Non-Ossifying Fibroma in Posterior Mandible, A Case Report

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Significance:
The significance of non-ossifying fibroma in the mandibular region lies in its unique combination of benign nature, rarity, asymptomatic presentation, distinct radiographic appearance, and the need for careful consideration of treatment and follow-up strategies. Continuous research and documentation of cases contribute to improving our knowledge of this rare entity in the mandible

Abstract
In this case report, we present a case of non-ossifying fibroma in the mandible of an eleven-year-old girl who reported to the Oral and Maxillo-Facial Surgery Department with facial asymmetry and painless swelling on palpation over the lower right mandibular region. In this paper, we review the radiographic features, clinical presentation, and histopathological features of non-ossifying fibromas. Non-ossifying fibromas present with a unique extra-gnathic appearance in the skull. It is usually asymptomatic and has diverse histological features. Co-related histological, radiological, and clinical features differentiate it from other odontogenic and non-odontogenic tumors and cysts of the oral cavity. Ortho-pantogram revealed a well-demarcated, expansile radiolucency with pronounced sclerotic borders with striations reaching the anterior and superior regions and thus slowly approaching a multilocular stage. The treatment plan comprised of excisional resection with curettage under general anesthesia. After excisional biopsy of the lesion, the specimen was sent for histopathological evaluation in neutrally buffered 10 % formalin solution, which confirmed it to be a Non-Ossifying Fibroma. Prior to surgery, informed consent and written permission to collaborate with a case report encompassing her findings were obtained from the patients' guardians. Confidentiality of patient has been taken care of before documenting the report. A meticulous literature review of previous case reports on non-ossifying fibromas revealed typical behaviors and characteristics of this lesion.

Introduction:
Non-ossifying Fibroma is a common, benign, non-neoplastic lesion that almost exclusively occurs in the metaphysis of the long tubular bones in children and adults, while a rare occurrence of non-ossifying fibroma has been reported in the mandible (1). Non-ossifying fibromas were first recognized by Sontag and Pyle in 1941 (2). According to World Health Organization it is defined as "a tumor like lesion grouped according to the histological classification of bone tumors'' (3). Non-ossifying Fibroma is one of the most common benign asymptomatic lesions of the skeletal system, and has an asymptomatic characteristic extra-gnathic radiographic appearance (4).

To date, only three cases of Non-Ossifying Fibroma have been reported in the cytogenic literature, which show the presence of diploid karyotypes with no structural abnormalities.(5) In the initial years when Jaffe and Lichtenstein recognized non-ossifying fibroma, it became evident after conducting a periodic survey analysis that non-ossifying fibroma starts as a cortical lesion, with a typical appearance in the metaphysis of long bones where it targets the cortical bone, and over the time of span, this lesion expands more.(6) It is widely accepted that these lesions are reactive or developmental anomalies but not true neoplasms (7).

In non-ossifying fibroma, the clinical and radiological features can help us to lead a provisional diagnosis of osteogenic or odontogenic lesions; for example, it would be asymptomatic more often, non-tender to palpation, an expansile uniform mass, thinning of the cortical bone, and sclerotic well-defined borders(8). In differential diagnosis, FCD, which typically presents as an eccentric osteolytic defect, is usually considered. Histological evaluation helped us reach a confirmatory diagnosis (9).

At present, the two most common terms used to describe these lesions are non-ossifying fibromas and fibrous cortical defects. They are among the most common benign lesions reported in the skeletal system, mainly affecting long metaphyseal bones. It has been noted that while non-ossifying fibromas have a great tendency to affect long bones in children or adults, they have rarely been reported to occur in the mandible or maxilla (10).

Case Report:
An 11-year-old girl reported to the Oral and Maxillofacial Surgery Department of Abbasi Shaheed Hospital, Karachi, Pakistan on 29th of March, 2022, with a chief complaint of facial asymmetry and a gradually increasing mass in the lower jaw for 1 year. The growth started as a small solitary mass in the right premolar of the mandible. It was, asymptomatic and presented as a slight swelling which was not tender to palpation or otherwise and measured about 7.0 x 3.0 x 1.6 cm in AP x CC x TR dimensions. It was firmly attached to the mandibular right premolar region. There was no paraesthesia of the surrounding skin nor there any history of facial trauma. None of the lymph nodes were palpable. Intraoral examination revealed grade 1 mobility of the right mandibular 1st and 2nd premolar tooth. Systemic history was not significant.

