CASE REPORT

Nodular Fasciitis over Maxillary Gingival Region - A Case Report

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ABSTRACT

In this report, we describe a unique rare case of oronasal nodular fasciitis (NF) over a maxillary sinus region. NF is a benign reactive proliferation of fibroblasts that emerge from the soft tissues, and mostly extending from the muscular fascia to the subcutaneous tissue. The cause of such proliferation is mostly believed to be trauma because of the location of these lesions over bony prominences. In this report, we describe a unique case of oronasal NF aggravated by trauma in a 20-year-old female child over a maxillary sinus region. NF is also known as pseudosarcomatous fasciitis, pseudosarcomatous fibromatosis, and infiltrative fasciitis owing to its rapid rate of growth, rich cellularity, and mitotic activity. Orofacial NF incidence is <20% and affecting primarily adults in 4^{th}–5^{th} decades of life along with review of literature.

Clinical Significance: NF can pose diagnostic dilemma so careful microscopic evaluation with clinical correlation required to differentiate this entity from other lesions to prevent unnecessary work-ups and overtreatment.

Keywords: Rapid mitotic activity, Trauma, Fibroblast, Orofacial nodular fasciitis.

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BACKGROUND

Nodular fasciitis (NF) is a very rare benign tumor-like condition consisting of fibroblast and myofibroblast. It emerges from soft tissue, most probably in response to some local injury and is commonly seen in upper extremities and rarely found in head and neck region. It is confused with myofibromatosis or a sarcoma owing to its rapid rate of growth, rich cellularity, and mitotic activity. It is important to distinguish NF from more aggressive condition. We are presenting a case of NF in a 20-year-old female patient over maxillary gingival region along with review of literature.

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CASE REPORT

A 20-year-old female patient reported, with a chief complaint of swelling and pain in the right upper gums from past 10 days with no relevant history. Local examination revealed a solitary well-defined firm swelling about 1.5 cm × 1.5 cm in size in maxillary gingival region. The teeth in the region showed no abnormality other than mild tenderness on percussion of tooth number 14.

Regional lymph nodes were not palpable. Intraoral periapical radiograph showed periapical radiolucency in relation to apex of 14. As per radiographical and clinical finding, a diagnosis of periapical abscess was arrived at and root canal treatment with simultaneous incision and drainage was done.

At the time of incision and drainage, it was found that the swelling was a tumor rather than an abscess. Incision biopsy was carried out. Histopathological examination gave a diagnosis of NF. The lesion was excised with adequate safety margin under local anesthesia.

Intraoperatively, the roots of 14 and 13 were found to be resorbed along with adjacent bone. Post-operative healing was uneventful and the patient was followed up for 12 months and no recurrence was noted.

On gross examination, specimen is round/oval in shape about 2.5 cm × 2.5 cm in size. Histopathological section revealed stroma with collagen fibers that are arranged irregularly giving it a feathered appearance. A few areas of stroma revealed immature fibroblasts forming short bundle and fascicles. Scattered distribution of mast cells and lymphocytes with few multinucleated giant cells could be discerned. Areas of myxomatous degeneration and many endothelial lined proliferating blood vessels could also be observed. The above clinical and histopathologic findings confirmed the tumor as NF that was present over the anterior wall of the maxillary sinus [Figures 1-6].

DISCUSSION

Nodular fasciitis was first described as subcutaneous pseudosarcomatous fibromatosis (fasciitis) by Konwaler

et al., in 1954.[1,4] It is a benign lesion, but it can often be confused with myofibromatosis or a sarcoma owing to its rapid rate of growth, rich cellularity, and mitotic activity.[1,2]

It can affect any age group but predominantly affect 4th to 5th decade of life with no gender prediction.[2]

Most commonly, it is located in the upper extremities and trunk except viscera.[3] However, the head and neck region is the most common site in children and is most commonly seen on the bony prominence of angle of mandible, zygoma, symphysis, and skull.[4,5]

Intraoral lesions are rare but most commonly seen in tongue, buccal mucosa, and alveolar mucosa.[6] Clinically, the lesion is submucosal, firm, nodular, painless, non-encapsulated, and well-circumscribed mass with variable growth rates. The pain in these lesions is due to perineural extension.[6,7] The lesion is suggestive of malignancy if clinically there is pain, ulceration, rapid growth, and adherence to the surrounding tissues; therefore, differential diagnosis is most commonly given as fibromatoses and fibrous malignancy.[8]

Based on its relationship with the fascia, it is divided into subcutaneous, intramuscular, and fascial. Other uncommon clinical and pathologic variants of NF, such as intravascular, intradermal, cranial, ossifying, and proliferative fasciitis, have also been described.[6]

