CASE REPORT

SUBMANDIBULAR SIALOLITHIASIS WITH SIALADENITIS PRESENTING AS LUDWIGS ANGINA - A CASE REPORT

Aneeta Mary Sabu¹, Sumir Gandhi², Manisha Solanki³, Ashwin Romy Sakharia⁴

¹Post Graduate Resident, ²Professor and Head, ³Associate Professor, Dept of Oral and Maxillofacial Surgery, Christian Dental College, CMC Ludhiana-141008, ⁴Oral and Maxillofacial Surgeon, Victoria hospital, Ministry of Health, Seychelles

ABSTRACT:
Ludwig’s angina is a potentially life-threatening, rapidly expanding, diffuse cellulitis of the submandibular, sublingual and submental space resulting in a state of emergency because of impending airway obstruction. Although 90% of ludwigs angina is caused by odontogenic infections, very rarely submandibular sialadenitis can also cause ludwigs angina. However, with early diagnosis, attention to airway maintenance, aggressive intravenous antibiotic therapy and judicious surgical intervention, the process would resolve without complications. Identifying the source of infection and eliminating it is also an important part of the treatment. In the present case report we have presented a rare case of ludwigs angina caused by submandibular gland sialadenitis with sialolithiasis.

Key words: Ludwigs angina, sialadenitis, sialolithiasis.

Corresponding Author: Dr. Aneeta Mary Sabu, Post Graduate Resident, Dept of Oral and Maxillofacial Surgery, Christian Dental College, CMC Ludhiana-141008, Punjab, India


CASE REPORT
A fifty five year old gentleman presented to the emergency department of a post graduate training institute in north India with chief complaint of swelling over the bilateral lower jaw since three days (figure -1). Six days prior to presentation at our hospital, the patient had constant severe pain with respect to the right lower jaw region, following which the patient had visited a dentist and got the faulty fixed acrylic prosthesis on the right lower jaw removed. On the third day after the onset of pain patient had noticed swelling over the right submandibular region which progressed towards the other side of the jaw. Patient had a history of associated fever, hoarseness of voice, cough and dyspnoea. On detailed examination, it was found that he was diabetic with blood sugar levels not within the normal range. Moreover, he gives history of reduced salivation. He was admitted for a day in a nearby local hospital and was administered intravenous antibiotics. On physical examination patient was toxic in appearance with a temperature of 101.1°F, blood pressure of 150/90 mm of Hg, pulse rate 78 beats /min, respiratory rate of 20 breaths /min and random blood sugar 188 mg/dl . On clinical examination, there was diffuse swelling of the submental and bilateral submandibular region. The swelling was firm with induration present in the right submandibular and submental region and soft in the left...
submandibular region. The swelling was tender on palpation with localised rise in temperature and erythematous overlying skin. Patient had a reduced mouth opening of 2 finger breadth and restricted neck movements. Intra orally the floor of the mouth was found to be raised, uvula was central in position with no edema of the faucial pillars. Right lower third molar was grossly carious and rootstump #47 were present .There was no intraoral pus discharge. CECT neck showed right submandibular space abscess with edematous changes in the fascial and subcutaneous planes. A submandibular sialolith 11x9 cm was located at the junction of the right submandibular gland and duct. A poorly visualised right submandibular gland was indicative of chronic obstructive gland atrophy.

An emergency incision and drainage was planned owing to the impending airway compromise from the progressive ludwigs angina. Patient was started on empirical intravenous antibiotic therapy. Bilateral submandibular sublingual and submental spaces were drained under local anesthesia under cardiac monitoring. Grossly decayed #48, #47 were extracted and all the involved spaces were decompressed and drains were secured in place (figure-2).

One week post incision and drainage patient was planned for submandibular gland excision and sialolith removal owing to continuing pus drainage from the right submandibular region despite symptomatic improvement. The gland and the sialolith were excised via a submandibular incision (figure -3) and it were dissected free from the surrounding fibrous tissue. The atrophied gland with the inflammatory tissue were submitted for histopathological examination (figure -4).

**Figure 1:** Diffuse indurated swelling of submental and bilateral submandibular region

**Figure 2:** Surgical decompression of submental and bilateral submandibular spaces

**Figure 3:** Excision of the submandibular gland via a submandibular incision

**Figure 4:** Excised sialolith and atrophied sumandibular gland along with the surrounding fibrous tissue

One week postoperative patient improved clinically and symptomatically.