After taking proper aseptic measures, the lesion was resected entirely along with right canine, 1st premolar and 2nd premolar of right mandible and was submitted in 10 % buffered formalin solution for histopathological investigation which confirmed it to be a Non-Ossifying Fibroma.

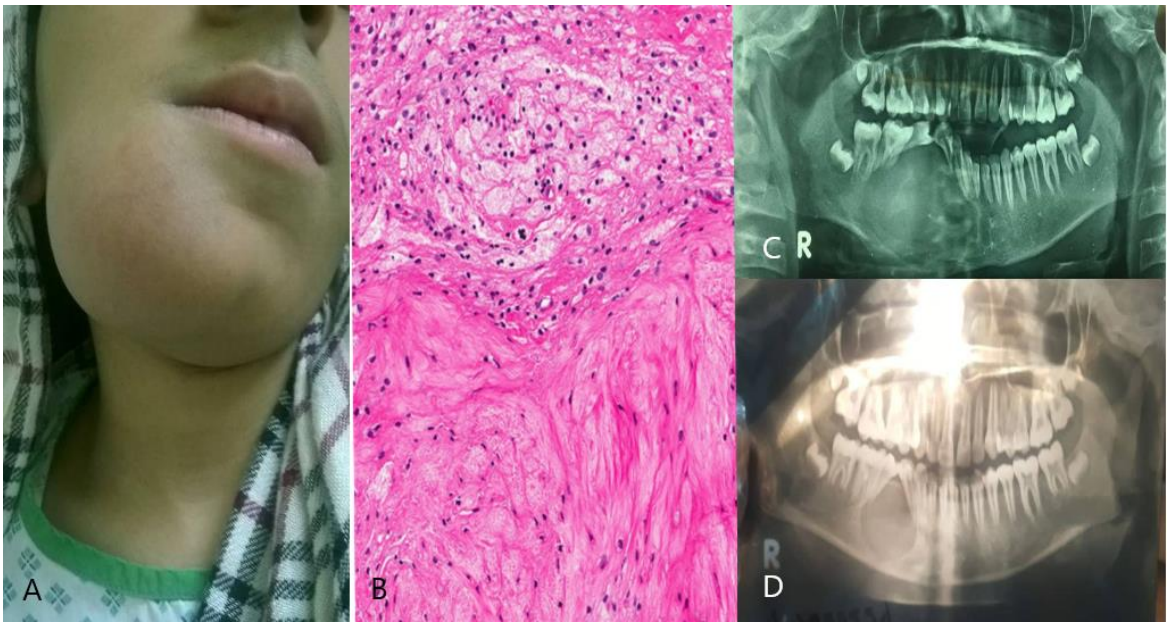

Figure 1 (A): Extra Oral Facial Asymmetry at right Side of Face. **(B):** The Non-Ossifying Fibroma, Enhanced magnification × 180: Hematoxylin and Eosin stain reveal clusters xanthoma cells adjacent to spindle cells. **(C):** Panoramic Investigation revealing more expansile destruction of the right posterior mandibular region on March 29, 2022. **(D):** Panoramic investigation performed by another dentist a year ago.

Histopathological Features

Histopathological examination revealed fragments of lesions composed of interlacing bundles and storiform areas composed of spindle-shaped cells adjacent to xanthoma cells with bland morphology and abundant eosinophilic cytoplasm with elongated nuclei. (Figure 1 (B)). Bundles of whorled connective tissue stroma are also observed. Some mitosis was seen. Scattered, multinucleated osteoclast-like giant cells were observed. The mitotic activity was appreciable. These cells were negative for immune-histochemical stain CD34.The lesion also encompassed bony tissue fragments exhibiting some degree of osteogenesis, and no evidence of malignancy was observed.

A meticulous histological examination by immunohistochemistry revealed patchy xanthoma cells along with scattered expression of CD68 and CD163.An unusual location and lack of factor 13 were noted. A confirmatory diagnosis of non-ossifying fibroma was concluded to be the dominant cell population consisting of storiform fibroblasts that differentiated it from reaching to a diagnosis of xathomatous histiocytes.

