Proliferative Myositis in Mandible: A Case Report

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ABSTRACT

Benign fibrous lesions are very common in the head and neck region and exhibit a variety of clinical and histologic features. Proliferative myositis is one such rare lesion. It is a benign fibroproliferative disease that develops rapidly within 1-3 weeks from onset, eventually resolving spontaneously in approximately six weeks. Cases of proliferative myositis developing in the head and neck are quite rare. Only 100 cases have been reported in the last 45 years. This disease usually affects muscles and soft tissues. It is a benign entity but its clinical presentation can simulate malignancy and was mistaken for sarcomas in the past. A 14-year girl presented to the Department of Oral and Maxillofacial Surgery with a complaint of pain in the anterior mandible in the midline. The panoramic x-ray revealed a radiolucency in the anterior mandible. Excisional biopsy under general anaesthesia was reported as proliferative myositis. This was an unexpected case because not a single case has been reported in the literature involving the bones.

Key Words: *Proliferative myositis, Anterior mandible, Pseudo sarcomatous tumour.*

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INTRODUCTION

Proliferative myositis is a benign reactive lesion that presents as a rapidly growing mass in soft tissues such as muscles and fascia.[1,2] This lesion affects patients ranging from 9 years to 82 years, with the peak incidence in the 50s. There is no gender predisposition.[3] This lesion is rare in the head and neck.[4] In the head and neck region, the most commonly affected sites are the sternocleidomastoid muscle followed by the masseter muscle.[5] Patients with proliferative myositis present with tender, well-defined, localised firm swelling, and trismus.[6] Symptoms depend on the anatomical area of involvement. It is a benign lesion, but because of its aggressiveness, it is often mistaken for malignancy and treated accordingly.[7] This entity can also affect the bone very rarely.

CASE REPORT

A 14-year female patient presented with a few weeks' history of pain in the anterior mandible. There was no history of trauma or any dental procedure in the past. Her medical history was non-significant.

On examination, there was no swelling extra-orally but mild swelling at the anterior mandible intraorally which was tender on palpation. Her mouth opening was adequate. There was no history of lip paraesthesia.

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The cervical lymph nodes were non-palpable. Intraorally, the teeth were vital, caries-free with no periodontal pathology. A panoramic radiograph revealed an irregular radiolucency 2 × 2 cm surrounding the roots of lower central and lateral incisors. The borders of the radiolucency were irregular and there was no root resorption (Figure 1).

Hence, differentials such as giant cell granuloma and odontogenic cyst/tumour were contemplated in this case on the basis of anatomical location and radiographic presentation. An excisional biopsy with curettage was executed and was sent for histopathology. Pathologic analysis revealed a cellular inflammatory infiltrate composed of lymphocytes, plasma cells, histiocytes, macrophages, a few eosinophils, and mast cells. The inflammatory cell infiltrate also included plump fibroblasts with large vesicular nuclei and abundant collagen and myofibro-blasts surrounding individual muscle fibres creating a checkerboard pattern. Evidence of dysplasia was not found. The overall morphological findings were consistent with proliferative myositis. After excisional biopsy and curettage, oral non-steroidal anti-inflammatory drugs (NSAIDs) were prescribed. Her anterior teeth were extracted as they were mobile. She was recalled after a week and then monthly for follow-up. The panoramic radiograph was repeated after a year and there was no sign of lesion recurrence. The radiolucency was replaced by new bone and the patient was symptom-free. Her teeth were replaced. Overall, an outstanding prognosis was maintained (Figure 2).

