Management of Osteomyelitis in Patient with Oral Submucous Fibrosis: A Case Report

Giri KY1, Sarwar Alam2* and Ruby Khan2

1Department of Oral & Maxillofacial Surgery, Institute of Dental Sciences, Bareilly-243006, Uttar Pradesh, India
2Department of Periodontics, Institute of Dental Sciences, Bareilly-243006, Uttar Pradesh, India

Abstract

We report an interesting case of chronic suppurative osteomyelitis of mandible in a patient with oral submucous fibrosis. The coexistence of two diseases in the present case differ in their etiology, clinical behavior and treatment modalities. Initial management of trismus associated with oral submucous fibrosis was done with fibrotomy of submucosal bands and post incisional fibrotomy coverage was done with buccal pad of fat. After achieving adequate mouth opening following fibrotomy, then management of osteomyelitis was carried out with intraoral extraction of offending tooth, debridement, curettage of necrotic bone, followed by extraoral sinus excision and post-operative antibiotic therapy. Following surgery, jaw exercises were initiated promptly after 3 days and continued for 6 months.

Keywords: Oral submucous fibrosis; Osteomyelitis; Trismus

Introduction

Osteomyelitis is an inflammatory condition of bone that involves the medullary cavity and has a tendency to progress along trabecular bone marrow and involve adjacent cortex, periosteum and soft tissues [1]. Oral submucous fibrosis is a collagen disorder common among Indians, Pakistanis, Sri Lankans and also the people of this origin settled elsewhere in the world [2]. Oral submucous fibrosis was first reported by Schwartz in 1952, reported five cases of Indian women from Kenya with Oral submucous fibrosis [3]. Oral submucous fibrosis is a well recognized potentially malignant condition, which is an insidious chronic disease affecting any part of the oral cavity and sometimes pharynx. Although, occasionally preceded by and/or associated with vesicle formation, it is always associated with juxtaepithelial inflammatory reaction followed by fibroelastic changes of the lamina propria, with epithelial atrophy leading to stiffness of the oral mucosa thereby causing trismus and inability to chew food [4]. The initial symptoms experienced by the patient include burning sensation to spicy food; hot food and rigidity of the tongue, palate, and lips are seen in advanced cases. Progress of oral submucous fibrosis into carcinoma has been reported in 3-7.6% cases [5,6]. The main concern in oral submucous fibrosis is the management of trismus and burning sensation of oral mucosa. The treatment modalities include both medical and surgical [7]. The occurrence of chronic suppurative osteomyelitis of mandible and oral submucous fibrosis in one patient is a rare coincidence.

Case Report

A 50 year old male patient reported to the department of oral & maxillofacial surgery with a chief complaint of pain, swelling and pus discharge in relation to right side of the mandible since one month. On extraoral examination, a sinus tract was seen on right side of the cheek 4.5 cm postero inferior to the right angle of the mouth with intermittent pus discharge. There was no paresthesia of the right lower lip and chin. Right sub-mandibular lymph node was palpable, tender and mobile. The patient gave a history of chronic gutkha chewing for the past 10 years. Intraoral examination was difficult due to stiffness and inelasticity of the oral mucosa. Trismus was severe with an interincisal opening of 5 cm (Figure 1a). The buccal mucosa was pale and firmly attached to the underlying tissues. Vertical fibrous bands could be palpated in the premolar area. The patient is unable to blow his cheeks and whistle. Medical history of the patient was non contributory. Vital signs appeared normal on examination. An orthopantomogram (OPG) revealed a remarkable moth eaten appearance and radiopacity periapically in relation to lower right posterior teeth (Figure 1b). Caries
in relation to 16, 45, 46, 47, 48 teeth were present. Teeth missing were 31 and 41. Periodontally compromised lower left posterior teeth and root pieces with 17 and 27 are seen on OPG. On routine blood investigations values are in normal limits except for haemoglobin level. The patient’s Hb level was 9 g/dl. Based on extraoral and intraoral examination, personal history and investigations a diagnosis of chronic suppurative osteomyelitis of the mandible with oral submucous fibrosis was made.

