Case Report

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E.coli Associated Extensive Bilateral Maxillary Osteomyelitis: A Rare Case Report

Dentistry Section

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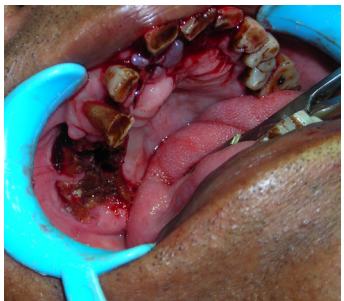
ABSTRACT

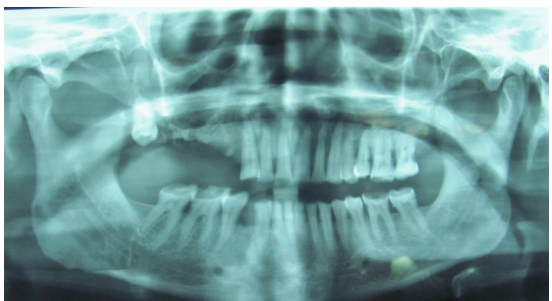
With the advent of broad spectrum antibiotics, chronic osteomyelitis of jaw, especially of maxilla, has become a rare lesion. Osteomyelitis of jaw is associated with a complex microbiota, the most common oral microorganism being, Staphylococcus sp. Reported cases of jaw osteomyelitis caused by enteric bacteria are very few in literature. Hereby, we are reporting a case of *E.coli* associated osteomyelitis in a diabetic individual who had presented with very aggressive bilateral maxillary necrosis. After extensive literature search, to the best of our knowledge, this is the first case of maxillary osteomyelitis associated with *E.coli* which we have come across.

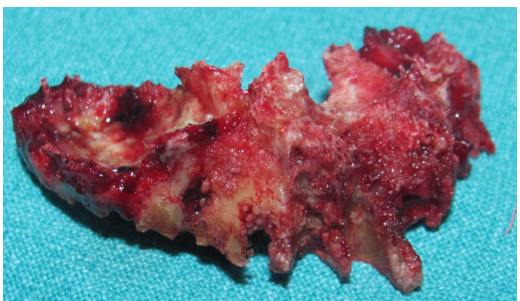
Key words: Osteomyelitis, Maxilla, *E.coli*

CASE REPORT

A 61–year–old male patient came to the Department of Oral and Maxillofacial Surgery, with complaints of painful, non healing extraction site in relation to right upper posterior teeth region, of 3 months duration. Patient's history revealed that there was pain in right maxillary first molar tooth region since the last 4 months. Due to the ignorance of the local physician and lack of optimum medical care, the patient had undergone repeated extractions of regional teeth and biopsy of local soft tissue, which revealed non specific infection with lots of inflammatory cells. The patient was referred to us for further evaluation and treatment. On clinical examination, a large area of exposed bone which extended from right maxillary first premolar to second molar region [Table/Fig-1] was found. Laboratory investigation revealed fasting blood sugar- 350 mg/dl, postprandial blood sugar – 470 mg/dl and elevated neutrophil and total leucocyte counts. Panoramic radiograph demonstrated diffuse radiolucencies extending from the periapical region of right maxillary second molar to left premolar region, with interradicular bone loss [Table/Fig-2]. Although clinically there was unilateral exposure of bone on the right side, the radiological features suggested diffuse radiolucencies in the left maxilla. Therefore, a provisional diagnosis of extensive bilateral maxillary osteomyelitis which was complicated with diabetes was made. A swab from the junction area of exposed bone and mucoperiosteum site was taken and it was sent for culture and antibiotic sensitivity. The patient was referred to an endocrinologist for controlling his blood sugar. The culture report revealed *E.coli* growth and the bacteria was resistant to most of the commonly used antibiotics, except amikacin and gentamycin. After the patient was stable, sequestrectomy under general anaesthesia was planned. Mucoperiosteal flap was reflected and necrotic bone pieces from deeper site were taken and sent for culture sensitivity. A wide area of necrotic bone, starting from right side second molar to left side second premolar region, was found. With slightest pressure from periosteal elevator, the premaxilla and right side posterior part of the maxilla got downfractured and came out [Table/Fig-3]. Almost entire palatal bone from right side and most part on left side (upto first molar) had to be removed because of necrosis. Buccally, root of the zygoma, anterolateral wall of the maxillary sinus (upto infraorbital foramen) on right side, left anterolateral wall of maxillary sinus and pyriform fossa on both sides were also found to be necrosed. So, they were excised, as was evident in post operative panoramic radiograph [Table/Fig-4]. Antral packs were given with ribbon gauge soaked in soframycin and brought to vestibule through a separate stab incision. The patient was given inj. Amikacin 250mg i.m. 12 hourly and inj. clindamycin 600 mg i.v. 8 hourly for seven days. Ryles tube feeding was started from immediate postoperative day and it was kept for seven days, to avoid secondary infections. The second culture report showed *E coli* growth, which was sensitive to amikacin, gentamycin and few third generation cephalosporins. There was a large area of suture line dehiscence on right side, which was noticed on 5th postoperative day, which was repaired under local anaesthesia. The patient was kept on oral cefixime – 200mg twice daily for another three weeks and closely monitored. Postoperative period was uneventful. The excision biopsy report confirmed the lesion to be chronic osteomyelitis of maxillae [Table/Fig-5], which is a rare condition. Culture reports on both occasions showed *E.coli* growth, which very rarely causes osteomyelitis of jaws. At 7 months of follow up, good healing of oral mucosa was noticed [Table/Fig-6].