Radiographic Features:

In this case, after receiving the panoramic investigation, Non-Ossifying Fibroma can be regarded as an expansile, well-demarcated, unilocular lesion with scalloped sclerotic borders causing expansion of the posterior mandible along with thinning of the cortical bone. Root resorption was not appreciable, but gross displacement of the posterior right mandibular teeth was evident. Computed tomography revealed destruction of the posterior right mandibular bone with marginal sclerosis and a heterogeneous mass expanding the posterior mandible. causing its expansion. Figure 1 (C)

The patient was also able to provide an ortho-pantogram that she had received a year ago, as prescribed by another dentist (Figure 2). When comparing these investigations, it was easily appreciable that an earlier lesion presented as a small oval-shaped unilocular lesion, with tilting of the right mandibular 1st and 2nd premolars towards each other, a well-defined sclerotic border along with less cortical thinning of the bone and less expansion of the lesion into the mandibular bone.

Treatment:

Non-ossifying fibroma, when occur in jaws, have shown to expand over a span of time and occur as an asymptomatic entity. A well-defined radiolucency occurring in the jaws, having sclerotic borders can be compared to the normal radiographic features in the long bones. In 2012, Bowers et al. performed peripheral ostectomy with curettage in a 22-year-old female patient diagnosed with non-ossifying fibroma of the mandible. [4] In 2011, Chrcanovic et al. treated a 15-year-old girl using simple curettage. Large lesions of non-ossifying fibromas can be treated with segmental resection (7).

In the present case, resection was performed to treat the lesion. Figure 2 A and B.

Follow Up:

The patient was advised to undergo a follow-up visit after six months. No evidence of recurrence was found on intra and extra oral examination (Figure 3A).

The face appeared symmetrical, and the patient was satisfied with the results. (Figure 3B).

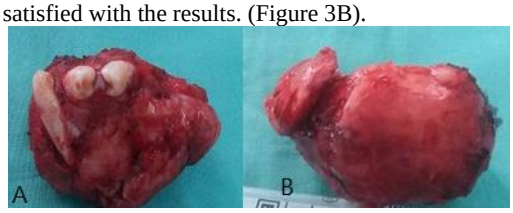

Figure 2: **A**: Resected lesion (Anterior View) **B:** Resected Lesion Posterior View

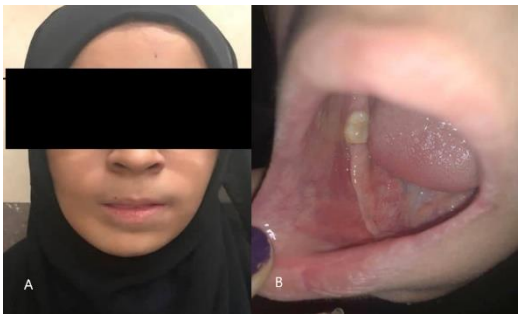

Figure 3A: Facial Symmetry was appreciable on follow-up appointment.

Figure 3B: Intraoral examination revealing a healed area where resection was performed.

Discussion:

Non-Ossifying Fibroma is a common benign, non-neoplastic lesion that usually occurs in the metaphysis of long bones in adults and children (5). The histopathological features of non-ossifying fibromas vary greatly because different terms were initially defined, including non-osteogenic fibroma, histiocytic fibrous defect, fibrous cortical defect (FCD), Central Giant Cell Granuloma, Metaphyseal Fibrous Defect (MFD), Benign Fibrous histiocytoma (BFH), histiocytic xanthoma, fibroxanthoma, histiocytic Xantho granuloma, and Fibrous Xanthoma. Later, more dedicated studies regarding Non-Ossifying Fibroma revealed differences between these terms as for instance, despite similar histopathological features of Non-Ossifying Fibroma and Focal Cortical Defect, it was seen that Non-Ossifying Fibroma is more elongated, larger, involves the medullary portion of bone, and lies parallel to the long axis of bone when compared to Focal Cortical Defect, so these two entities can be differentiated on the basis of these features (6).

Various studies have suggested that Non-Ossifying Fibroma can occur as a result of trauma that can disturb the ossification centers of the bone, thereby altering the calcification process (1).

