Case Report

Nonfamilial cherubism: A case report and review of literature

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Abstract

Cherubism is a rare hereditary developmental condition of the jaws and generally inherited as an autosomal dominant trait. It is also known as familial fibrous dysplasia of the jaws, familial multilocular cystic disease and hereditary fibrous dysplasia of the jaws. The gene for cherubism is mapped to chromosome 4p16.3 may lead to pathologic activation of osteoclasts and disruption of jaw morphogenesis. The lesion usually appears between 2 and 5 years shows a predilection for the mandible and causes a bilateral swelling giving rise to a cherubic chubby appearance. The eosinophilic cuffing of blood vessels appears to be specific for cherubism. The diagnosis is based on clinical, radiographic and histopathologic findings. The purpose of this article is to present a rare case of nonfamilial cherubism as there are very few cases reported and to review the literature with its cone beam computed tomography findings.

Keywords: Eosinophilic cuffing, familial fibrous dysplasia, floating teeth appearance, multilocular cystic disease

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INTRODUCTION

The face is something that we take for granted – more than just another body part; it functions as an important index of identity, a point of reference in our relations with other people.[1] Hence, it is for the dental fraternity to look into its cosmetic purpose in the form of appearance and anatomical variations, the purpose of which could be of great significance in forensic dentistry. Cherubism is a genetic disorder that affects the face and develops early in the childhood. It was first described in 1933 by Jones as "familial multilocular disease of the jaws," but the term "cherubism" was later coined to describe the rounded facial appearance resulting from jaw hypertrophy that resembled cherubs depicted throughout Renaissance art. The condition was initially characterized as familial, but both hereditary and sporadic cases have been described.[2]

It presents as an autosomal dominant trait with 100% penetrance in males and 50%–70% penetrance in females. The typical age of onset being 2–5 years. The jaw lesion progresses gradually until puberty, stabilizes spontaneously and then eventually regresses.[3]

Mutations in the SH3BP2 gene cause cherubism have been identified in about 80% of people with cherubism. The

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SH3BP2 gene provides a command for making a protein whose exact function is unclear. The protein plays a role in transmitting chemical signals within the cell, particularly cells involved in the replacement of old bone tissue with new bone (bone remodeling).

The overactive protein likely causes irritation in the jaw bones and triggers the production of osteoclasts, which are cells that break down the tissue during bone remodeling. An excess of these bone-eating cells contributes to the destruction of bone in the upper and lower jaws.[4]

Perivascular fibrosis leading to mesenchymal disorder and decreased oxygenation is widely accepted theory for its pathogenesis.[5]

The gene underlying cherubism was mapped to chromosome 4p16.3, between D4S 127 and 4p-telomere.[6]

Gene SH3BP2 for cherubism has been mapped to chromosome region 4q16.3. Normally, SH3BP2 encodes the adaptor protein SH3 - domain binding protein. It is required in several intracellular protein tyrosine kinase – dependent signaling pathways during hematopoietic cell differentiation and function. SH3BP2 positively regulates the activity of the transcription factor (nuclear factor of activated T-cells) which is involved in osteoclastogenesis. (Lietman *et al.* 2006). However, the abnormal gene products are formed in cherubism by mutations in the SH3BP2 gene with its location on chromosome 4q16.3 this mutation activates tumor necrosis factor expression in myeloid cells, causing both bone loss and inflammation.[3]

CASE REPORT

A 9-year-old female child reported with the chief complaint of bilateral swelling of the face since 2 years.

Extraoral examination of the face showed bilateral swelling in the mandibular ramus region. The skin over the swelling was normal, intact and freely movable [Figure 1].

Intraorally, the swelling was seen extending from the mandibular canine to the molar-ramus area, bilaterally [Figure 2].

The swelling was painless, slow growing with distinct expansion of the buccal cortex.

Submandibular lymph nodes were bilaterally palpable.

