Case Report

AN INCIDENTAL FINDING OF SQUAMOUS CELL CARCINOMA FROM OSTEOMYELITIS: A DIAGNOSTIC DILEMMA

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Abstract
Osteomyelitis is an inflammation of the bone cortex that begins as a medullary cavity infection, quickly jeopardizing the haversian system and spreading to the bone's periosteum. It could come from trauma, inadequate care for a fracture, radiation therapy following surgery, or a persistent odontogenic infection. A 61-year-old man was diagnosed with squamous cell carcinoma (SCC) arising from chronic osteomylitis in his left mandible. The pathological findings of the excised mandible showed an ulcerated tumor extending to the medullary cavity of the right mandible continuously. A careful pathological evaluation was done for early detection of malignant neoplasms arising from osteomyelitis. Surgery was performed as a treatment modality for extensive osteomyelitis and reconstruction was done. Osteomyelitis is managed by both conservative and surgical management. With the availability of more potent antimicrobials and treatment under aseptic conditions, management of osteomyelitis has been possible.

Keywords: Chronic osteomyelitis, Squamous cell carcinoma, Reconstruction plates, Mandible, Rehabilitation

INTRODUCTION –

Osteomyelitis of the jaws is now defined by the presence of exposed bone in the mouth, which fails to heal after appropriate intervention [1]. Osteomyelitis is an inflammation of bone cortex and marrow that develops in the jaw usually after a chronic infection [2-5]. Marx and Mercuri defined acute osteomyelitis as lasting for less than 4 weeks and chronic osteomyelitis as lasting for more than 4 weeks. Because of poorly vascularized cortical plates, the mandible shows a higher incidence of osteomyelitis. The mandible gets its blood supply from the inferior alveolar neurovascular bundle. Maxilla being less dense shows less incidence as it is highly vascularized and gets its blood supply from numerous feeder vessels. The maxillary posterior region is less affected than the mandibular posterior region. Diminished host defense mechanisms whether local or systemic contribute to its emergence. Diabetes, malignancies, autoimmune diseases, and malnutrition are some of the other causes that may be associated with osteomyelitis [1,6-10].

Predisposing Factors: (1) Any illness that compromises the host's resistance or a delayed host defense mechanism; (2) Local or systemic lack of vascularity in the host bone; and (3) Microorganism virulence.

Any immunocompromising condition, such as diabetes mellitus, leukemia, malnourishment, agranulocytosis, severe anemia, drug abuse, chronic alcoholism, sickle cell disease, typhoid, chemotherapy, radiation, steroid use, hepatitis, HIV, glomerulonephritis, systemic lupus erythematosus, etc., can activate the host defense system. Treatment-related radiation, Paget's disease, osteoporosis, fibrous dysplasia, arsenic, and bismuth (which cause metallic bone necrosis) all affect the vascularity of the host bone. In the maxilla, osteomyelitis can occur due to pansinusitis or facial cellulitis. Because of lysosomal action, certain organisms form thrombi. A protective barrier is formed around the infective foci by the pathogen-born bioactive peptides and the chemoattracted leukocytic purulence which leads to lysosomal and enzymatic destruction of the tissues along with microvascular thrombi formation. The organisms further proliferate in the host medium but remain protected from the host defense mechanism [10,11]. The differential diagnosis of these lesions includes Paget disease, hypercementosis, fibrous dysplasia, early-stage malignant bone tumor, osteogenic sarcoma, and fibrous dysplasia [12,13].

Case report

A 61-year-old male patient reported to the outpatient wing of the Department of Oral and Maxillofacial Surgery, KVG Medical College, Sullia with the chief complaint of pain in the lower left back tooth region for one week. The patient gave a history of pain for one week, which was localized and increased on having food and relief on taking medications. The patient also gave a history of swelling with the left buccal mucosa for 12 days, which was initially smaller in size and has attained the present size. The patient was a known diabetic but not under any medications. The patient gave a history of beedi smoking, approximately 10 beedis/day, from the last 10 years. The patient was moderately built, moderately nourished and well-oriented to time, place and person with stable vitals. On extraoral examination, mild facial asymmetry was noted on the left side of the face, measuring approximately 3x1cm in its greatest dimension, extending supero-inferiorly 2cm below the corner of the mouth to 3cm below the lower border of the mandible and anteroposteriorly 2cm away from the midline to 4cm from the angle of the mandible on the left side. On palpation, all the inspector's findings were confirmed. The swelling was hard in consistency and fixed to the underlying submandibular gland. Paresthesia was noted and tenderness over palpation was elicited over the left lower third of the face. There was no local rise in temperature, no pus discharge or bleeding elicited on palpation. The lymph nodes were non-palpable. The clinical problems reported by the patient were persistent pain, swelling, facial asymmetry, difficulty in mastication.

