous metronidazole and piperacillin-tazobactam therapy, and recommended against steroids to prevent further maternal immune suppression. As the patient was not medically optimized at the time of presentation, and had severe anaemia, thrombocytopenia with acute renal shutdown, immediate decision for I & D was not taken. NCCT of the head and neck was planned which revealed a hypodense lesion in the Para glossal region (Figure 1).



**Figure 1: Hypodense lesion in the para glossal region (Ludwig's angina).**

Nephrologist consultation was sought for anuria and deranged renal function, and recommended in favor of renal replacement therapy, hemodialysis (HD) and same was commenced immediately. Antenatal testing was conducted periodically during this time and was found to be reassuring.

The patient also received blood components during HD. After 36 hours, the patient was medically optimized, and was reviewed by an otorhinolaryngologist and a plan of incision and drainage (I&D) was made. The time taken to decide for surgical management was compensated by the timely initiation of antibiotics, HD and blood component transfusion. At the same time, fetal heart rate tracing became persistently non-reassuring with repetitive late decelerations, and the decision for cesarean section along with I&D was taken. She underwent repeat lower transverse cesarean section under general anesthesia and delivered a male infant weighing 1490 grams with Apgar score of 8 and 9. Simultaneously I&D of submandibular abscess was done, draining 15 ml of malodourous discharge followed by removal of her endotracheal tube and placement of a tracheostomy tube. On second post-operative day, the tracheostomy tube was changed. The oral maxillary facial service performed an extraction of four of her infected teeth after 4 days of cesarean section. Her antibiotic treatment was continued after an abscess culture revealed gram-negative rods and *E. coli*. The newborn did well in the neonatal intensive care unit. During admission, the patient received 5 cycles of HD. The patient was discharged on postoperative day 17 with improving trends in renal function and tracheostomy tube in situ. RFTs at discharge were urea-49 mg/dl and creatinine of 3.6 mg/dl. Follow up visit was scheduled for tracheostomy decannulation.

## DISCUSSION

Ludwig's angina is a life-threatening condition because of asphyxia and septicemia. Infection from the oral cavity spreads contiguously to the pharyngo-maxillary and retropharyngeal spaces. Since the infection and edema are limited by deep cervical fascia, mandible and hyoid, the tongue and floor of the mouth is elevated and displaced posteriorly with compromise in airway leading to airway obstruction and asphyxia.<sup>3,4</sup> Dental caries and abscesses in the oral and maxillofacial region can release various exotoxins, cytolytic enzymes, as well as gram-positive and gram-negative bacteria, exhibiting their harmful effects throughout the physiological system leading to septicemia.<sup>5</sup> Due to various physiological and immunological changes, pregnant woman is predisposed to odontogenic infections and their faster progression can lead to serious life-threatening conditions. The diagnosis of Ludwig's angina is made on the basis of clinical presentation. Higher imaging like Computed tomography or magnetic resonance imaging scans can help in defining the extent of infection. In 1939, Grodinsky developed criteria for the diagnosis of Ludwig's angina.<sup>6</sup> There must be cellulitis, not an abscess, of the submandibular space that never involves only one space and usually is bilateral; produces gangrene with serosanguineous, putrid infiltration but very little frank pus; involves connective tissue, fascia, and muscles but not glandular structures; and is spread by continuity and not by lymphatics. Intrauterine fetal death (IUD), abruptio placentae, uterine hemorrhage, and puerperal sepsis are among the obstetrical problems

