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A Rare Case of Ossifying Fasciitis at The Lower Boarder of the Mandible in A 19-Months-Old Child: Case Report

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1. Abstract

Ossifying Fasciitis (OF) is a soft tissue benign tumor with rare occurrence, that affect most commonly the trunk and extremities with infrequent head and neck involvement. It is considered as a form of nodular fasciitis with similar clinical behavior but different histological findings. It can mimic malignant lesions due to its behavior and reactive characteristics. Ossifying fasciitis affect adults more commonly, and rarely affect children. The primary symptom of ossifying fasciitis has been described as local inflammation associated with pain, but asymptomatic presentation has also been reported. We herein present a rare case of a 19-months-old boy, with history of a rapidly growing hard painless swelling at the right side of the mandible. Resection of the mass, and a subsequent correlation with histopathological and radiological findings resulted in the final diagnosis of ossifying fasciitis affecting the submandibular region at the lower boarder of the mandible. Recovery was uneventful, with more than five-years follow-up showing no sign of recurrence. Despite the resemblance of to malignant lesions, accurate clinical and histopathological examination is crucial to avoid misdiagnosis and consequently unnecessary aggressive treatment.

2. Introduction

Ossifying fasciitis is a benign tumor of the fascia with rare occurrence and close proximity to nodular fasciitis clinically and histpathologically except for the formation of calcified metaplastic bone and chondroid tissue differentiation [1]. Ossifying fasciitis can easily be clinically and histologically misdiagnosed as malignancy, due to its rapidly growing behavior. These lesions affect most commonly the trunk region as well as the upper and lower extremities, and rarely affects the head and neck region. Ossifying fasciitis has the ability to regress following partial resection, and has no tendency for recurrence or metastasis. [2] Accurate clinical and histopathological correlation of this rare benign lesion to avoid misdiagnosing it as a malignancy is crucial for proper patient management and avoiding unnecessary aggressive treatment [2].

We report a very rare case of ossifying fasciitis in the right submandibular region at the lower boarder of the mandible in a 19-months-old male patient.

3. Case Report

We present a case of a 19-months-old male patient brought to the hospital clinics by his parents giving a history of rapidly growing hard painless swelling at the right side of the submandibular region, started four months ago, rapidly increased in size within the last two weeks. Clinical examination revealed bony mass expanding both buccal and lingual cortices, measuring around 3x3 cm in diameter, fixed to the right submandibular region with normal overlying skin appearance, without any sign of infection or inflammation.

Facial bone CT scan with IV contrast was obtained with 3-D reconstruction, revealing submandibular ill-defined soft tissue mass at the right mandibular body and angle, extending to areas of unerupted teeth number 84 and 85, measuring 3.6 x 3 x 2.7 cm.

Incisional biopsy was obtained under general anesthesia using submandibular extraoral approach. Spacemen was examined Histopathologically, revealing variably cellular areas of short irregular bundles and fascicles of fibroblasts and my fibroblasts associated with dense reticulin mesh work in a fibrous and myxoid stroma rich in alcian blue positive mucopolysacchride, along with scattered foci of microhemorrhage. Foci and fragments of reactive and metaplastic bone formation were found within the lesion. Scattered normal mitotic figures and lymphocytes were also seen with no evidence of malignancy, diagnosed as myxoid fibroblastic/myofibroblastic lesion, consistent with ossifying fasciitis.

Accordingly, patient was taken to the operative room for mass resection with peripheral ostectomy using submandibular approach (Risdon), followed by soft tissue layered closure including fascia and subcutaneous tissue (Figure 1). Lesion was excised and tissue collected in four separate specimens marked as, (1) superior tissue biopsy, (2) right submandibular tumor, (3) inferior bone margin, (4) posterior bone margin.

