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Case report

Rare pediatric presentation of aneurysmal bone cyst with trabecular juvenile ossifying fibroma and ossifying fibroma

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ABSTRACT

Aneurysmal bone cyst (ABC) is a benign intraosseous lesion characterized by blood filled spaces of varying sizes associated with a fibroblastic stroma containing multinucleated giant cells, osteoid and woven bone. ABC can present either singly or in association with osseous neoplasms such as ossifying fibroma (OF), giant cell granuloma etc. Juvenile ossifying fibroma has two variants: psammomatoid JOF (PJOF) and trabecular JOF (TJOF). ABC formation in TJOF is very rare in pediatric patients with only three cases reported in literature till date to the best of our knowledge. We hereby report three pediatric cases of ABC, two of which were associated with TJOF and one associated with OF.

1. Introduction

Aneurysmal bone cyst (ABC) was first described as a distinct entity in 1942 when Jaffe and Lichtenstien described two cases of a peculiar blood containing cyst of large size [1]. The World Health Organization defines ABC as a benign intraosseous lesion characterized by blood filled spaces of varying sizes associated with a fibroblastic stroma containing multinucleated giant cells, osteoid and woven bone [2]. The presence of these tumors in facial bones is infrequent with a 2–12% prevalence among all the ABCs of the body [3].

ABC is found more frequently in the mandible than in the maxilla (3:1) with preponderance for the body, ramus and angle of the mandible. It affects young individuals under 20 years of age with no gender predilection [4].

ABC can occur de novo or can be seen secondary to other associated lesions like ossifying fibroma, chondroblastoma, giant cell tumor of the bone, osteoblastoma, giant cell granuloma, fibrous dysplasia, myxofibroma and solitary bone cyst [5].

We report three cases of ABC associated with ossifying fibroma (OF), two of which were associated with typical features of trabecular juvenile ossifying fibroma (TJOF), which is a very rare occurrence. All the three cases were involving pediatric patients in the age group of 10–14 years.

2. Case 1

A 10 year old female reported to the outpatient department with the chief complaint of swelling involving the middle third of the face on the left side. She first noticed a pea sized swelling a year earlier which then rapidly progressed to the current size. There was no history of pain. Extraoral examination revealed a swelling 4 cm × 3 cm in dimension extending from the left ala of nose, 3–4 cm laterally up to the malar prominence. Superoinferiorly, the lesion extended from lateral aspect of the nose to an imaginary line at the level of the corner of the mouth. The margins of the swelling were ill defined. The swelling was hard and non-tender. Intraoral examination revealed labial expansion obliterating the vestibule from permanent left central incisor to deciduous second molar (Fig. 1a). No palatal expansion was observed. A differential diagnosis of fibrous dysplasia and OF was made.

Computed tomography showed an expansile, lobulated lesion with ground glass matrix in left maxilla having an abrupt zone of transition (Fig. 1b). The incisional biopsy report was given as fibro-osseous lesion, since plenty of immature bony trabeculae were seen in a fibrocellular stroma.

The patient underwent complete surgical excision by intraoral approach. The excised tissue was dome shaped measuring 3 cm × 3 cm × 2 cm with irregular superior surface, bony hard in consistency (Fig. 1c). Microscopic examination of the decalcified tissue showed highly cellular stroma composed of proliferating fibroblasts intermingling with areas of immature bone formation with osteoblastic rimming. The trabeculae of bone were irregularly shaped and found to be interconnected with each other in some areas (Fig. 1d). Large cavities filled with erythrocytes was seen

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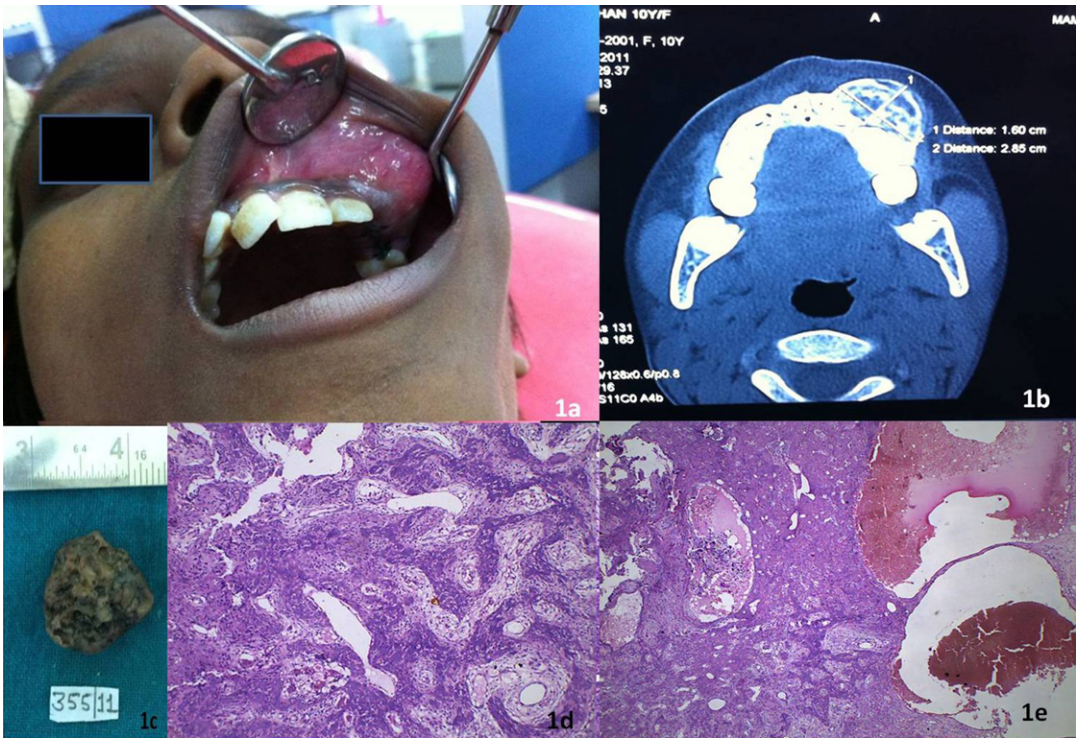