Hematological investigations were performed. Blood profile was within normal limits.

The orthopantomogram (OPG) revealed a bilateral irregular radiolucency in the rami area sparing the condyles [Figure 3 – First visit and Figure 4 – After 2 years of follow-up).

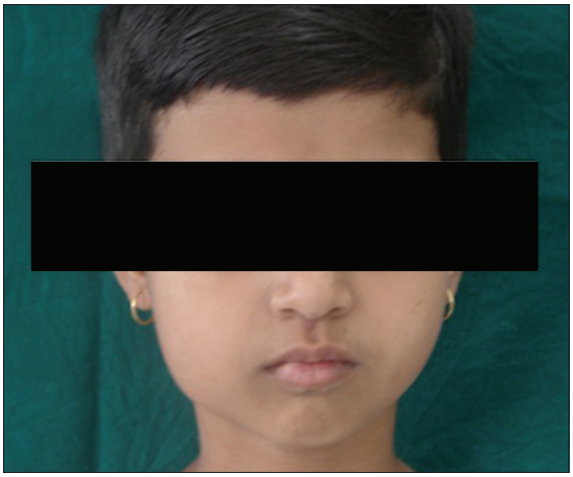

Figure 1: Bilateral swelling seen in the body of the mandible

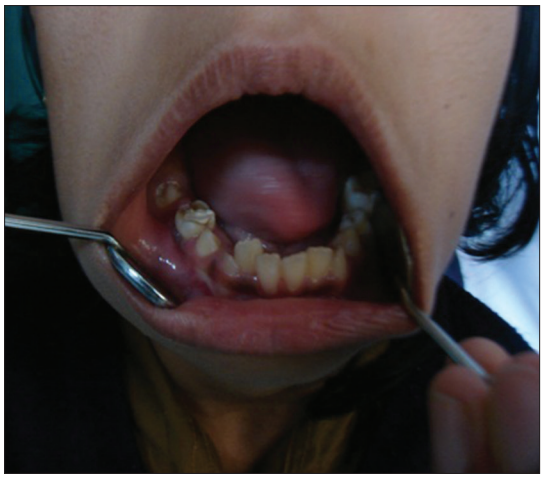

Figure 2: Intraoral findings

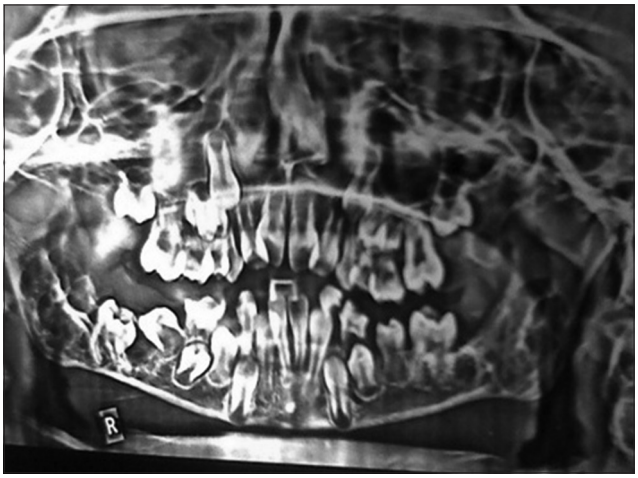

Figure 3: Orthopantomogram revealed a bilateral irregular radiolucency in the rami sparing the condyles

A computerized tomographic scan showed well-defined, multilocular, bilateral and expansile lesion in the rami of the mandible with multiple areas of cortical thinning and discontinuity [Figure 5].

Fine-needle aspiration biopsy was negative and hence incisional biopsy was performed.

Histopathological findings revealed highly cellular fibrous connective tissue [Figure 6].

Multinucleated giant cells resembling osteoclasts were seen in the dense fibrous stroma [Figure 7].

