Recurrent Non-Ossifying Fibroma in Shaft of Distal Radius – A Rare Case

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Learning Point of the Article:

Non ossifying fibroma can be curetted when it becomes symptomatic. Extended curettage will not be necessary as residual tumour spontaneously regress with growth.

Abstract

Introduction: Non-ossifying fibroma are is a common benign lesions found in children. Usually, they are found in cortical bone in metaphysis, but they can also be seen in cancellous bone. Theses lesions are known to regress spontaneously. Usually, it is asymptomatic and an incidental finding on radiograph. No intervention is required. Here, we are reporting a case of symptomatic non-ossifying fibroma of radius with unusual characteristics which required surgical intervention.

Case Report: An 11-year-old skeletally immature girl presented to us two 2 years back with pain and swelling over the right forearm without any restriction of moments and other constitutional symptoms. She was diagnosed to have non-ossifying fibroma and had underwent surgery for the same 4 years back. Radiographic and higher imaging studies suggested non-ossifying fibroma. Since the lesion was painful and the child had difficulty in carrying heavy objects, we decided to intervene. Tumour tissue was thoroughly curetted and the defect was filled with artificial bone substitute. Biopsy confirmed the diagnosis of non-ossifying fibroma and post-operative radiograph showed some residual tumour which was noted even at one 1-year follow-up X-ray. On 3-years follow-up, the patient was symptom-free with no residual lesion and complete incorporation of the artificial bone substitute.

Conclusion: Though Although most of the NOF regress completely without intervention, large and symptomatic lesions in unusual location require to be surgically treated. The natural history of the lesion – spontaneous regression also aids in the management.

Keywords: Diaphysis of radius, management, non-ossifying fibroma, pediatrics, unusual presentation.

Introduction

The term fibrous cortical defect and non-osteogenic (non-ossifying) fibroma were coined by Jaffe and Lichtenstein, in 1942, when they found fibrous tissue in the lesion, they had biopsied from distal femur [1]. These fibrous defects are common benign lesions found in children. Usually, they are found in cortical bone in metaphysis, but they can also be seen in cancellous bone. Other terms used to describe these fibrous lesions include fibrous metaphyseal defect and fibrous endosteal defect. Huvos [2] defined the tumor as a well-delineated lytic lesion in the metaphyseal region of long bones. The vast majority of the lesions develop in the metaphysis of the

long bones of the lower extremities [3]. Histologically, it is characterized by a fibroblastic, dense cellular proliferation in a mottled, whorled growth pattern admixed with multinucleated giant cells, and foamy xanthomatous cells[4]. The fibrous presentation is following post-infarction necrosis and is probably not a true neoplasm [5]. The lesion is known to regress [6]. Usually, it is asymptomatic and an incidental finding on radiograph. No intervention is required [7, 8]. Very rarely, when the lesion involves a significant width of the bone (more than 50%), the patient can have pain and pathological fracture [9]. Here, we are reporting a case of symptomatic non-ossifying fibroma of radius with unusual characteristics.

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Figure 2: Pre-operativeX-ray showing 1cm diameter multiloculated lesion involving the complete diameter of with no periosteal reaction and cortical breach.

Case Report

An 11-year-old skeletally immature girl presented to us 2 years back with pain and swelling over the right forearm without any restriction of moments and other constitutional symptoms. The patient had a similar problem 4 years back (Fig. 1), for which she underwent surgery elsewhere and she completely recovered. Available biopsy reports suggested a non-ossifying fibroma. On examination, the patient had a tender bony thickening of radius extending to about 2cm underneath the previous surgical scar which was a 5cm anterolateral longitudinal surgical scar in the region of the middle third and distal third junction of forearm. The patient had no neurovascular deficit.