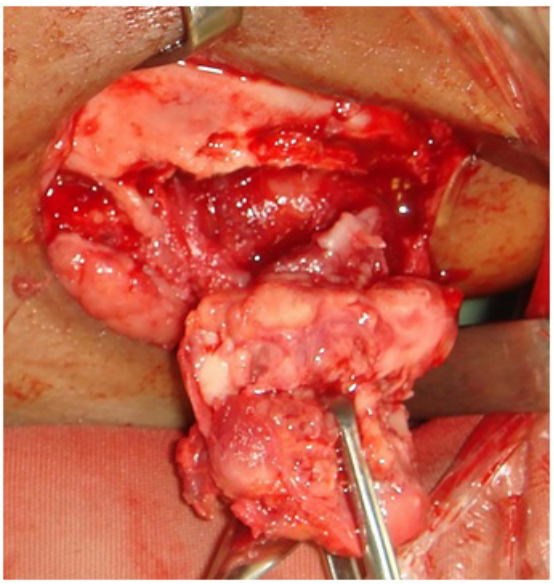

Figure 1: Surgical site following the removal of the OF and peripheral ostectomy

Microscopic description of the specimens came as follow: (1) Fragments of fibrocollagenous tissue and fat, free from tumor. (2) Sections reveal proliferation of uniform plump spindle cells with vesicular nuclei and small nucleoli arranged in long gently undulated C and S-shape fascicles in a myxoid background, alternating with cells with smaller nuclei in a more collagenized stroma. Many arborizing thin walled blood vessels with scattered extravasated RBCs and scattered lymphocytes were also found. Few foci of reactive metablastic bone formation were seen as well. The tumor infiltrates the adjacent muscles and fat, with entrapped skeletal muscles seen in some areas. Occasional non-viable spicules of bone were present at the periphery, (3) Normal active tumor free bone, (4) Normal active cancellous and compact active bone with focal tumor tissue causing pressure on the bone surface (Figure 2). Immunostaining was performed, tumor reacted positive with Smooth muscle actin (SMA), and vitamin, where reacted negative to: S-100P, B catenin, and desmin protein.

Diagnosis came back as right submandibular ossifying fasciitis, concurring the incisional biopsy results. Follow up of the patient was conducted for more than five years with no sign of recurrence (Figure 3).

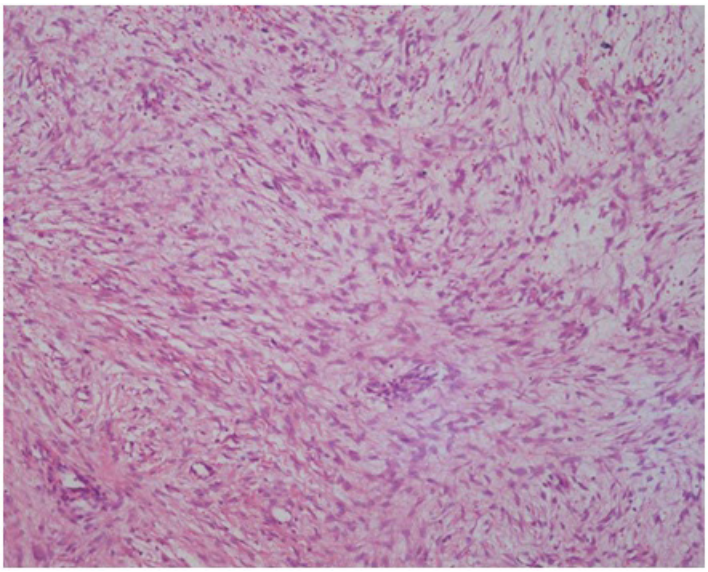

Figure 2: Immature uniform, plump spindle cells arranged in a tissue culture-like pattern with scattered lymphocytes and extravasated red blood cells (20 HPF)

