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

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Abstract

Ossifying fasciitis is a very rare benign tumor of reactive character that can mimic malignant lesions, especially osteosarcoma. We report a case of a 19-months-old boy, who experienced a rapidly growing hard painless swelling at the right side of the mandible. Resection of the mass, and a detailed correlation with a clinicopathological and radiological analysis led to the final diagnosis of ossifying fasciitis of the submandibular region at the lower boarder of the mandible.

Keywords

Ossifying fasciitis• histopathological• treatment' fasciitis

Introduction

Ossifying fasciitis is a rare benign tumor similar histopathologically to nodular fasciitis, but composed morphologically of metaplastic bone with calcification and chondroid differentiation. This lesion can easily be misinterpreted as malignancy, clinically and histologically, because it presents as a rapidly growing mass originating from subcutaneous or deep fascial tissues. These lesions are usually located in upper and lower extremities and trunk. Ossifying fasciitis typically has no tendency to recur or metastasize; with tendency for regression following partial resection. Accurate clinical and histopathological examination of this rare benign lesion is important to avoid misdiagnosing it as a malignancy resulting in unnecessary aggressive treatment. This report describes a rare case of ossifying fasciitis in the right submandibular region at the lower boarder of the mandible in a 19-months-old male patient.

Review

A 19-months-old male patient presented to the hospital clinics, with 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 (Figure 1).

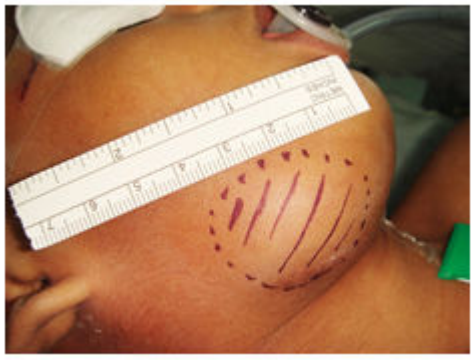

Figure 1. Clinical Dimensions and Presentation of the Right Submandibular Mass.

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 (Figure 2).

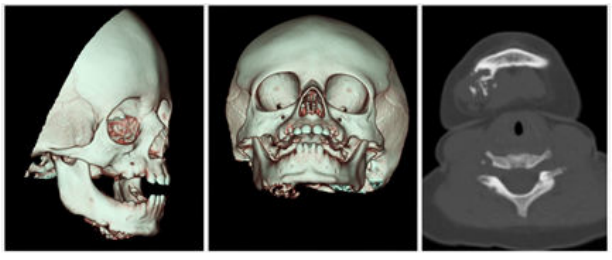

Figure 2. Computerized Tomography with 3-D Reconstruction.

Incisional biopsy was obtained under general anesthesia using submandibular extraoral approach. Spacemen was examined

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Histopathologically, revealing variably cellular areas of short irregular bundles and fascicles of fibroblasts and myofibroblasts 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 3-5).

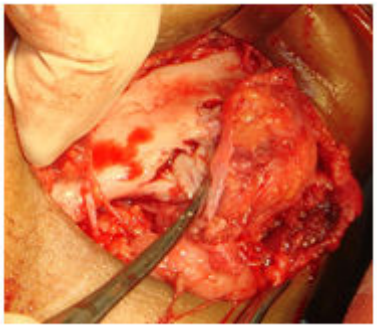

Figure 3. Intraoperative Exposure of the Ossifying Fasciitis Extending from the Inferior Boarder of the Mandible.

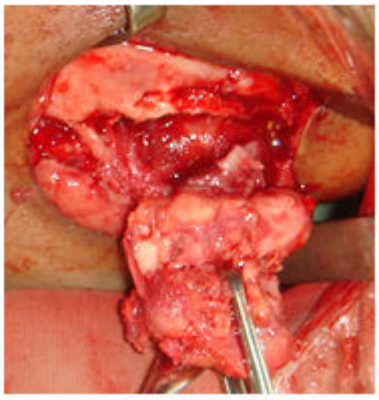

Figure 4. Intraoperative Dissection of the Ossifying Fasciitis from the Inferior Boarder of the Mandible.

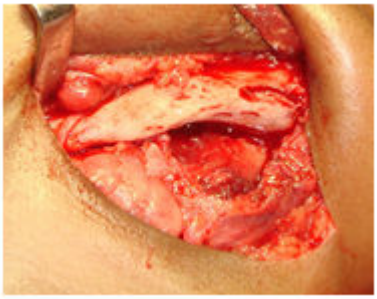

Figure 5. Surgical Site Following The Removal of the OF and Peripheral Ostectomy.

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. 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 spindles 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 6-9).

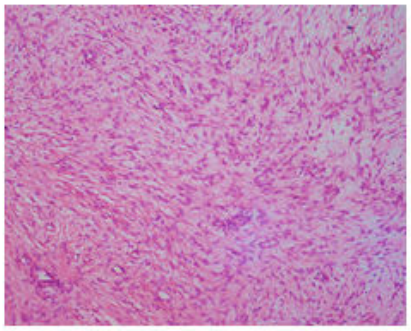

Figure 6. Immature uniform, plump spindle cells arranged in a tissue culture-like pattern with scattered lymphocytes and extravasated red blood cells (20 HPF).