Blood vessels showed evidence of eosinophilic collagen around them described as perivascular cuffing [Figure 8]. Some hemorrhagic foci were also observed. Mixed type of inflammatory cells was present such as lymphocytes and plasma cells.

Peripheral bone also seen along with the inflammatory stroma [Figure 9].

On the basis of clinical, radiographic and histopathological findings, final diagnosis of cherubism was made.

After 2 years of follow-up, the OPG revealed well-defined, multilocular, bilateral and expansile lesion.

The swelling of the mandible also showed increase in size when the patient reported after 2 years [Figure 10]. Hence, it was decided that we take a cone beam computed tomography (CBCT) and check for details.

CBCT scan was taken only after 2 years, and the report showed the following.

Reconstructed panoramic and three-dimensional view of CBCT scan [Figure 11].

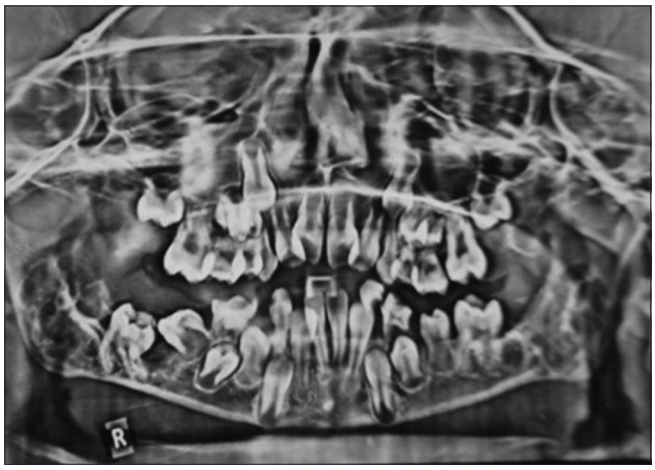

Figure 4: Orthopantomogram after 2 years of follow-up showing bilateral osteolytic lesions in the posterior mandible

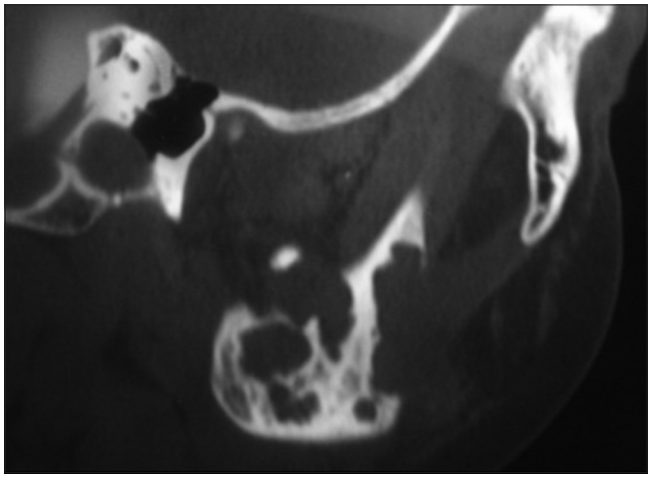

Figure 5: A computerized tomographic scan showed well-defined multilocular, bilateral expansile lesion in the rami of the mandible with multiple areas of cortical thinning and discontinuity

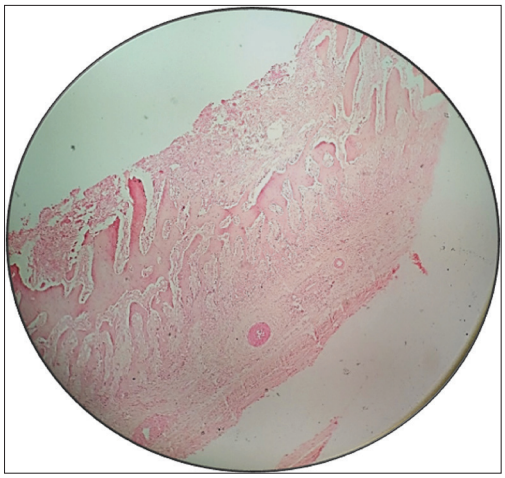

Figure 6: Photomicrograph under ×4 showing peripheral bone, perivascular cuffing, fibrocellular stroma, and multinucleated giant cells