X-ray radiography showed a 1cm diameter multiloculated lesion involving the complete diameter of bone with sclerotic rim in the distal diaphysis of radius, with no periosteal reaction and cortical breach (Fig. 2). Magnetic resonance (MR) imaging showed a well-defined expansile lytic, trabeculated lesion with multiple internal septations region (Fig. 3a and b). Since the previous biopsy report from a reputed institute suggested nonossifying fibroma 4 years back, we did not go for further studies like bone scan. As the lesion was painful and the child had difficulty in carrying heavy objects, we decided to intervene. Based on the previous biopsy report and imaging features, we decided to perform direct intralesional excision (curettage) of

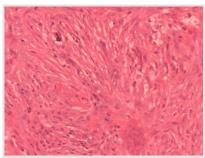


Figure 4: Photomicrography showing tissue with spindle cells arranged in fascicles and bundles in a storiform pattern along with osteoclast-like giant cells, no evidence of cytological atypia/atypical mitosis/necrosis suggestive of non-ossifying fibroma.



Figure 5: Two-month post-operativeX-





Figure 3: (a) Pre-operativemagnetic resonance imaging(MRI). (b) Pre-operativeMRI, welldefined expansile lytic, trabeculated lesion with multiple internal septations region.

the lesion. Radius was exposed through the previous surgical bone with sclerotic rim in the distal diaphysis of radius, scar (Henry's approach) without tourniquet control. During exposure, we found gross adhesions around radial artery due to the previous exposure. Radial artery was accidentally cut, repair was not attempted and hence ligated. Under image intensifier guidance, the lesion was identified and bone window was made. Tumor tissue was thoroughly curetted and the defect was filled with artificial bone substitute (STIMULAN calcium sulfate beads).

> Tumor tissue was sent for biopsy. Above elbow, POP slab was applied for 6 weeks. Histopathology showed tissue with spindle cells arranged in fascicles and bundles in a storiform pattern along with osteoclast such as giant cells and no evidence of cytological atypia/atypical mitosis/necrosis suggestive of nonossifying fibroma (Fig. 4). The patient was put on the above elbow POP slab for 2 weeks and converted to POP cast for the next 4 weeks. At 6 weeks, POP was removed and an active range of motion exercises of wrist started. The patient was advised not to involve in sports activities for nearly 6months until bony union was seen radiologically. The patient was serially followed up. Post-operative radiograph showed some residual tumor involving the dorsal cortex which was noted even at 1-year follow-up X-ray (Fig. 5). The patient was followed up for 3 years. Tumor was completely regressed and was not seen on X-ray (Fig. 6). She had a full range of movement with no functional deformity (Fig. 7a-d).

Discussion



Figure 6: Three-year postoperativeX-ray.

Hatcher [10] stated that the lesion was developmental rather than neoplastic and called it "metaphyseal fibrous defect." At present, if the lesion is small and localized to the cortex, it is called "fibrous cortical defect." On its extension to the medullary canal, it is called "non-ossifying fibroma [4]." Non-ossifying fibroma is commonly found in the metaphyseal region and very unusual in the diaphysis. MR coronal images give the maximum









Figure 7: (a-d) The clinical images after 3-year follow-up.

extension of tumor lesion [11]. In our case, it was found in the diaphysis and also had recurrence after initial surgery which is rare. Non-ossifying fibromas are usually asymptomatic as the lesion is present within the cortex of bone, but in our patient, the lesion was involving the whole diameter of bone and therefore symptomatic. Although the residual lesion was present post-curettage until 1 year of follow-up, complete regression of the tumor was noted radiologically in the follow-up radiographs. We encountered a major complication of accidental radial artery cut during dissection of the lesion. Ligation of the artery was done. However, the patient developed good radial pulse in follow-up probably due to ulnar artery collaterals. The patient is symptom-free 3-year post-operative and with no residual lesion and complete incorporation of the artificial bone substitute.

Conclusion

Although most of the NOF regress completely without intervention, large and symptomatic lesions in unusual location require to be surgically treated. The natural history of the lesion – spontaneous regression also aids in the management.

Clinical Message

Non-ossifying fibroma can be curetted when it becomes symptomatic. Extended curettage will not be necessary as residual tumor spontaneously regresses with growth.

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