On intraoral examination, the mouth opening was unrestricted. An irregularly shaped reddish-white lesion, measuring approximately 1cmx1cm was noted on the floor of the mouth. The lesion was firm in consistency and fixed to the floor of the mouth. There was no pus discharge, bleeding or sinus opening noted. There was a completely edentulous maxillary and mandibular arch and tenderness over palpation was elicited. Provisionally, it was diagnosed as Osteomyelitis, and OPG and CBCT were advised.

Material and method

OPG revealed bone resorption extending from the midline to the ramus region on the left side anteroposteriorly and from the alveolar crest to the inferior border of the mandible supero-inferiorly. It also depicted a lytic lesion causing a pathological fracture of the body of the mandible on the left side. CBCT depicted an altered trabecular pattern with a reduction in the bone density on the right mandibular body region, and lytic areas were observed near the crest

(fig 1). There was cortical discontinuity with fragmentation of bone periosteal thickening. There was resorption of the buccal and lingual cortical plates and sequestra formation on the left mandibular body.

FNAC smear showed the presence of moderately cellular and a polymorphous population of lymphoid cells showing predominantly small mature lymphocytes admixed with activated lymphocytes, plasma cells, and macrophages, which favoured the presence of reactive lymphadenitis. To confirm the diagnosis an incisional soft tissue biopsy was done on 33, 34 vestibular region and the specimen was sent for histopathological examination, the report suggested Chronic Osteomyelitis. Following the biopsy, a bilateral parotid gland ultrasound was taken which revealed osteolysis of the left mandibular ramus with bony fragments, periosteal thickening surrounding soft tissue changes, suggestive of left mandibular ramus osteomyelitis, and left submandibular lymphadenopathy (fig 2).

An additional Bone Mineral Densitometry was advised which gave an impression of Osteoporosis in the lumbar spine and the bilateral femora was within the normal limits. The hematological investigation indicated raised ESR (Erythrocyte sedimentation rate), elevated C reactive protein (CRP), moderate leukocytosis (PMNL). Based on investigatory findings and reports osteomyelitis was confirmed as final diagnosis. The patient was planned for mandibular resection of the involved bone under general anesthesia (fig 3-6). Using a modified Risdon incision, the subplatysmal layer was reflected, facial artery and vein were located and ligated, mandible was exposed showing pathological fracture was resected and submandibular lymph nodes were removed. Excised section was send for pathological evaluation (fig 7). Resconstruction plates were used to stabilize the mandible fragments and closed with layer-wise suturing. Drain was placed for 10 days. Gold standard antibiotic protocol was followed, Penicillin (2 million IU Q4H) with Metrogyl (500mg Q6H for 48 hours) for 5days followed by Penicillin 2g with Metrogyl 0.5gm Q8H for 2-4 weeks. Post-op healing was satisfactory. Post 7 days pathology report confirmed Squamous cell carcinoma of left mandible. Ethical approval was taken from the institute (kvg23/0030/3930).

Discussion

The term osteomyelitis was given by Nealston in 1844 which meant infected bone. Before the introduction of penicillin in the 1940s, in the pre-antibiotic era, the classical presentation of jawbone osteomyelitis was an acute onset, usually followed by a later transition to a secondary chronic process (Wassmund 1935; Axhausen 1934) [2,4]. Massive clinical symptoms with widespread bone necroses, neoosteogenesis, large sequester formation, and intra- and extraoral fistula formation were common presentations, sometimes leading to significant facial disfigurement.

Osteomyelitis primarily affects the mandibular region, often in patients with weakened immune systems. Diabetes, malignancies, autoimmune diseases, and malnutrition can contribute to its development. It can arise hematogenously or nonhematogenously, through adjacent infection or direct bone inoculation. Radiographically, sequestra formation, dead cortical bone surrounded by pus, is evident. New bone formation, forming involucrum, starts around two-three weeks post-infection. Diagnosis relies on painful sequestra and suppurative areas in the jawbone unresponsive to conservative therapy [6]. Tissue specimens should be cultured for the presence of microorganisms. Once soft tissue and bone specimens have been obtained, they must be sent to the microbiology laboratory immediately to identify the microorganisms [12]. Histologically, an increased number of osteoblasts, thickened bony trabecula, and fibrous marrow replacement are found.