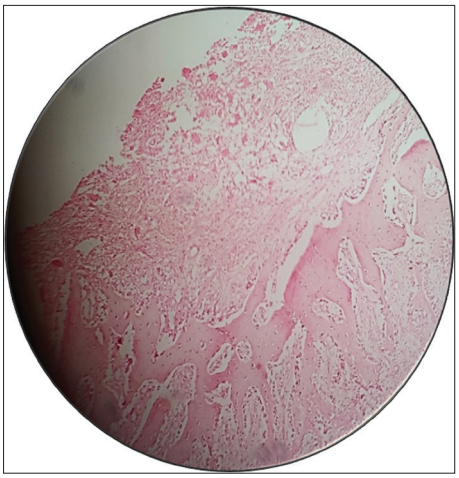

Figure 7: Photomicrograph under ×10 showing peripheral bone, fibrocellular stroma, and multinucleated giant cells

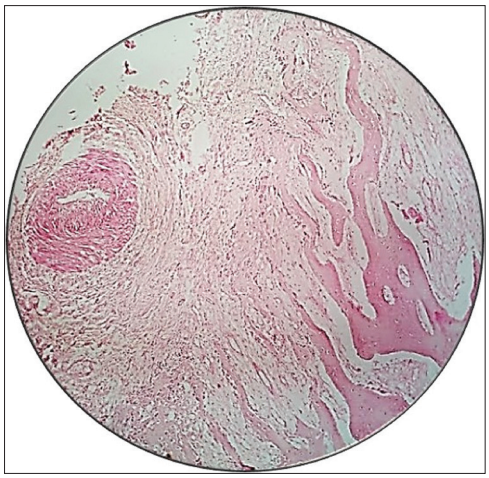

Figure 8: Photomicrograph under ×40 showing perivascular cuffing, fibrocellular stroma, and peripheral bone

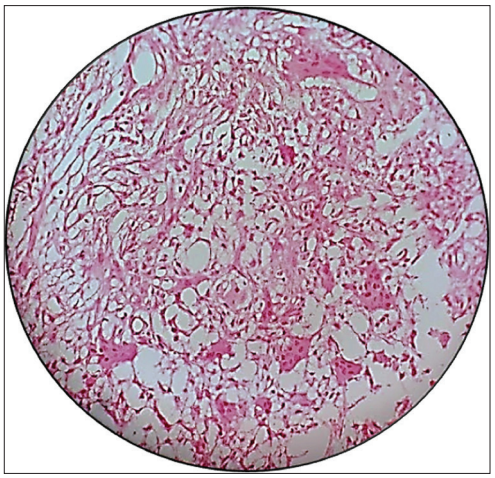

Figure 9: Photomicrograph under ×40 showing multinucleated giant cells in a fibrocellular stroma

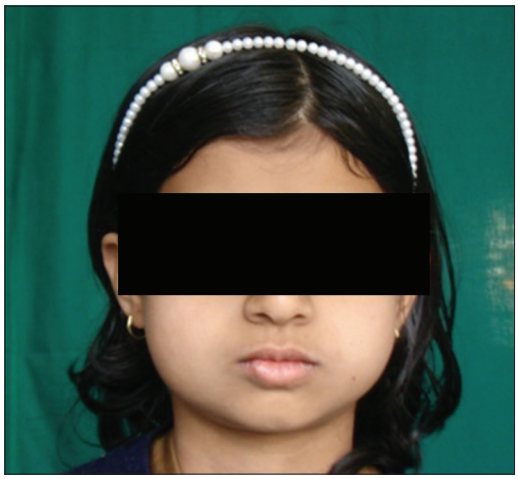

Figure 10: Swelling of the mandible seems to be increased after 2 years of follow-up