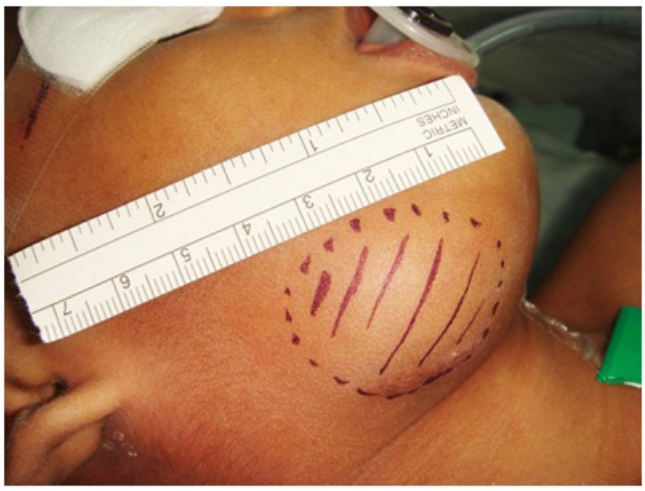

Figure 3: Clinical Dimensions and Presentation of the Right Submandibular Mass

4. Discussion

Ossifying Fasciitis (OF) is a reactive fibroproliIerative lesion of the fascia that can easily be misdiagnosed as malignancy, most commonly sarcoma. The rapid growing behavior along with the abnormal histological features may result in an inaccurate diagnosis and aggressive unnecessary treatment. Konwaler and Weiss in 1955 described fibroblastic and myofibroblastic lesions as inflammatory reactions with multiple variants that most commonly occur following trauma. The currently recognized variants include; nodular fasciitis, proliferative fasciitis, cranial fasciitis of childhood, parosteal fasciitis, and intravascular fasciitis (Figure 4).

It is classified as a type of Nodular Fasciitis (NF), which is a relatively frequent soft tissue pseudo-tumor. [3] However, unlike nodular fasciitis, ossifying fasciitis is very uncommon. [4] Ossifying fasciitis has more prevalence for adults, but it can also affect young children as well as infants. [5] Presenting mainly in the extremities and trunk region with up to15% of cases reporting history of trauma. It has been shown to affect females more than males between 20 and 30 years of age (Figure 5).

The pathological appearance of osteoplastic bone formation differentiates ossifying fasciitis from nodular fasciitis. Intralesional ossification etiology is unclear, which is found to be unrelated to either bony structures or muscle tissues. [10] described the histological appearance of ossifying fasciitis to compose of uniform fibroblasts and immature myofibroblasts, cartilage, bone and osteoid in a rich fibrous (Figure 8, 9, 10) connective tissue. He also described the Intralesional ossification as zonal pattern ossification, presenting focally with osteoid or mature lamellar bone trabeculae. [6] Diversiform cells group in “C” or “S” shaped bundles are also present. Histopathological staining of ossifying fasciitis cells with Smooth Muscle Actin (SMA) and vimentin will reveal positive results, (Figure 6) where cytokeratins, desmin, myogenin, S-100 protein and Anaplastic Lymphoma Kinase (ALK) stain negative, and low Ki-67 proliferation index. [4].

Described soft tissue hyperplasia to take place during the early phase of ossifying fasciitis development, followed by various stages of bone tissue differentiation. [7] The primary symptom of ossifying fasciitis has been described as local inflammation associated with pain. [5] However, (Figure 7) other symptoms such as peripheral neuropathy although rare have been described. [8] Ossifying fasciitis appears radiographically as a soft-tissue lesion with varies degrees of calcification and contrast enhancement, depending on its evolution phase [9].

According to the literature, ossifying fasciitis does not have the tendency to reoccur or metastasize with no reported cases indicating malignant transformation. Therefore, ossifying fasciitis is treated surgically using simple excision. Nevertheless, shrinkage and regression has been reported with partial resection followed by the Intralesional administration of anti-inflammatory medication [10].

According to the literature and up to our knowledge, there are no other reported cases of ossifying fasciitis affecting the head and neck region in young children.