that typically result in acute kidney damage (AKI) during pregnancy. Epidemiological characteristics of pregnancy-related AKI with respect to incidence, causes, and outcome differ significantly between developing and developed world.<sup>7-9</sup> Septic abortion is still the commonest cause of pregnancy-related AKI in developing countries. The incidence of AKI is as high as 15% among patients with skin and soft tissue infections.<sup>10</sup> Ludwig's angina patients must be treated with early with aggressive antibiotic medication, any abscess must be cut and drained, and the airway must be generally protected.<sup>11</sup> Pregnant patients become a challenge to treat as two lives are involved. The microbes may reach the amniotic cavity through the placenta and can affect the fetus by causing septicemia and asphyxia, so the management should focus on minimizing fetal risk without compromising the maternal condition. For any treatment to be successful, it is crucial to address the source of infection immediately. However, this was not possible in our patient because of her compromised renal status along with severe anemia and thrombocytopenia. So, the first step in the management of our patient was optimizing renal status, starting on I/V antibiotics and transfusion of blood and blood products. At the same time, the fetal well-being was being monitored with NST and Biophysical profile. Termination of pregnancy was decided 2-days after admission due to non-reassuring fetal status. The incision and drainage of abscess was done in the same sitting. The timing of the surgical intervention was vital for the outcome of our patient. There is very scarce literature on acute renal failure as a consequence of Ludwig's angina in a pregnant patient. Our patient presented with acute kidney injury and required urgent renal replacement therapy in addition to the commencement of broad-spectrum antibiotics. In addition to AKI, this patient also had severe anemia and thrombocytopenia. During dialysis we could transfuse blood components, as a consequence well timed surgical intervention was possible on a medically optimized patient. Medical optimization within time frame along with aggressive surgical management led to positive outcome in the management of the presented case in our centre.

## CONCLUSION

In pregnancy, Ludwig's Angina is a rare presentation and focus of infection. It is important to recognize that Ludwig angina can present as acute kidney injury also if associated with overwhelming systemic sepsis in immuno-suppressed patients, like pregnancy. A good interaction of a multi-professional team in the treatment of Ludwig's Angina is essential. Obstetric care should include periodontal evaluation and maintenance of good oral hygiene during

the prenatal period in order to reduce the likelihood that dental infections will advance to life-threatening disorders.

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## REFERENCES

1. Vincent U, Kizito N, Fadekemi O. Ludwig's Angina: an analysis of sixteen cases in a suburban nigerian tertiary facility. *Afr J Oral Health.* 2005;2(1):12-9.
2. Lee JK, Kim HD, Lim SC. Predisposing factors of complicated deep neck infection: An analysis of 158 cases. *Younsei Med J.* 2007;48:55-62.
3. Moreland LW, Correy J, McKenzie R. Ludwig's angina. Report of a case and review of the literature. *Arch Intern Med* 1988;148(2):461-6.
4. Saifelddeen K, Evans R. Ludwig's angina. *Emerg Med.* 2004;21(2):242-3.
5. Kamath AT, Bhagania MK, Balakrishna R, Sevagur GK. Ludwig's Angina in Pregnancy Necessitating Pre Mature Delivery. *J Maxillofac Oral Surg.* 2015;14: 186-9.
6. Patterson HC, Kelly JH, Strome M. Ludwig's angina: an update. *Laryngoscope.* 1982;92:370-8.
7. Jayakumar M, Prabahar MR, Fernando EM, Manorajan R, Venkatraman R, Balaraman V. Epidemiologic trend changes in acute renal failure: A tertiary center experience from South India. *Ren Fail.* 2006;28:405-10.
8. Prakash J, Kumar H, Sinha DK, Kedalaya PG, Pandey LK, Srivastava PK, et al. Acute renal failure in pregnancy in a developing country: Twenty years of experience. *Ren Fail.* 2006;28:309-13.
9. Selcuk NY, Tonbul HZ, San A, Odabas AR. Changes in frequency and etiology of acute renal failure in pregnancy. *Ren Fail.* 1998;20:513-7.
10. Chertow GM, Burdick E, Honour M, Bonventre JV, Bates DW. Acute Kidney Injury, mortality, length of stay, and costs in hospitalized patients. *J Am Soc Nephrol.* 2005;16(11):3365-70.
11. Marple BF. Ludwig angina: a review of current airway management. *Arch Otolaryngol.* 1999;125(5):596-600.

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