The pathogenesis of NF in maxillofacial region is uncertain, but two main causative factors found to be associated with the lesion are trauma and over exuberant reaction to innocuous injury or infection.[9] Apparently, this hypothesis is based on two evidences: (1) The presence of these lesions at certain anatomic locations vulnerable to repetitive trauma such as the zygoma, tongue, buccal mucosa, and others[10] and (2) the histological features often mimic atypical reparative granulation tissue.[11] It is also seen that in maxillofacial region the mass is often adherent to the periosteum and causes resorption/erosion of the bone.[13] In our report, it is clear that infection was the initiating factor and later the growth was suddenly accelerated after an insult (i.e., biopsy). As a result, it attained a large size over a short period of time (>2 cm) in association with pain, and the resorption of the bone was seen in the involved tooth.

Histologically, NF is characteristically composed of myofibroblast and fibroblast haphazardly arranged in myxoid stroma composed of mild predominately lymphocytic infiltrate and erythrocyte extravasations. Mitosis is abundant, but cellular atypia is seen. NF is positive to vimentin (fibroblast marker) and actin (smooth muscle marker) and histiocyte marker. To establish differential diagnosis with other myxoid tumors, NF should show muscle specific actin and smooth muscle actin positivity.[12] In our case, it showed positivity to vimentin.

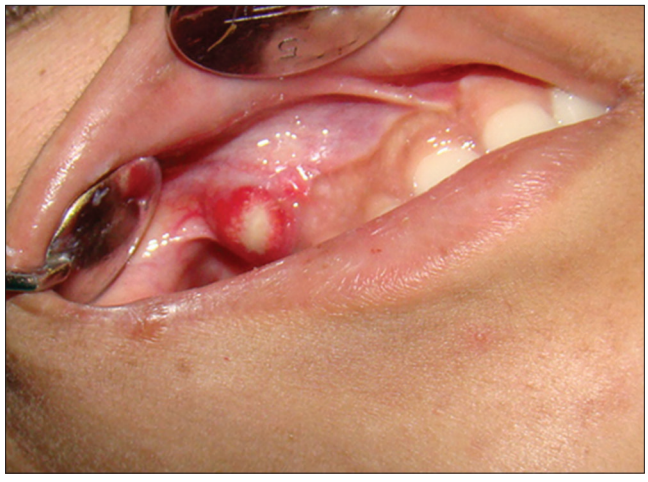

Figure 1: Swelling about 1.5 cm × 1.5 cm in size in maxillary gingival region in relation to tooth number 14

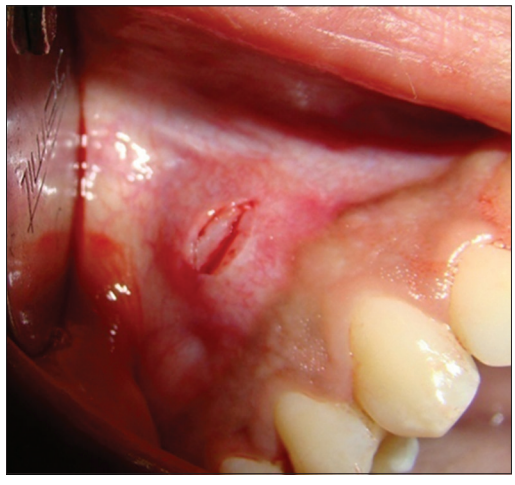

Figure 2: Incision biopsy shows tumor-like swelling than abscess

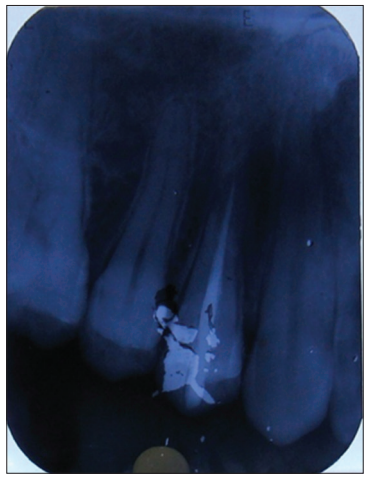

Figure 3: Resorption of root in relation to 14 and 13 along with adjacent bone

At the histological level, variety of benign and malignant lesion can be considered as differential diagnosis. The benign tumors with spindle cells include fibrous histiocytoma, pyogenic granuloma, myofibroma, and