The patient was hospitalized and started with empirical antibiotic therapy. Extraoral sinus was derided daily and sterile dressing placed. The Hb concentration of the patient was raised following infusion of 2 units of packed red blood cells. Pre anaesthetic evaluation was done with all necessary investigations and patient was posted for surgery under general anaesthesia.

GA was administered through fibre optic nasal intubation. Surgical management of osteomyelitis in this patient was difficult due to trismus associated with oral submucous fibrosis.

Initial management of trismus was done with a horizontal incision placed bilaterally along the occlusal plane on the buccal mucosa below the Stenson’s duct orifice. Anteriorly, the incision was limited just lateral to the angle of the mouth and posteriorly the incision was carried to pterygomandibular raphe. The fibrous bands were released using finger dissection. A guarded mouth gag was placed between the occlusal surface of the posterior teeth and mouth was then forced to open upto 3.5 cm (Figure 2). Incisional biopsy was performed bilaterally from the lesion for histopathology. The raw areas on the cheek mucosa following fibrotomy were covered with buccal fat pad bilaterally. The buccal fat pad was approached through the posterosuperior margin of the surgical wound submucosally, that is posterior to the zygomatic buttress. The fat was gently teased out by using small artery forceps interposed to the raw area and was secured to the wound margins with simple interrupted sutures using 3-0 Vicryl.

Later treatment for chronic suppurative osteomyelitis was performed by extraction of 45, 46, 47, 48 teeth with thorough debridement of the necrotic bone and curettage. The teeth were extracted due to the focus of infection and not amenable to endodontic treatment. Finally the sinus tract was tracked and excised extraorally (Figure 3a and b).

Ryle’s tube was kept intraoperatively for feeding and was removed on 7th postoperative day. Post operative i.v. antibiotics were given for 7 days. Post-operative irrigation was done on the operated site twice daily using 15 ml Betadine. Postoperatively by the end of the first week, healing was uneventful (Figure 4a). Jaw exercises were initiated promptly after 3 days following surgery and continued for 6 months. Strict instructions were given to the patient to stop gutkha chewing. Following discharge oral Penicillins for 1 week and vitamin tablets were prescribed for 15 days. Recalls were done twice a month with a 15 day interval upto 6 months. During review at the end of the second month, the scar produced by extraoral sinus excision is unnoticeable (Figure 4b). At the end of the 6th month, the final interincisal distance was maintained at 30 mm (Figure 5).

**Discussion**

Osteomyelitis is an extensive inflammation of the bone. It involves the cancellous portion, bone marrow, cortex and periosteum. Osteomyelitis of the jaws is more common than that of long bones.
mainly due to odontogenic infections. Osteomyelitis is classified into chronic suppurative, chronic focal or diffuse sclerosing osteomyelitis and chronic osteomyelitis. Osteomyelitis lasting for over 6 weeks duration is defined as chronic osteomyelitis [8]. Chronic osteomyelitis of the mandible represents most complicated infection. The etiological factors for jaw osteomyelitis include odontogenic infections, traumatic injuries to jaw bones, irradiation, and chemicals such as Phosphorus, Arsenic, Mercury and specific diseases such as Tuberculosis, Actinomycosis and Syphilis. Osteomyelitis of the mandible has also been reported as a complication of sickle cell anaemia by Girasole and Lyon (1977) [9]. Osteomyelitis is more commonly affected in mandible than in maxilla. Extensive blood supply of the maxilla makes it less prone to osteomyelitis.