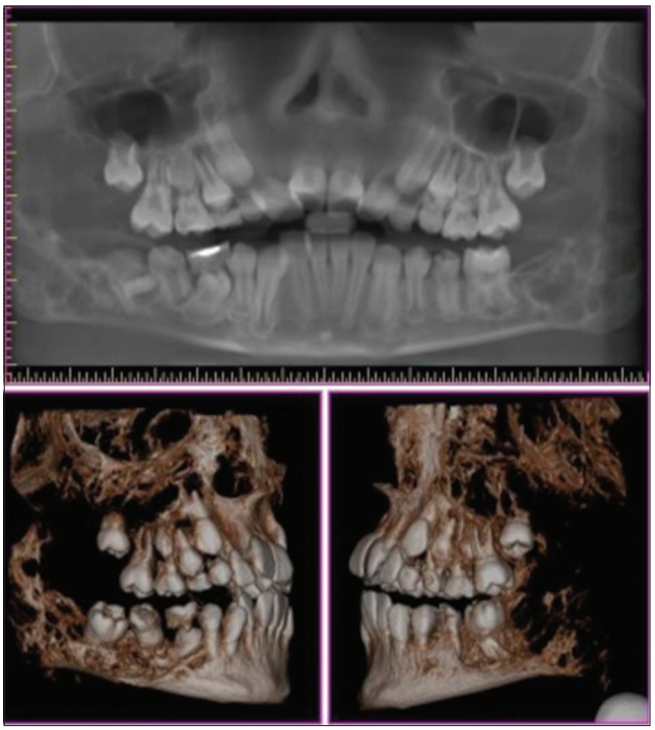

Figure 11: Reconstructed panoramic and three-dimensional view

Characterization of jaw lesion with right mandibular posterior region

- A large roughly oval, osteolytic, expansile lesion with regular outline is evident in the ramus of mandible extending from the apical region of #45 to apical region, distal to posteriorly into ramus
- Superiorly, the lesion extends from anterior border of ramus to the lower border of mandible
- The lesion has grown predominantly buccally with complete resorption of lateral cortex at few sites (black arrows)
- Lesion is circumscribed by thin radiodense bone. Multiple loculation seen
- Inferior alveolar canal cannot be traced [Figure 12].

Characterization of jaw lesion with left mandibular posterior region

- A large roughly oval, osteolytic and expansile lesion with regular outline is evident in the ramus of mandible extending from the apical region of #36 to apical region, distal to posteriorly into ramus
- Superiorly, the lesion extends from anterior border of ramus to the lower border of mandible
- The lesion has grown predominantly buccally with complete resorption of lateral cortex at few sites (black arrows)
- Lesion is circumscribed by thin radiodense bone. Multiple loculation seen
- Inferior alveolar canal cannot be traced [Figure 13].

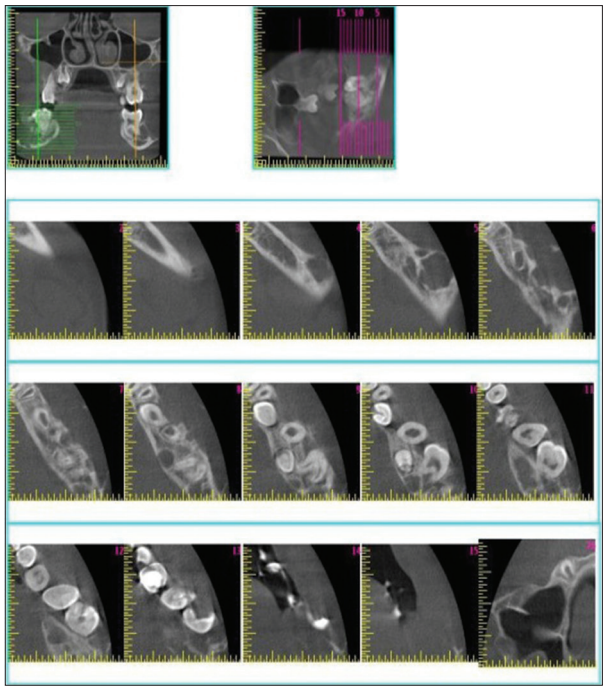

Figure 12: Right mandibular posterior - reconstructed sagittal and 1 mm axial sections at 2 mm interval