Fig. 1. (a) Intraoral bony mass obliterating the facial vestibule. (b) CT scan showing lobulated lesion with ground glass matrix in left maxilla. (c) Gross specimen – dome shaped with numerous cavities of varying sizes. (d) Photomicrograph showing irregular and immature bony trabeculae (H and E 100×). (e) Photomicrograph showing cavities filled with RBC's associated with giant cells adjacent to area of immature trabeculae (H and E 40×).

adjacent to the areas of ossification. Giant cells were seen in association with the cystic cavity, suggesting the coexistence of an ABC (Fig. 1e) with TJOF. The patient is on regular follow up and no recurrence has been observed with follow up of 1 year and 3 months.

3. Case 2

A 14 year old female reported to the outpatient department with the chief complaint of swelling over the right lower jaw since 3 years. Gross facial asymmetry was apparent on extraoral

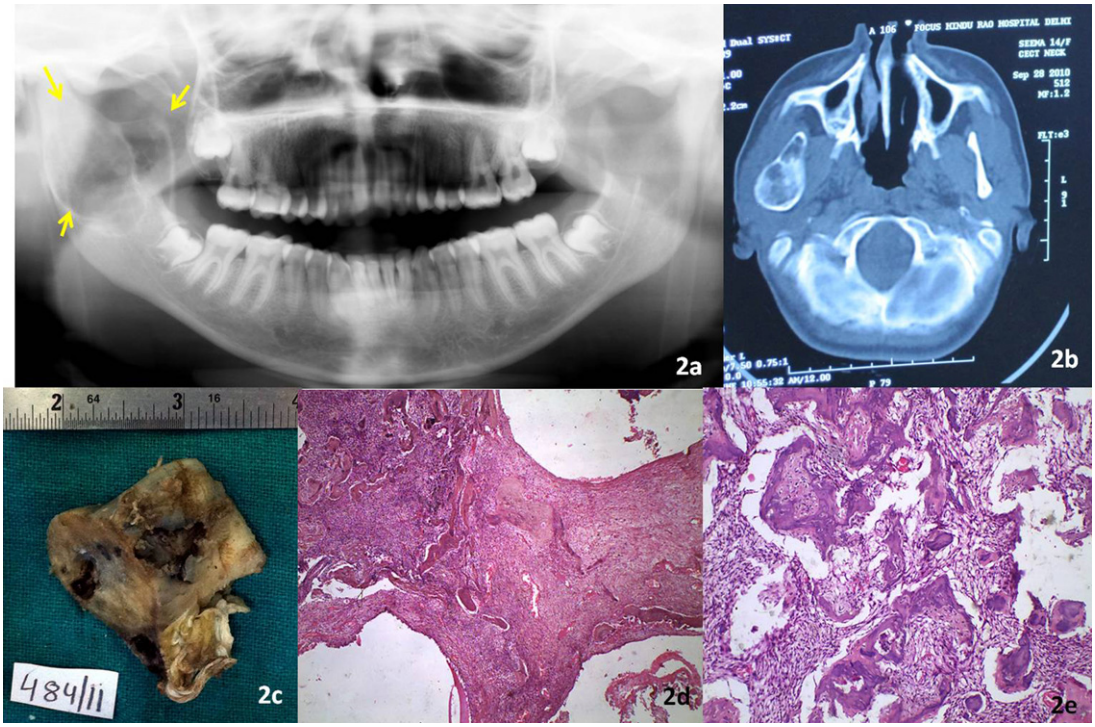

Fig. 2. (a) Panoramic view showing multilocular lesion in the right ramus and condyle. (b) CECT showing expansile destructive osteolytic lesion with internal loculation. (c) Cut surface of gross specimen showing hemorrhagic cavities. (d) Photomicrograph showing large cavities containing erythrocytes (H and E 40×). (e) Photomicrograph showing immature trabeculae of bone in highly cellular stroma (H and E 100×).