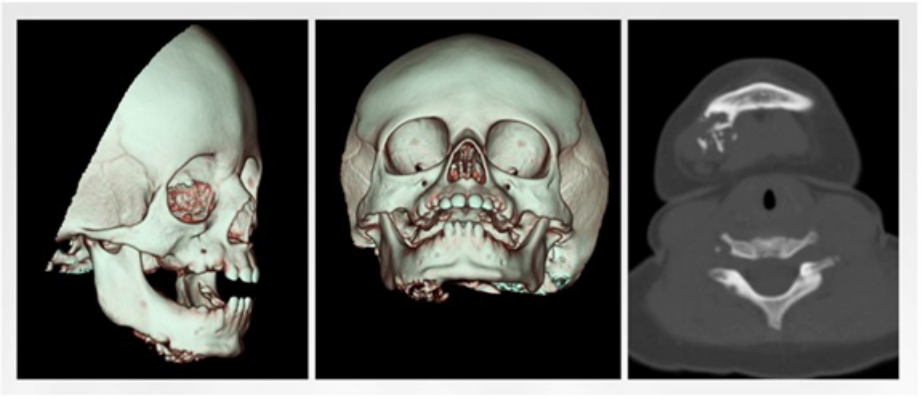

Figure 4: Computerized Tomography with 3-D Reconstruction

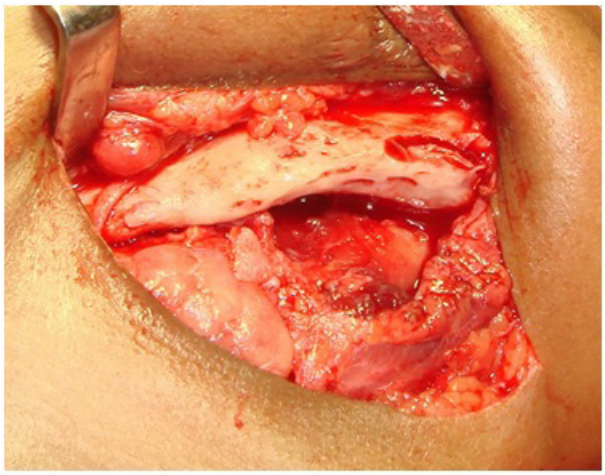

Figure 5: Intraoperative Exposure of the Ossifying Fasciitis Extending from the Inferior Boarder of the Mandible

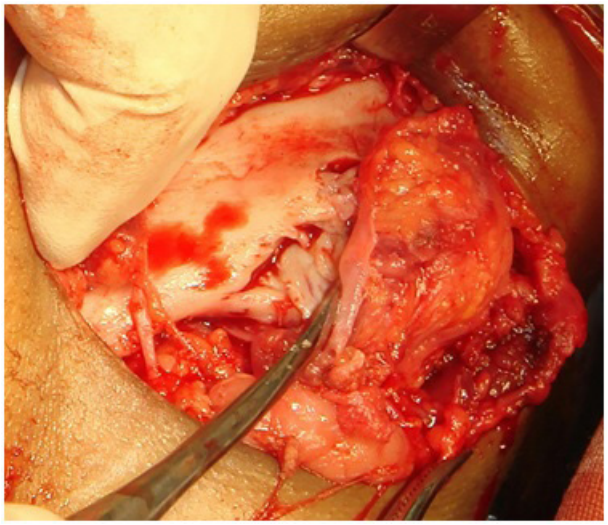

Figure 6: Intraoperative Dissection of the Ossifying Fasciitis from the Inferior Boarder of the Mandible