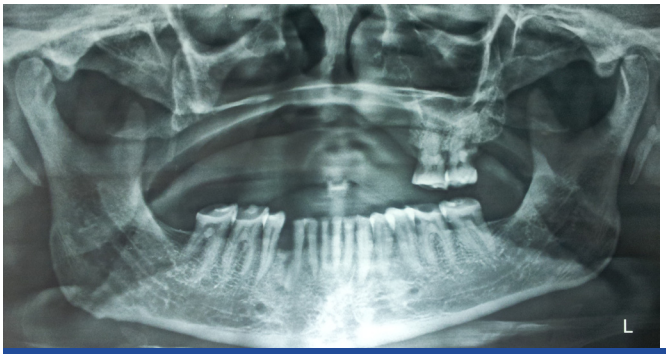


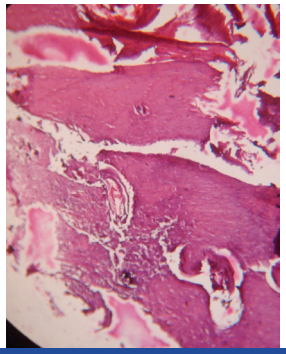


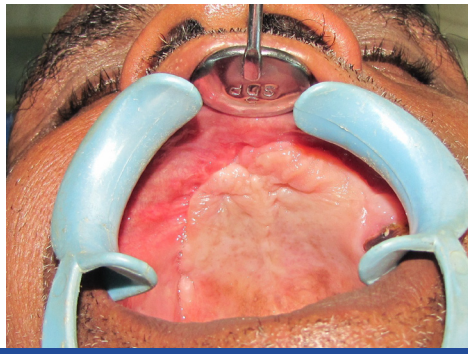
[Table/Fig-1]: Exposed necrotic bone extending from second molar to first premolar region on right side maxilla

[Table/Fig-2]: Preoperative panoramic radiograph demonstrated multiple periapical radiolucencies extending from right maxillary second molar to left premolar region

[Table/Fig-3]: Excised necrotic maxilla, with maxillary sinus floor on both the sides







[Table/Fig-4]: Postoperative panoramic radiograph showing extent of bone loss after extensive sequestrectomy
[Table/Fig-5]: Photomicrograph showing necrotic bone and chronic inflammatory cells (10x)
[Table/Fig-6]: Healing after seven months postoperative follow-up

DISCUSSION

Osteomyelitis of the maxilla is much less frequent than that of the mandible, because of extensive blood supply, thin cortical plates and a relative paucity of medullary tissues in the maxilla [1]. *Staphylococcus aureus* and *S.epidermidis,* together, are responsible for jaw osteomyelitis in around 80 to 90 % of the cases. Other bacteria like haemolytic *streptococci*, *pneumococci*, typhoid and acid fast bacilli, *Escherichia coli* and Actinomyces sp. are considered as causative organisms in 10-20% of the cases [1,2].

The bacteria was named as '*Escherichia coli*' after Escherich, who first described it. *E.coli* is an enteric, gram negative bacterium which is commonly found in human and animal intestines. It is an aerobe and a facultative anaerobe [3]. The enteric bacteria generally do not cause disease, and in the intestine, they may contribute to normal functions and nutrition. The bacteria become pathogenic only when they reach tissues outside of their normal intestinal or other less common normal flora sites [3,4].

