Nodular Fasciitis (Pseudosarcomatous) is a non-encapsulated, rapidly proliferating, benign non-neoplastic lesion arising from the atypical fibroblasts called myofibroblasts of superficial fascia. It was first described by Konwaler et al. in 1955. It is prevalent in upper extremities, followed by head and neck, lower extremities and trunk in the decreasing order. Only 7 to 20% of lesions of upper extremities, followed by head and neck, lower extremities and medical histories were noncontributory.

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INTRODUCTION
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CASE REPORT
A 42-year-old female patient was referred by a general dentist, to the Department of Oral Medicine and Radiology, for evaluation of an asymptomatic, painless swelling refractory to treatment in the left maxillary region, since one month (Fig. 1). Family, personal, and medical histories were noncontributory.

Extraorally, a solitary diffused swelling was evident in the left middle-third of the face measuring 2 × 2 cm in size, extending superio-inferiorly from infra-orbital margin to the angle of the mouth, anterio-posteriorly from naso-labial fold to about 1 cm short of external acoustic meatus. The skin over the swelling was stretched and shiny. On palpation, there was local rise in temperature and it was soft to firm in consistency.

Intraorally measured 1 × 1 cm, extending antero-posteriorly from distal aspect of 24 to the distal aspect of 27. Medio-laterally it was obliterating the buccal vestibule and extended on to the palate 1 cm short of mid-palatal raphe. Overlying mucosa appeared erythematous. Swelling was soft and tender on palpation. In relation to 26, there was a deep carious lesion involving the pulp with tenderness on vertical percussion. Root stump of 24 was noted (Fig. 2).

Based on history and clinical assessment, a provisional diagnosis of 24, 26 related periapical pathoses were arrived at. Intraoral radiograph of 24, 26 along with OPG and PNS radiographs were advised.

Radiographs revealed loss of lamina dura and widened PDL in the periapex of 24. OPG and PNS revealed complete haziness of left maxillary sinus.

Radiographic differential diagnosis of chronic maxillary sinusitis and empyema were considered.

Endodontic treatment was planned for 26 and 24 was extracted under the cover antibiotics with thorough periapical curettage. Post-extraction was uneventful and posted for review after a week.

Patient reported back to the OPD in 3 days time, with an intraoral, non-tender pedunculated reddish brown, foul smelling mass, arising from the 24 extracted site. It obliterated the left upper and lower buccal vestibule. The lesion measured 2 × 2 cm, and extended anterio-posteriorly from the left angle of the mouth to retromolar region, was soft to firm in consistency.

Keeping in mind the enormity and rapidity of the lesion, a differential diagnosis of fibrosarcoma, central reactive giant cell granuloma,
fibromatosis and nerve sheath tumors were considered. An incisional biopsy was carried out.

After the biopsy, patient reported back again within two days, with a further increase in the swelling; the pre-existent swelling had increased further into a huge proliferative reddish brown mass, measuring about 14 × 6 cm, soft to firm in consistency, protruding out of the mouth, causing inability to close the mouth (Fig. 3).

Patient was subjected for CT – contrast of the maxillary region. It revealed huge expansile infiltrating mass arising from the lateral aspect of the superior alveolus of the left maxilla, extending into the maxillary sinus, causing erosion of all the walls of the sinus (Fig. 4).

Histologically, it was composed of cellular lesions of mature proliferating spindle shaped fibroblastic cells, arranged in interlacing bundles with rare typical mitosis, numerous slit-like spaces, often containing erythrocytes, were observed, along with scattered lymphocytes and macrophages, collagenous component with presence of inflammatory cells indicated a reactive process than tumor and the lesion was nonencapsulated (Fig. 5). Findings were indicative of “Pseudosarcomatous fasciitis” of cellular type.

To confirm the diagnosis, the tissue specimens were subjected to immunohistochemistry for markers S-100, and Vimentin. IHC reported the specimen to be negative for S-100 and positive for vimentin. Hence, the nerve sheath tumors were ruled out and origin from myoblastic cells was confirmed.

Due to its highly aggressive behavior and rapid growth, as revealed by the CT, partial maxillectomy was planned to avoid further encroachment of the base of the skull. Postoperative period was uneventful. After three months, a temporary functional and esthetic reconstruction was done by giving an obturator. No recurrence was noted. After a follow-up of one year, a permanent functional reconstruction was planned.

DISCUSSION

Nodular fasciitis is a benign non-neoplastic well encapsulated lesion, arising from the deep fascia, the dense fibrous connective tissue that interpenetrates and surrounds the muscles, bones, nerves and blood vessels of the body. Exaggerated secretion of inflammatory and anti-inflammatory cytokines, growth factors, by fibroblasts (myofibroblasts) in hypoxic conditions can play an important role, in pathological tumor like proliferation of nodular fasciitis. Is also known as ‘pseudosarcomatous fasciitis’, ‘proliferative fasciitis’ and ‘subcutaneous fibromatosis’. Its occurrence is rare in the region of the oral cavity, the reported cases are very few. Males and females are affected equally. Any age group can be affected, but seen most often between 20 to 40 years of age. The size of the lesion varies between 0.5 to 10 cm. Occurs within a short duration and shows rapid growth without evidence of associated infection.

Nodular fasciitis on the basis of its location is divided into three types:

‘Subcutaneous’, ‘Intramuscular’ and ‘Intermuscular’ types. Rare types are ‘Intravascular’ and ‘Intradermal’. Histologically is classified into: ‘Myxoid’, ‘Cellular’ and ‘Fibrous type’ matures from Myxoid type to fibrous type over a period of time.
Contrast CT imaging of nodular fasciitis is seen as a relatively well-defined, soft tissue mass of superficial location. Though histologically benign, deep-seated lesions may show an aggressive clinical behavior simulating malignancy. These invade and destroy the structures, including bone. As a result of rapid proliferative, destructive behavior, it can be clinically mistaken for malignancy.

Surgical resection is the treatment of choice although some authors advocate use of intralesional corticosteroids. Recurrence is rare.

In the present case, the lesion could be characterized into the type of subcutaneous (submucosal) category, of a “cellular” subtype. The lesion was of a very aggressive variety and grew into an enormous size of 14 × 8 cm, weighed 250 gm causing destruction of the left maxilla and antrum, extending up to the infraorbital margin. Histologically was reported as pseudosarcomatous fasciitis. Immunohistochemistry showed positive reaction to ‘Vimentin,’ which is a myofibroblast marker. In the present case, a partial maxillectomy was carried out. Postoperative follow-up was done and there was no evidence of any recurrence for past eight months.

A rare intraoral lesion of nodular fasciitis involving maxillary sinus, with rapid proliferative and destructive behavior, of enormous size (14 × 8 cm) and weight (250 gm) is being reported.

REFERENCES