Jaw osteomyelitis usually is more commonly due to odontogenic infection or traumatic injuries to jaw bones and is a mixed infection with oral bacteria that involves all layers of bone in which widespread necrosis occurs [10]. The disease is diagnosed on parameters such as patient history, clinical evaluation, radiographs, CT scans and histopathological examination. Diagnosis of the disease can also be made by 3-phase skeletal scintigraphy using 99MTC-MDP as a radiotracer and SPECT-technique [11]. But cost effectiveness and compliance should be considered. Treatment of the osteomyelitis of the jaws includes removal of the cause like extraction of the carious tooth which are a source of infection for the disease and not amenable for endodontic treatment, incision and drainage, curettage, sequestrectomy, saucierization, decortication, resection of the jaw, antibiotic and HBO therapy. Most cases of the osteomyelitis of the jaws responds well to debridement, sequestrectomy and treatment with antibiotics. Publications regarding treatment for osteomyelitis have been inconclusive, but a protocol consisting of surgical debridement supported by antimicrobial therapy given intravenously for 1 week followed by oral penicillin for 1 week has been shown to be successful [12]. In our case, the cause of chronic suppurative osteomyelitis was due to odontogenic infection. The treatment was performed by extraction of carious tooth 45, 46, 47 and 48 with thorough debridement of necrotic bone and curettage. The teeth were extracted due to not amenable to endodontic treatment. Finally sinus tract was tracked and excised extratorially. Postoperatively, i.v. antibiotics were given for 1 week, followed by oral penicillin for 1 week and vitamins for 2 weeks. Treatment has given complete healing of the disease without any adverse long term consequences.

Oral submucous fibrosis is a well recognized potentially malignant condition, which is an insidious chronic disease is affecting any part of the oral cavity and sometimes pharynx and rarely the larynx followed by muscular degeneration and trismus. The main etiological factor for oral submucous fibrosis is chewing areca nut [13]. Arecoline, an alkaloid component of areca nut stimulates fibroblast proliferation and collagen synthesis, rendering them resistant to degradation by collagenase. The other etiological factors that trigger the disease include consumption of cells, nutritional deficiencies and defective iron metabolism [14]. The initial symptom is burning sensation on eating spicy food, hot food or on intake of hot beverages. Oral submucous fibrosis is diagnosed on clinical criteria including mucosal blanching, burning, hardening and the presence of characteristic fibrous bands and is associated with gradual inability to open the mouth [15]. Measurement of interincisal distance on mouth opening gives the severity of disease (Functional Stage), Clinically, it may be classified by the site of involvement and distribution of fibrous bands (Clinical Staging) (Table 1) [16].

Treatment for Oral submucous fibrosis is broadly divided into medical and surgical. The main concern in the treatment of oral submucous fibrosis is management of trismus and burning sensation of oral mucosa. The non- surgical or medical management includes discontinuation of habit and counseling, multiphase injections of local hyaluronidase, local corticosteroids, local placental extract, and supportive care with vitamins. The choice of treatment greatly depends upon functional and clinical staging of the disease. Extensive review of literature on medical management was reported by Kerr et al. [17]. Patients with marked trims are suitable for surgical treatment [18]. Surgical management is carried out by fibrotomy of submucosal bands and with post incisional fibrotomy coverage. Additional procedures like temporals myotomy, coronoidectomy and masseter muscle stripping have also been described to enhance mouth opening. Post incisional fibrotomy coverage can be done with buccal fat pad, nasolabial flaps, dorsal tongue flaps, and split thickness skin grafts. A new technique of utilizing bilateral palatal island flap to cover the post fibrotomy defect was described by Khanna and Andrade [19]. Different surgical procedures described in the literature claim different success rates. The application of buccal fat pad to cover the post incisional fibrotomy defect was first described by Yen [20]. In our case as there is marked trismus with interincisal distance of less than 5 mm, treatment for oral submucous fibrosis was done with bilateral fibrotomy of mucosal bands and post incisional fibrotomy defect was covered with buccal fat pad.

**Conclusion**

Management of osteomyelitis in mandible and oral submucous fibrosis as two different entities in different patients are carried on with the established management protocols. In our case, due to coexistence of osteomyelitis with functionally significant oral submucous fibrosis in the same subject, we experienced difficulty in surgical management of osteomyelitis due to the unyielding nature of the vocal mucosa. This experience of ours also hints about the surgical difficulty posed by the trismus due to oral submucous fibrosis in minor procedures like extractions also and necessitates primary management of trismus in trivial procedures also.

**References**