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CASE REPORT

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Massive ossifying fibroma of mandible: A case report and review of the literature

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Key Clinical Message

Benign fibro-osseous neoplasm. Massive size is rarely reported. May be confused with other pathologies such as fibrous dysplasia or osteosarcoma. Aggressive nature and high recurrence pose management challenges. Treated by surgery.

Abstract

Ossifying fibroma is a benign fibro-osseous neoplasm. It can affect both mandible and maxilla. Precise diagnosis can be challenging due to significant overlap of clinicopathological features with other neoplasms. Case reports with massive tumor sizes as presented in our case are uncommon. Huge tumor size can cause alarm for other pathologies such as osteosarcoma. The radiological tests should reassure the attending practitioner and histological examination confirm the diagnosis. The aim of the present report is to discuss a case of a giant ossifying fibroma in a 13-year-old male child. He presented with a progressive mandibular mass for 4 years. Clinical, radiological, and pathological characteristics and surgical treatment approaches are further discussed. This is one of the rare cases of massive ossifying fibroma in the English literature.

KEYWORDS

benign, fibro-osseous neoplasm, mandible, ossifying fibroma

1 | INTRODUCTION

Ossifying fibroma (OF) is a benign fibro-osseous lesions. Other fibro-osseous lesions in the jaw bones include fibrous dysplasia, cemento-ossifying fibroma, florid osseous dysplasia, and focal osseous dysplasia.[1,2] The cause is unknown; however, studies have demonstrated mutations of HRPT2 gene in fibroma ossificans, suggesting a genetic factor in its occurrence.[3] OF is derived from progenitor cells of the periodontal membrane, capable of differentiation into fibroblasts, osteoblasts, and cementoblasts.[4] Juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF) are two histologic variants of OF.[5] This neoplasm exhibits progressive slow enlargement and bone expansion that can result in asymmetry, facial disfigurement, and malocclusion. The commonly affected age varies from first to fifth decade of life with a definite female predilection. Clinically, OF is characterized by an asymptomatic lesions of highly variable sizes and generating bone expansion in the cortices and osteolysis of the affected area. Cases with massive sizes as presented in our patient are rare. The mucosa

overlying the lesion usually is of normal appearance. Mandible is commonly affected than maxilla. The case study of unusual massive mandibular OF in a teenager successfully treated by an en bloc jaw resection in our institution is presented. A brief overview of the literature has been provided.

2 | CASE PRESENTATION

A 13-year-old male child was brought to dental clinic of our institution because of a mandibular new growth. The father of the child was conscious of the mass since 4 years. It was slow in onset, enlarging over the years to the present size and was not associated with history of toothache or trauma of the jaw. At first, the mass was asymptomatic but since a year ago, it became painful on closure of the jaws because of contact of upper teeth with the lesion. The patient had no significant past medical or surgical history.

Clinical examination revealed a huge mass measuring 16 × 12 cm in its greatest dimension on the left side of his mandible. Extraorally, the mass was extending from zygomatic arch to 5 cm below the lower border of mandible in superior inferior direction. It was associated with displacement of corner of the mouth to the right side, drooling of saliva leading to gross facial asymmetry (Figure 1A–C). The mass was bone hard in consistence, nontender, and not ulcerated. The overlying skin was normal with traditional marks and not fixed to the underlying structures. Intraorally, the mass was solitary, well-defined, bone hard in consistency, and nontender and was extending from behind the left retromolar trigon to the level of second lower molar on the right side. Because of huge size of the lesion, the patient presented with difficulty in chewing and speaking. Marked alveolar asymmetry was noted. The associated teeth were displaced to peripherally, and the tongue was displaced buccally to the right side. No clear vision of oral cavity due to huge size of the lesion that covered the whole oral cavity and protruded extraorally. The oral mucosa that cover the lesion was of normal in color and not ulcerated. There were no palpable cervical lymph nodes in both sides. All vital signs and baseline laboratory blood workup tests were within normal limits.

A CT scan of head and neck revealed a large expansive mass arising from the left mandible causing bone erosion with multiple calcifications. There was erosion of left mandible, loosening of teeth, and unilocular radiolucency with well-defined borders and central multiple opacification (Figure 2A,B). Vallecula, epiglottis, and vocal cords appeared normal. Parapharyngeal and masticator spaces appeared normal. The carotid and jugular vessels appeared normal. Both lobes of thyroid appeared normal. No significant lymph node enlargement identified. Prevertebral soft tissues appeared normal. The cerebral hemispheric parenchyma was normal. Posterior cranial fossa and its contents were normal. Bilateral basal ganglia and thalami appeared normal. Ventricles and cisterns appeared normal. No evidence of midline shift seen. Bony skull vault and skull base appeared normal. CT scan of chest and abdomen were essentially normal.