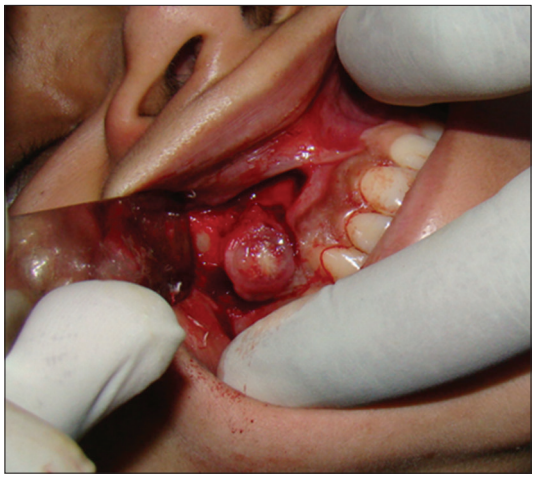

Figure 4: Excision of lesion, mass is adherent to periosteum and causes resorption of bone

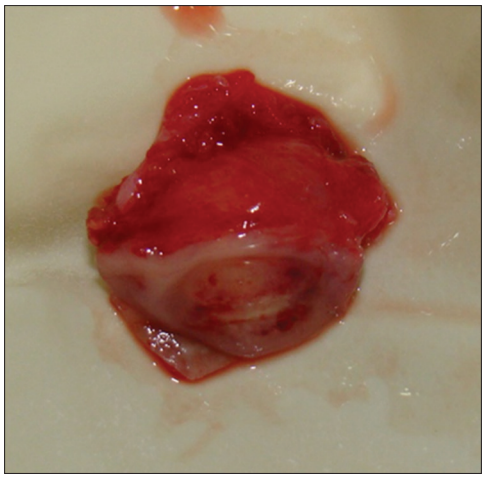

Figure 5: Gross specimen oval in shape

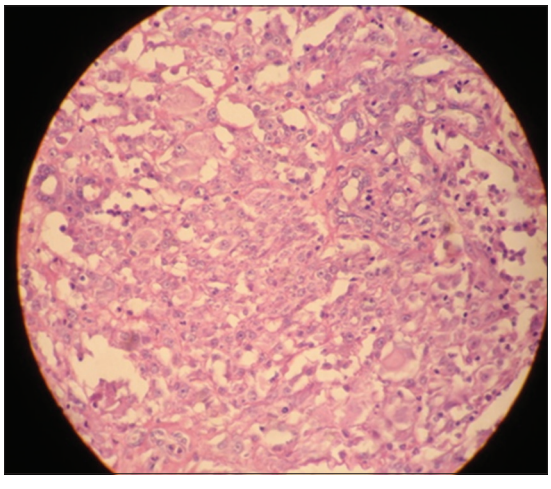

Figure 6: Histopathological section

post-operative/post-traumatic spindle cell nodule. The main differential diagnosis of this tumor is fibrohistosarcoma, presence of the vascular component, the mucinous changes, and the inflammatory infiltrate helps to differentiate NF from fibrosarcoma or soft tissue tumor.[14]

Radiographic findings in computed tomography (CT) and magnetic resonance imaging (MRI) scan are suggestive NF based on its anatomic and histological subtypes. Hussain *et a*l. suggested that lesions which are subcutaneous or submucosal origins appear as well-defined homogenous masses on CT scan and pick up higher signal intensity than that of the surrounding tissues on T2-weighted MRI.[15] On the other hand, intramuscular lesions with fibrous histologic subtype were poorly defined on CT scan, picked up markedly low signal intensity on T2-weighted MRI.[16]

Surgical resection of this lesion is the treatment of choice. Recurrence is rare. If recurrence does occur after excision, an incomplete resection should be suspected or a malignant process should be reconsidered.

CONCLUSION

The sum of short history, rarity of the lesion in orofacial region, non-specific clinical presentation, histologic variations, and variability in anatomic locations, and epidemiologic data make the diagnosis of orofacial NF very difficult.

Unfortunately, many cases of NF were misdiagnosed as more aggressive ones, mainly sarcomatous lesions. Certainly, careful histological assessment is the best available diagnostic tool to distinguish this lesion from more aggressive counterparts.

However, recent advancements in radiography are promising. Several studies have demonstrated characteristic radiographic findings suggestive of these lesions, based on their anatomic locations and histologic subtypes.

Clinical Significance

NF can pose diagnostic dilemma for surgeon pathologists due to its histological similarity with other soft tissue tumors of fibroblastic/myofibroblastic differentiation. Careful microscopic evaluation with clinical correlation required to differentiate this entity from other lesions to prevent unnecessary work-ups and overtreatment.

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