At present, Non-Ossifying Fibroma is considered to be a benign, asymptomatic non-neoplastic lesion originating in the metaphysis of long bones, but in 1942, Lichtenestein and Jaffey suggested that Non-Ossifying Fibroma can affect any bone and not just the long bones (7). Non-ossifying Fibroma has rarely been reported to affect the mandible, with a greater tendency to occur in the ramus and posterior region of the mandible, as these areas are believed to contain mandibular growth centers; thus, etiology of Non-Ossifying Fibroma occurring in mandible mimics the pathogenesis of Non-Ossifying Fibroma occurs in any long bone (8). The case presented in this report describes the features of Non-Ossifying Fibroma occurring in the premolar region of the mandible of an 11-year-old girl. Non-Ossifying Fibroma has been reported to affect individuals who are usually less than 20 years of age, but can also occur in adults. Various studies have revealed that the mean age for Non-Ossifying Fibroma to occur in the mandible is 21 years, and men tend to have these lesions in long bones, while most mandibular NOFs occur in females. Mandibular lesions can be asymptomatic or lead to slight to moderate swelling (4). Due to their rare occurrence in the jaw, these lesions can often be misdiagnosed as odontogenic tumors or jaws (8).

Radiographically, they present as oval or round radiolucent lesions with smooth lobulated edges and a well-demarcated sclerotic border, along with thinning of the cortical bone. Although on radiographic studies it has seen that Non-Ossifying Fibroma can be multi-locular too but in most of the cases, it presented as a uni-locular lesion (9)

In this case, Non-Ossifying Fibroma can be regarded as a well-demarcated, unilocular lesion with scalloped sclerotic borders causing expansion of the posterior maxilla. Initially, a differential diagnosis of odontogenic myxoma, odontogenic keratocyst, ameloblastoma, and focal cortical defect was made; however, based on radiographic and histological findings, it was concluded to be a Non-Ossifying Fibroma.

Some researchers believe that in the jaw, central giant cell granuloma (CGCG) is the mandibular presentation of the long bones. Non-ossifying Fibroma. However, histopathological studies revealed differences between these two entities, as resected specimens of Non-Ossifying Fibroma usually reveal bundles of whorled connective tissue stroma, whereas only minor patches of connective tissue are seen in central giant cell granuloma. Moreover, Non-Ossifying Fibroma has foam cells that are absent in central giant cell granuloma, and the absence of osteogenesis in central giant cell granuloma can be diagnosed more conclusively (10).

Table 1: Summarized Table of clinical and radiographic features of Non-Ossifying Fibroma in Mandibular Body

S. No.	Author Name	Year	Age/Gender	Location	Unilocular/Multilocular	Treatment
1	Liaw et al.(12)	1979	17/F	Posterior mandible	Unilocular	Resection
2	Makek (13)	1980	20/M	Condyle	Multilocular	Resection
3	Ide et al. (14)	1982	37/F	Body	Multilocular	Curettage
4	Mirra et al. (15)	1982	12/F	Body	Unilocular	Curettage
5	Park et al. (16)	1982	21/F	Body	Unilocular	Curettage
6	Elzay et al.(11)	1984	11/F	Ramus	Multilocular	Curettage
7	Bessho et al. (17)	1986	28/M	Body	Unilocular	Curettage
8	Aldred et al. (18)	1989	18/F	Condyle	Multilocular	Resection
9	Mizukawa et al. (19)	1997	7/M	Body	Unilocular	Curettage
10	Uçkan et al (20)	1999	16/F	Body	Multilocular	Curettage
11	Bailey et al. (9)	2001	6/F	Angle	Multilocular	Curettage
12	Hudson et al. (21)	2003	13/M	Condyle	Unilocular	Curettage
13	Chrcanovic et al. (1)	2010	15/M	Angle	Unilocular	Curettage
14	Abdelsayedet al. 1st Case (8)	2010	14/F	Ramus	Multilocular	Curettage
15	Abdelsayedet al. 2nd Case (8)	2010	27/M	Ramus	Multilocular	Curettage
16	Leah M Bowers (4)	2011	22/F	Ramus	Multilocular	Curettage
17	Present Case	2019	11/F	Posterior Mandible	Unilocular	Resection

Non-ossifying fibroma is usually treated by curettage or enucleation and no matter how extensive the surgical intervention had been, re-occurrence has not yet been reported.

Conclusion:

Non-ossifying fibroma is benign and less aggressive lesions that usually occur in metaphysis of long tubular and its occurrence in mandible has been reported to be rare. Due to its radiographic and clinical features, it poses a challenge for surgeons to diagnose accurately; however, histopathological investigations enable us to reach a confirmatory diagnosis. The reoccurrence of these lesions has not yet been reported.

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