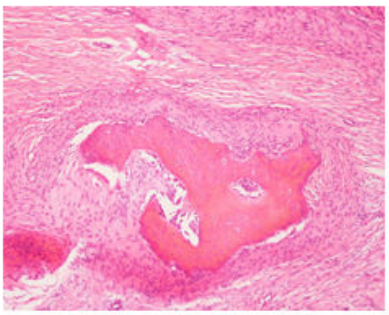

Figure 7. Metaplastic bone formation.

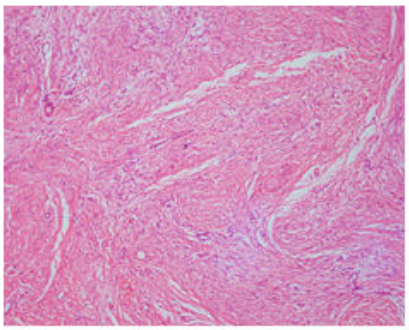

Figure 8. Myofibroblastic cells arranged in short, intersecting fascicles with a storiform pattern.

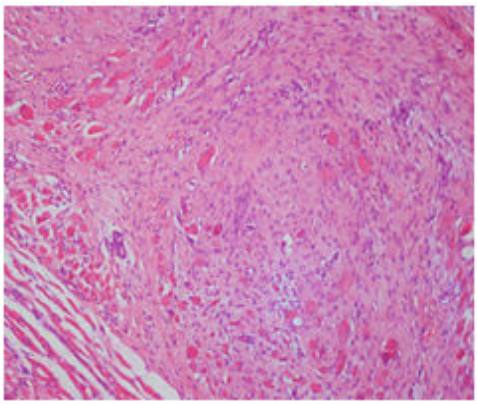

Figure 9. Skeletal muscle entrapment within the plump tumor cell.

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.

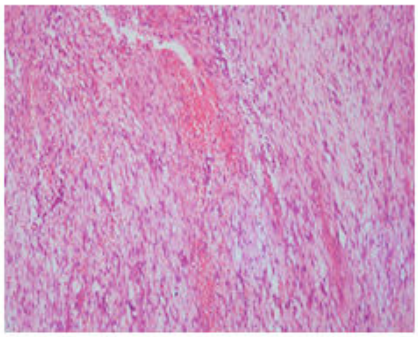

Figure 10. Immature uniform, plump spindle cells arranged in a tissue culture-like pattern with scattered lymphocytes and extravasated red blood cells (10 HPF).

Discussion

Reactive fibroprolilerative lesions of the fascia have long been recognized as diagnostic challenge that can occasionally be misinterpreted as sarcomas. Erroneous interpretation can take place in cases with history of rapid growth and disturbing histologic features. The currently recognized variants include; Nodular fasciitis proliferative fasciitis, cranial fasciitis of childhood, parosteal fasciitis, and intravascular fasciitis. Nodular Fasciitis (NF) is a relatively frequent pseudo-tumor of soft tissue; and Ossifying Fasciitis is classified as one of its variants. However, ossifying fasciitis is extremely rare. Both nodular fasciitis and ossifying fasciitis affect adults more commonly, but the disease can also affect children, including infants. It has been reported that OF most commonly affect females between 20 and 30 years of age, occurring mainly in the upper and lower extremities with a history of trauma in 10-15% of patients. Fasciitis histogenetic mechanism, has been postulated in most reports as a reparative process secondary to soft tissue injury.A history of trauma was elicited in only the minority of cases. The difference between (OF) and (NF) is the pathological appearance of bone formation from osteoblast. It is an uncommon post-traumatic benign lesion of subcutaneous tissue with an unclear etiology of ossification which is neither related with a bony structure nor muscle tissue. Ossifying fasciitis, occurs as a superficially located tumor. It is composed of richly cellular fibrous connective tissue with uniform fibroblasts and immature myofibroblasts, cartilage, bone and osteoid. Ossification in ossifying fasciitis contrast to zonal pattern ossification, occurring in ossifying myositis. It is present focally and it consists of osteoid or mature lamellar bone trabeculae. Diversiform cells group in “C” or “S” shaped bundles are also present. Ossifying fasciitis cells are stained positive with Smooth Muscle Actin (SMA) and vimentin, and negative for cytokeratins, desmin, myogenin, S-100 protein and Anaplastic Lymphoma Kinase (ALK).

Conclusion

The Ki-67 proliferation index is usually low. During the first week of ossifying fasciitis development (early phase), soft tissue hyperplasia takes place. In the following weeks, various stages of bone tissue differentiation become visible in the architecture. The tumor reaches a characteristic macroscopic size of 3 cm on average. Pain and local inflammation is considered to be the primary symptom to initiate a diagnostic process. However, rare symptoms, such as peripheral neuropathy, were also described. A radiological examination may reveal a soft-tissue lesion, characterized by various degree of swelling, calcification and contrast enhancement-depending on the phase of evolution Ossifying fasciitis has no tendency to recur or metastasize, and usually treated by simple surgical excision. However, cases of ossifying fasciitis shrinkage after administration of anti-inflammatory medication, and regression following partial resection have also been reported. Metastases are extremely rare, with no reported cases in the literature indicating malignant transformation. According to the literature, this is the first reported case of ossifying fasciitis affecting the head and neck region in young children.

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