**DISCUSSION**

In 1836, Wilhelm Frederick Von Ludwigs described a gangrenous induration of connective tissues of the neck and floor of mouth. The name adopted for this infection is a reminder of its potentially lethal outcome. *Angina* is derived from the latin word *angere* meaning to strangle. Ludwig’s angina is a severe diffuse cellulitis that presents an acute onset and spreads rapidly, affecting the submandibular, sublingual, and submental spaces bilaterally resulting in a state of emergency because of impending airway obstruction. In the preantibiotic era, Ludwig's angina frequently caused asphyxiation and death. But with the advent of antibiotics, mortality due to ludwigs angina has significantly reduced from 50% to 8%-10%.

In 1939, Grodinsky developed criteria for the diagnosis of Ludwig’s angina. 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.

Typically, Ludwig’s angina is characterized by fever, malaise, dyspnoea, dysphagia as well as a brawny hard tender swelling of the floor of the mouth and neck. In most instances, it develops as a complication of an odontogenic infection usually from the second and third
molar as the teeth have root at/below the level of insertion of the mylohyoid muscle and hence abscesses here can spread to submandibular spaces.\textsuperscript{3} Other less commonly reported cases include sialadenitis peritonsillar abscess, open mandibular fractures, infected thyroglossal duct cyst, intravenous injection of drugs into neck, traumatic bronchoscopy, endotracheal intubation, oral laceration, tongue piercings, upper respiratory infection and trauma to floor of mouth.

Bacterial sialadenitis can be a potential rare cause of ludwigs angina. Bacterial infections of the salivary glands typically result from retrograde propagation of bacteria through their ducts from oral cavity. This process is promoted by stasis of salivary flow. Predisposing factors for the ductally ascending infection are dehydration, xerogenic drugs and salivary gland diseases associated with reduced saliva secretion or ductal obstructions. Other possible modes of infection are through transitory bacteremia, especially in the neonatal period, or direct spread from adjacent infectious processes.

The parotid gland is the most common site of acute supplicative salivary infection. Saliva of the parotid gland is primarily serous and therefore provides less protection against ascending bacteria.

On the other hand, mucoid saliva produced by the submandibular and sublingual glands contains many antimicrobial protective elements, including lysozymes and IgA antibodies. Mucins also contain stialic acid, which agglutinates bacteria, preventing its adherence to host tissues. Specific glycoproteins found in mucins bind epithelial cells, competitively inhibiting bacterial attachment to these cells. Submandibular sialadenitis is less frequent and accounts for approximately 10% of all cases of sialadenitis of the major salivary glands.

Majority of submandibular gland infections are related to sialolithiasis of Wharton’s duct. Submandibular secretions are more mucinous, and therefore more viscid; they also are more alkaline, containing a higher percentage of calcium phosphates. Some factors inherent to submandibular gland like longer and larger calibre duct, flow against gravity, slower flow rates and higher alkalinity along with higher mucin and calcium content of saliva contribute to the fact that 85-90% of salivary calculi are located in the submandibular duct.\textsuperscript{5}

Long term obstruction in the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis.\textsuperscript{7} In most cases of acute submandibular sialadenitis removal of duct obstruction and conservative therapy including proper hydration, stimulation of saliva flow, analgesics and local heat application to ease the discomfort are sufficient to resolve the disease. Repeated acute supplicative infections lead over time to permanent damage characterized by sialectasis, ductal ectasia, and progressive acinar destruction combined with a lymphocytic infiltrate. The structure of parenchyma and function of the gland are gradually destroyed, hence leading to decrease in salivary secretion and further promotes recurrences in a vicious circle.

The microbiology of Ludwigs’s angina is polymicrobial and includes many gram positive and negative aerobic/anaerobic organisms, but commonly isolated are streptococcal spp, staphylococcus aureus, prevotella spp and porphyromonas spp.\textsuperscript{8}

Treatment invariably consists of securing the airway where necessary, aggressive broad-spectrum antimicrobial therapy, and surgical decompression of the facial planes with removal of source of infection. In select cases, clinically resembling ludwigs angina where submandibular sialadenitis is the etiology, Lenner DN and Troost T advocate gland excision be included with the definitive incision and drainage procedure.\textsuperscript{9} Even in our experience, performing gland and sialolith excision as a secondary procedure after incision and drainage was a technically difficult surgery due to the extensive fibrosis and inflammatory tissue that had formed due to the infection.

**CONCLUSION**

Careful history taking and detailed examination and investigation in identifying the cause of the infection is very important in the management of ludwigs angina. Chronic long standing sialolithiasis can result in sialadenitis, which even though a rare case can result in ludwigs angina. Recognition of ludwigs angina and aggressive management with maintenance of airway, antibiotic therapy and removal of the foci of infection is prudent in the management of ludwigs angina.

**REFERENCES**