Pathologic bone remodeling and the presence of chronic inflammatory cells are often cited as indicators of osteomyelitis [13]. Pathogenic organisms usually implicated in chronic mandibular osteomyelitis are normal oral flora, Staphylococcus aureus and aerobic gram-negative bacilli [11]. The histopathology revealed fibrocellular connective tissue stroma with necrotic bony trabeculae with empty lacunae. Dense fibrous tissue with inflammatory cells, including lymphocytes and plasma cells, confirmed chronic osteomyelitis.
Radiographic changes in osteomyelitis appear after 3 weeks, with at least 40–60% bone loss required for detection. Early stages show mixed radiodensities and a "mottled appearance" due to Volkmann's canal growth and marrow space widening. Later, cortical bone involvement occurs. Imaging modalities include radioisotope scanning, PET/CT, and CT. CT offers clearer images with less radiation exposure. Treatment involves medical and surgical approaches, often starting with empirical antibiotics based on gram stain results. Penicillin and Metrogyl are commonly prescribed along with hyperbaric oxygen therapy. Good oral hygiene, including rinses like sodium fluoride gel and chlorhexidine gluconate, is essential.

We also investigated the possibility that some additional instances of SCC may have resulted from osteomyelitis that had spread to or been localised inside bone marrow. However, it may be difficult to locate such cases if extensive pathological findings based on a thorough study of the surgical materials were not made. Furthermore, malignant tumours that form in the deep bone marrow may be detected late, resulting in a bad prognosis. Indeed, our patient had metastases to the clavicle shortly after amputation. As a result, rigorous clinical observation along with pathological testing will be required in order to identify similar situations in the future and enhance the prognosis of each patient.

Conclusion

We saw a case of SCC on the left mandible caused by persistent osteomyelitis following an initial fracture and infection. The SCC extended continuously from the medullary cavity. According to the current and earlier publications, rigorous pathological assessments, such as cytology from bone marrow washing, may be effective for early diagnosis of malignant neoplasms developing from osteomy.

Ethical approval and consent to participate

Not required

Conflict of interest

None

Acknowledgments and disclosure statements

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Patient consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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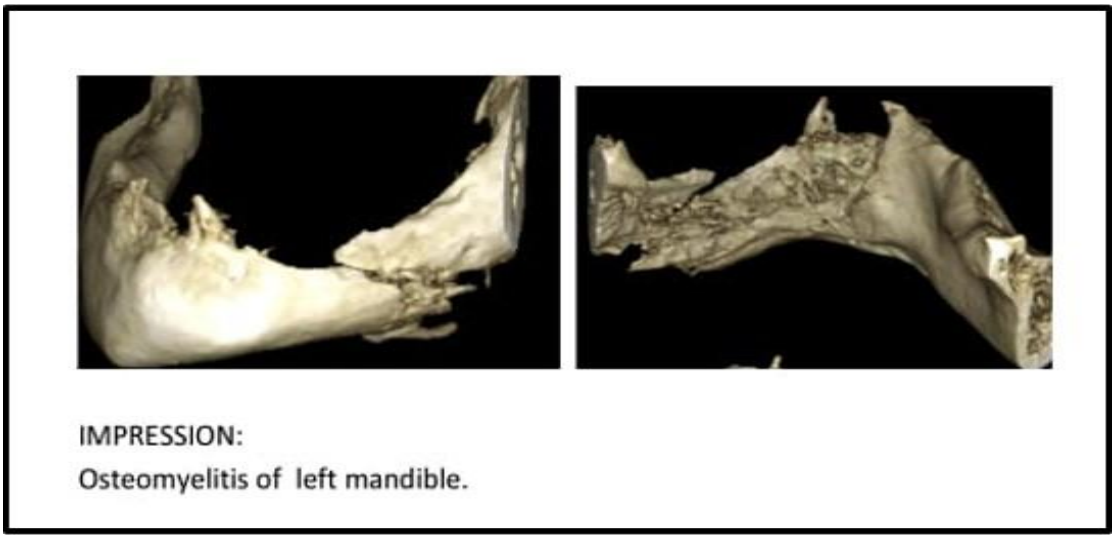