Based on the clinical findings, history of presenting illness and radiological features, differential diagnoses of OF, ameloblastoma, and fibrous dysplasia were entertained. Histopathology of incisional biopsy from the lesion revealed a benign fibro-osseous tumor made up of diffuse hyperchromatic stromal fibroblastic cell proliferation. The matrix was mineralized with woven and lamellar bone deposits or cementum-like calcifications distributed throughout the lesion (Figure 3). The findings were in favor of mandibular OF with differential diagnoses of osteoblastoma, fibromatosis, desmoplastic fibroma, cementoblastoma, and osteoid osteoma. The patient's family was counseled for surgery, and written informed consent from the legal guardian was obtained.

3 | SURGICAL PROCEDURE

Under general anesthesia, the tumor resection surgery was undertaken. Due to the enormous size of the tumor filling the oral cavity, and compressed nasal apertures, it was difficulty to pass a nasoendotracheal or oral tube

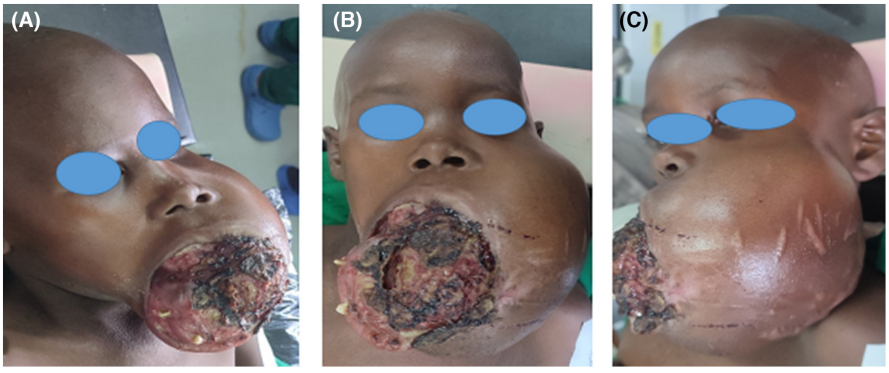

FIGURE 1 Photographs of a patient displaying a massive mandibular tumor causing gross facial asymmetry (A), intraorally the mass was solitary, well-defined, firm in consistency (B), the overlying skin was normal with traditional marks on it and not fixed to underlying structures (C).

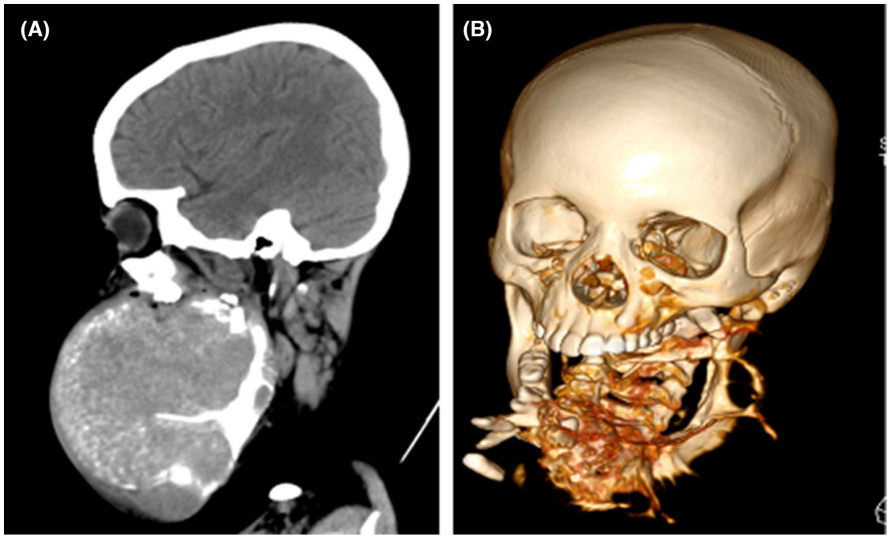

FIGURE 2 CT scan imaging highlighting a huge expansile mass arising from the left mandible with well-defined borders causing bone erosion, loosening and loss of teeth, unilocular radiolucency, and central multiple opacification and calcifications (A), 3D appearance of the mass (B).