DISCUSSION

Proliferative myositis is a benign pseudosarcomatous entity that was first defined by Kern in 1960. It represents the intramuscular equivalent of proliferative fasciitis. In the current case, it represents its osseous equivalent which is a very rare condition.[1]

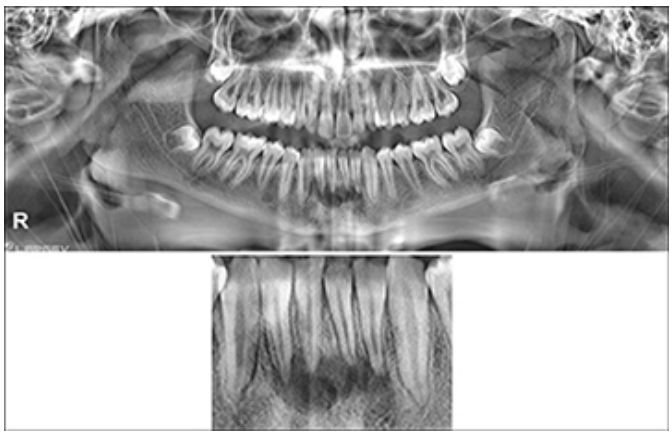

Figure 1: Panoramic radiograph revealing an irregular radiolucency (2 × 2 cm) surrounding the roots of lower central and lateral incisors.

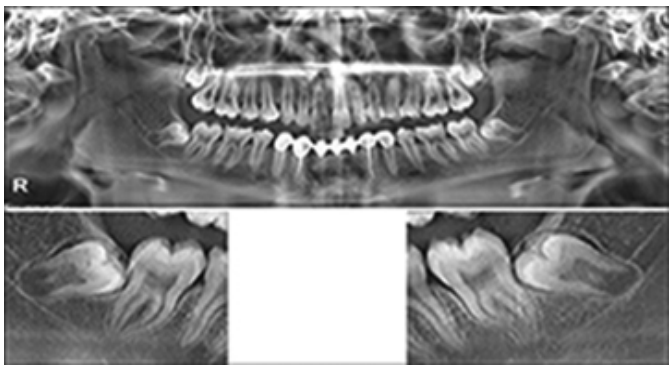

Figure 2: Postoperative panoramic radiograph of the same patient after a year.

Most cases of proliferative myositis reported in the literature affected muscles of the trunk and extremities. It is quite rare in head and neck region, with only around 20 cases documented in the literature involving head and neck musculature or fascia.[2] What makes this case distinctive is the involvement of bone. The lesion involved the anterior mandible and there is not a single case in the literature of proliferative myositis within bone anywhere in the body. However, a very few cases have been reported involving mylohyoid, trapezius, and tongue muscles.[2,5] The aetiology of this lesion is unknown. The prevailing view suggests that proliferative myositis is a reactive lesion to mechanical stimuli in the form of injury or trauma. About 30% of cases of proliferative myositis have a positive history of trauma in the past.[4] Patients present with tender well-defined localised firm swelling. The symptoms also depend on the area involved, as in the case of masseter involvement, there is trismus.[6]

As it involves musculature most of the time, magnetic resonance imaging (MRI) is an investigation of choice.[8] Histopathological features of this lesion show the proliferation of immature fibroblasts and myofibroblastic spindle cells; there is an increased rate of mitoses in these cells but no atypia. Ganglion-like giant cells and lymphocytes are also found.[9]

The treatment options vary from symptomatic therapy with medications such as NSAIDs or steroids to surgical extirpation. According to the literature review, 70% of patients underwent excisional biopsy while 15% of patients had an incisional biopsy, which resulted in spontaneous resolution, so it served as both a diagnostic as well as a therapeutic modality.[2,7,10] Curettage was done in this case, followed by painkillers.

In conclusion, proliferative myositis is a benign entity, but its clinical presentation can simulate malignancy. It should be included as a differential diagnosis when patients present with facial swelling and bone resorption of unknown aetiology, to avoid unnecessary invasive treatments. To the best of our knowledge, not a single case has been reported in the literature of interosseous proliferative myositis.

PATIENT'S CONSENT:

Informed consent has been obtained from the patient's parents to publish this case.

COMPETING INTEREST:

The authors declared no conflict of interest.

AUTHORS' CONTRIBUTION:

ARI: Contributed to the literature search.
AZ: Contributed to the literature search and wrote the report with guidance and refined the report script.
MI: Conceived the idea.
All authors approved the final version of the manuscript to be published.

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