There has been reports of *E.coli* associated osteomyelitis in infants or older patients suffering from underlying medical illnesses or in intravenous drug users and is exceedingly rare in the maxillofacial region [5]. Two cases of acute mandibular osteomyelitis evolving towards chronicity associated with enteric bacteria have already been reported and one of which was associated with *E.coli* [6]. In the maxillofacial region, osteomyelitis primarily occurs as a result of contiguous spread of odontogenic infections or as a result of trauma. The adult process is initiated by an inoculation of bacteria into the mandible or maxilla. This can occur with extraction of teeth, root canal therapy, or fractures of the maxilla or mandible [7]. In our case, an extraction history before 3 months, strongly suggests this theory of bacterial inoculation into the extraction socket.

Voided in the faeces, *E coli* remains viable in the environment only for some days. Detection of *E.coli* in drinking water, therefore, is taken as evidence of recent pollution with human or animal faeces. This phenomenon can be exploited by public health microbiologists as an indicator of faecal pollution of water sources, drinking water and food [8]. Three possible causes have been stated by Scolozzi et al., [6], that can lead to the bone's being exposed to enteric bacteria: (1) contamination due to a contiguous focus of infection such as an open fracture, an abscess, or cellulitis or chronic apical periodontitis; (2) haematogenous osteomyelitis, which develops mostly in prepubertal children and in elderly patients and is usually located in the metaphyseal area of long bones; and (3) the possibility of self-injection of soil or faecal material, such as in the case of chronic factitious disease. The 2nd and 3rd reasons mentioned above were ruled out in our case, considering the site and thorough history of the patient. There was abundant clinical evidence in the previously case reported by Scolozzi et al., [6] that the patient was suffering from chronic factitious disease. This includes the probability of the patient contaminating himself with the substances containing organisms, which remains the reasonable explanation of isolation of *E.coli*. In conjunction with this report, we could hypothesise the mechanism of inoculation of enteric bacteria into the oral cavity through contamination of drinking water and food items with animal faeces in this particular case. As the extraction socket didn't heal because of uncontrolled diabetes, the exposed bone would have acted as entry point for the bacteria to migrate into the deeper tissues. The proposed hypothesis is of more concern to public health workers, especially in rural and suburban areas of developing countries like India, where poor sanitation prevails.

Necrosis of maxilla can be caused by fungal infection, trauma, irradiation, Herpes zoster, necrotizing sialometaplasia, midline lethal granuloma, Gaucher's disease, bisphosphonate associated osteonecrosis and bacterial infection [8-11]. In our case, there was no history of trauma and radiotherapy. A smear was taken to rule out fungal component. Herpes zoster usually presents with painful rashes and vesicular eruptions in the distribution of a sensory nerve. This disease usually affects middle-aged and older patients, it is confined to specific dermatomes, and typically, it does not cross the midline [10, 11]. So, Herpes zoster induced maxillary osteomyelitis was ruled out on basis of bilateral involvement of maxilla and absence of vesicular eruptions and rashes. The histopathology of this case was not corroborative with the typical findings of necrotizing sialometaplasia. Clinical presentation and history of this case were not consistent with midline lethal granuloma and Gaucher's disease. Patient had no history of bisphosphonate exposure. Bacteriological cultures from pus sample and necrotic bone had shown *E.coli* growth on both the occasions.

In the absence of acquired resistance, *E.coli* is susceptible to many antimicrobial agents, including ampicillin, cephalosporins, tetracyclines, quinolones, aminoglycosides, trimethoprim and sulphonamides. Many strains, however, have acquired plasmids conferring resistance to one or more of these drugs, and antimicrobial therapy should be guided by laboratory tests of sensitivity [3, 12]. As in this case, the first bacterial culture and sensitivity report showed sensitivity only for amikacin and gentamycin. The empirical antibiotics given by the dentists before the patient came to us were not so helpful to the patient.

The most frequent sites of clinically important infections caused by *E. coli* are the urinary tract, biliary tract, and other sites in the abdominal cavity [4]. It is exceedingly rare in maxillofacial region [5]. An insight to the patients' socio economic status may help in assessing the possibilities of the *E.coli* infection, due to sanitary practices followed by the patient, as we have hypothecated in this case. Despite its rare finding in mandibular and maxillary osteomyelitis, surgeons should always keep *E.coli* in mind when they formulate a differential diagnosis, especially when they deal with aggressive osteomyelitis, as in our case.

CONCLUSION

Patients having known risk of vascular diseases, with risk factors

like diabetes, hypertension, alcohol abuse, long term steroid medication, decompression diseases, sickle cell disease, etc, are prone to osteomyelitis. Due to the varied bacteriology in case of maxillofacial osteomyelitis, no study has been done on pathogenicity of less commonly associated bacteria. Further studies in this field are highly recommended, to elucidate the pathogenicity of bacteria in context to massive bone destruction, even in highly vascular maxillary region, and to evaluate the host and bacterial factors responsible for bone necrosis.

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