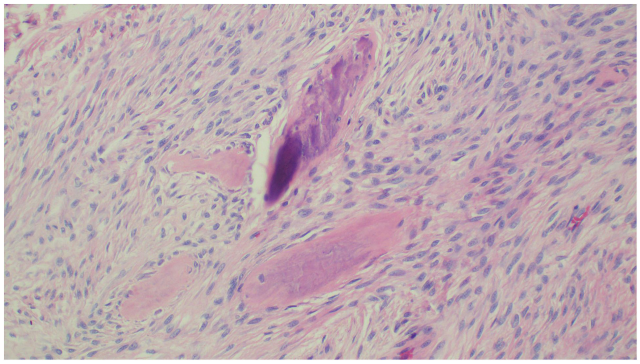

FIGURE 3 Histopathology of ossifying fibroma demonstrating a benign fibro-osseous nature of lesion composed of diffuse hyperchromatic stromal fibroblastic cell proliferation, without atypia, or mitoses. The matrix is mineralized with woven and lamellar bone deposits or cementum-like calcifications distributed throughout the lesion, H & E staining 10×original magnification.

for intubation. Thus, a tracheostomy was fashioned to facilitate endotracheal intubation to administer general anesthesia and maintain the airway. The surgery was performed by a team of experienced experts in the field of oral and maxillofacial surgery. The tumor was managed through an intraoral and extraoral approach. Submandibular incision and resection from the body of mandible between tooth 47 and disarticulation of condyle on left side were made. Soft tissue reconstruction was done primarily both via intra and extra oral approach. Reasonable functional and aesthetics were obtained (Figure 4A,B). Resected specimen (Figure 4C) was submitted for histopathology study, the results concluded it to be OF with negative surgical resection margins. Postsurgery recovery was uneventful (Figure 4D,E). There was no breathing problems noted during postoperative course of the patient. Six month later, the child reported back with good aesthetics and better chewing function at least, and facial appearance (Figure 4F–H). No recurrence has been observed to date. Future plan is to do reconstruction of the jaw with autologous bone and replacement of lost teeth with fixed prosthesis.

4 | DISCUSSION

Our report describes a case of massive mandibular ossifying fibroma (OF) in a 13-year male successfully treated at our institution. OF cases studies with unusual huge sizes are relatively uncommon. A number of them have been reported in the English literature.[1–11] Predominantly, OF affects facial bones, most commonly in the mandible, where it arises from apical to premolars and molars, and superior to the mandibular canal.[4] In the index case, the lesion was located on the body of mandible and clinically presented as a massive jaw bone expansion on buccal and lingual cortical plates. Due to the slow growth, the overlying mucosa or skin and the cortical plates of the bone are invariably intact. However, teeth in proximity with the lesion usually preserve their vitality, pain or paresthesia may occur if pressure is exerted on a neighboring nerve. In our case, despite the huge lesion size, the oral mucosa and skin were not ulcerated, and the patient did not experience paresthesia. These tumors progress slowly and essentially are asymptomatic with initial clinical indication being a thickening of cortical layer, resulting in significant extraoral facial asymmetry.[5,6] However, huge tumors as it was mirrored in the index case, symptoms are present due to mass effect associated with deformity. The radiographic findings vary according to the maturity of the lesion, with an increase in radiopacity associated with an increase in maturity.[2,7] As it was evidenced in our patient, radiological features reveal a well-defined unilocular radiolucency with or without radiopaque foci associated with the degree of calcification.