Fig 1

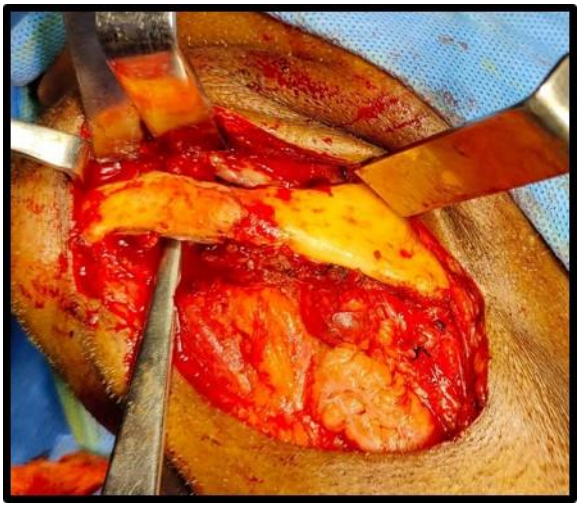
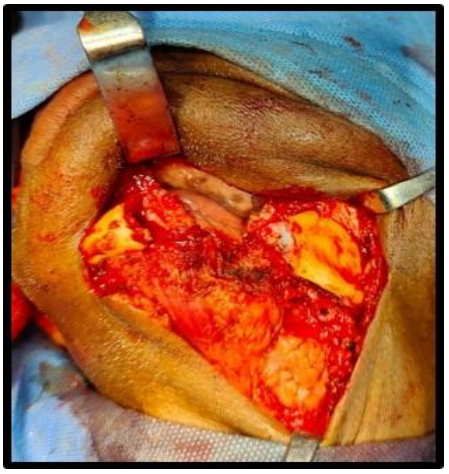


Fig 2



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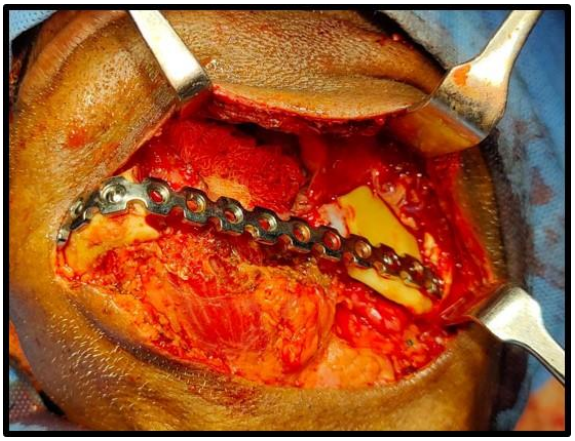

Fig 4

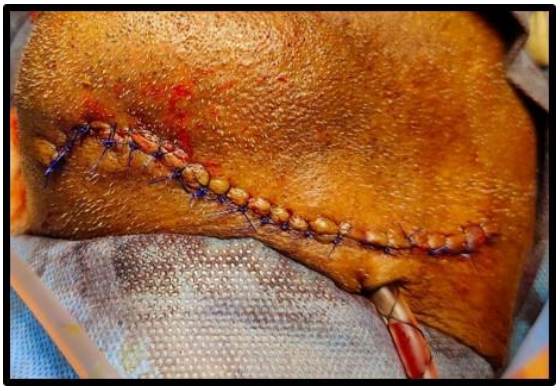

Fig 5

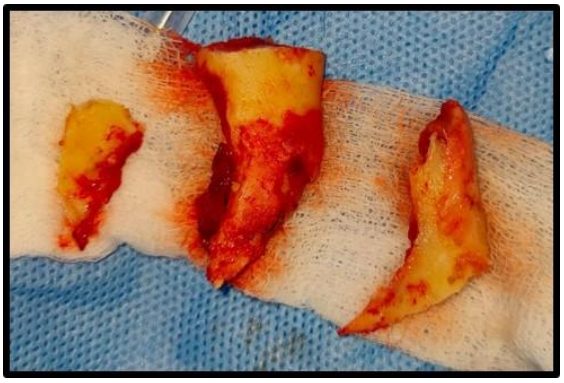

Fig 6

Figure legends

Figure 1: A 61-year-old male patient diagnosed with Osteomylitis, CBCT shows fracture of the left mandible

Figure 2: intra op showing pathological fractured bone

Figure 3: mandibular resection of involved bony lesion

Figure 4: Reconstruction plates used for stabilization of fragments

Figure 5: Layer wise skin closure done

Figure 6: Excised bony fragment after excision