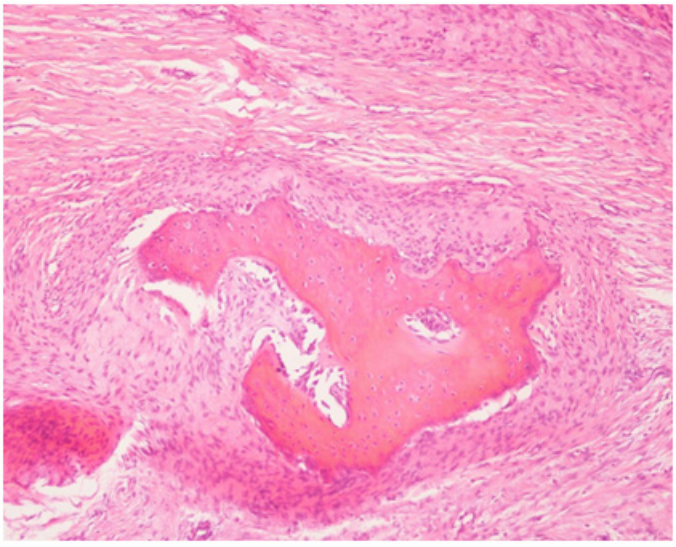

Figure 7: Surgical Site Following the Removal of the OF and Peripheral Ostectomy

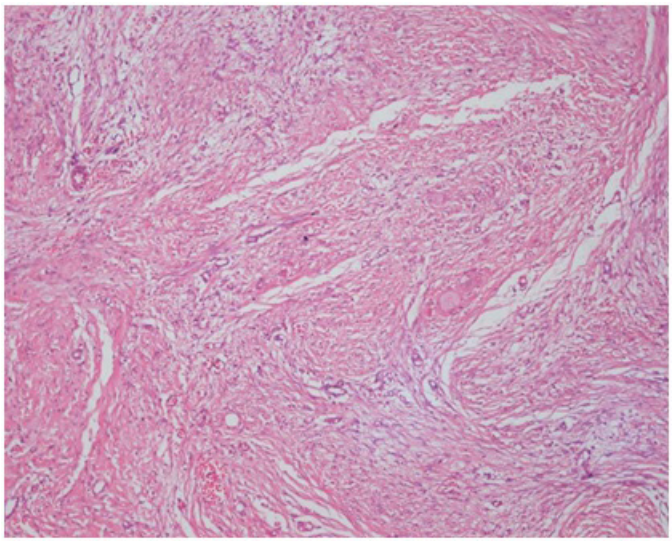

Figure 8: Immature uniform, plump spindle cells arranged in a tissue culture-like pattern with

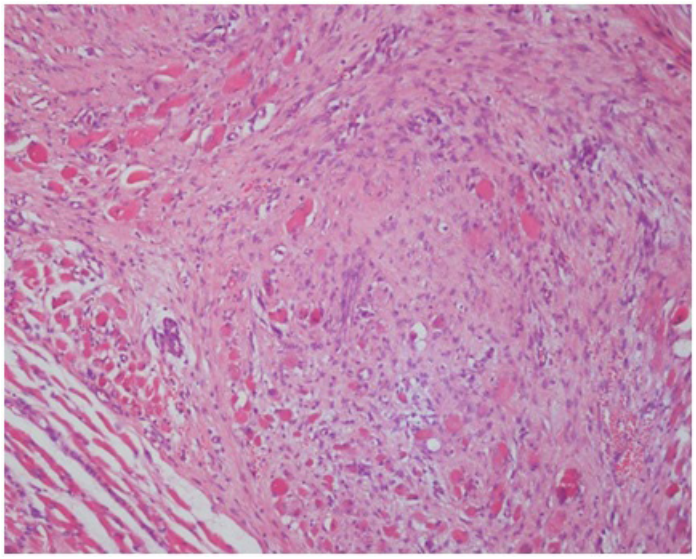

Figure 9: Metaplastic bone formation

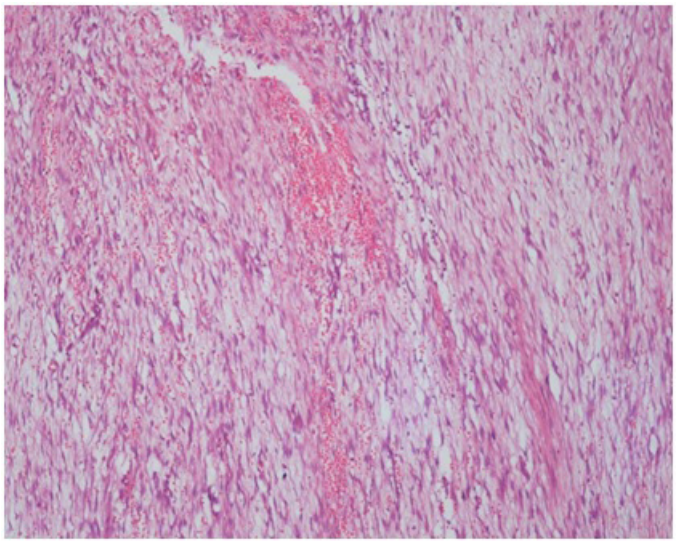

Figure 10: Myofibroblastic cells arranged in short, intersecting fascicles with a storiform pattern

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