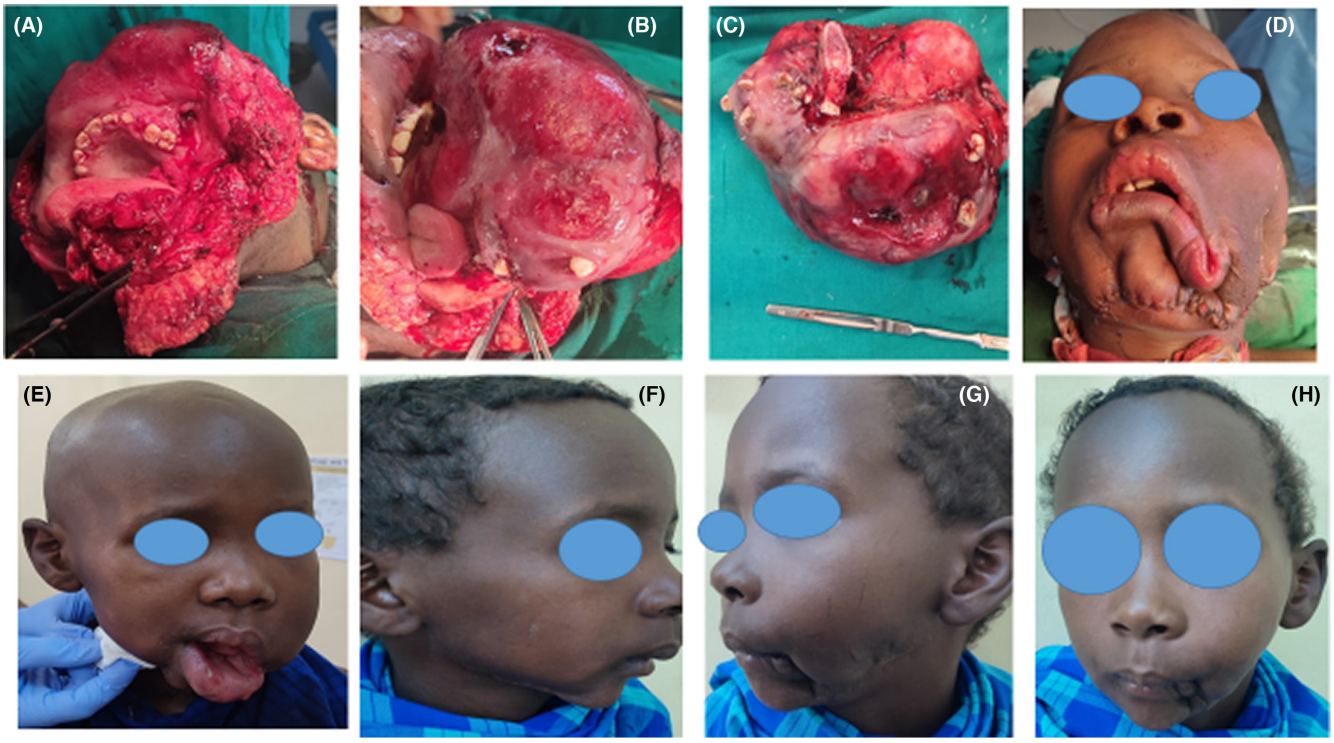

FIGURE 4 Photographs of the patient during surgical resection of the tumor through an intraoral and extraoral approach (A,B), an en bloc resection of the jaw specimen (C), appearance immediately after the surgery (D), on 7th day (E) and 6 months postsurgery (F–H).

As it was the case in our patient, OF continues to enlarge to massive size when left untreated. Principally, OF is managed by conservative surgical excision. However, massive lesions as evidenced in our patient require en bloc surgical removal. Likewise, to reduce the possibility of recurrence for such large lesion, an en bloc resection of the jaw was ideal to our case with the delay reconstruction of the defect. Management of massive lesions tend to be difficult due to its aggressive nature, as well as the high recurrence rates.[8,9] However, radical surgery treatment results in major hard and soft tissue defects that affect not only esthetics and function, but also craniofacial development as well. These defects can be reconstructed by treatments such as free bone grafts in small defects or micro vascularized grafts in large defects.[10,11] The reconstruction of these post-tumor ablative defects is a great challenge, especially in developing patients such as pediatric patients.[5] In our patient due to massive tumor size, bone graft was not done at the same sitting. However, the plan is to do jaw reconstruction using costochondral and iliac bone grafts with titanium metal plates in near future.

5 | CONCLUSION

Gigantic ossifying fibromas as presented in our case is uncommon. It may complicate surgical procedures, postoperative complaints, or prosthetic needs as a result of extraction of teeth associated with the lesion. Patients should be instructed not only about the importance of early detection of the lesion, but also the necessity of routine periodic checkups for avoidance of recurrence or future complications after treatment.

AUTHOR CONTRIBUTIONS

Kanankira A Nnko: Conceptualization; data curation; writing – original draft. **Deogratius S Rwakatema:** Data curation; writing – review and editing. **Jackson M. Mariki:** Data curation; writing – review and editing. **Calvin J Baraka:** Data curation; writing – review and editing. **Sosthenes Damas:** Data curation; writing – review and editing. **Raphael T Pima:** Data curation; writing – review and editing. **Alex Mremi:** Conceptualization; data curation; investigation; writing – original draft; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare that there are no conflicts of interest regarding the publication of this paper.

DATA AVAILABILITY STATEMENT

There is no data generated from this study.

CONSENT

Written informed consent for publication of clinical details and images were obtained from the patient's legal guardians.

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