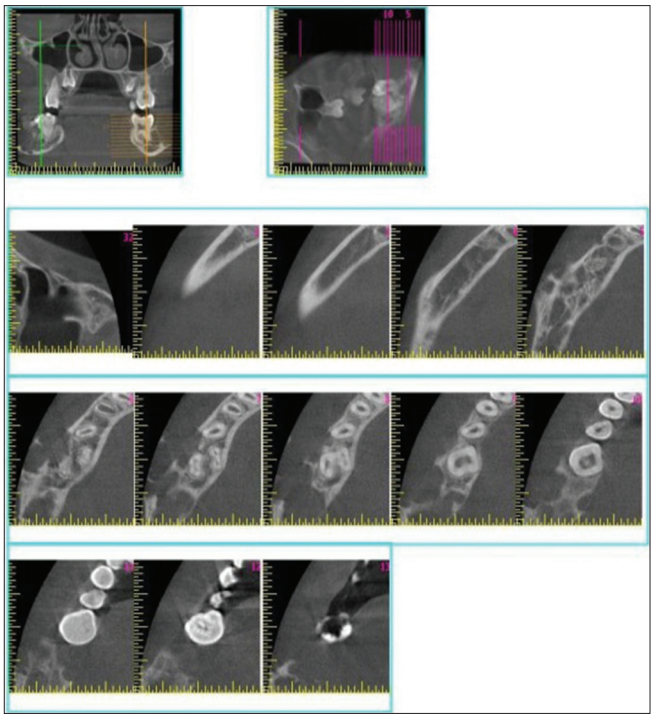

Figure 13: Left mandibular posterior - reconstructed sagittal and 1 mm axial sections at 2 mm interval

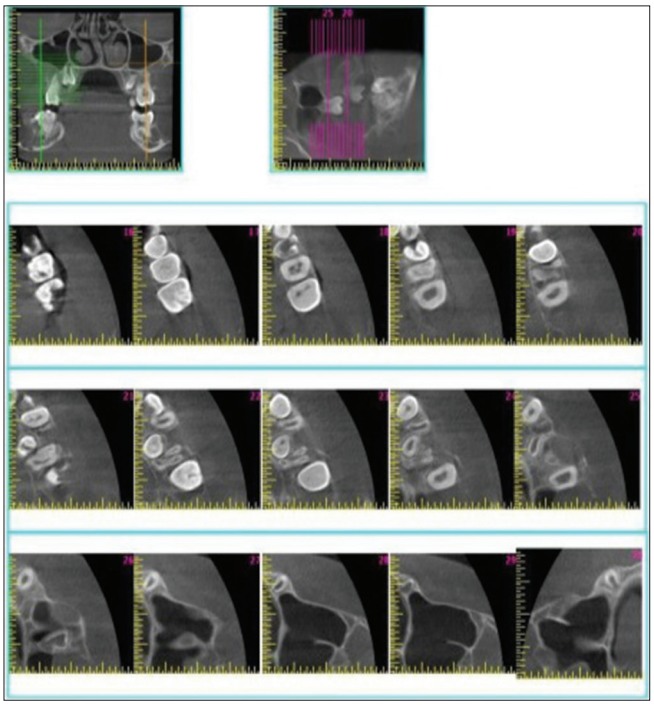

Figure 14: Right maxillary posterior - reconstructed sagittal and 1 mm axial sections at 2 mm interval

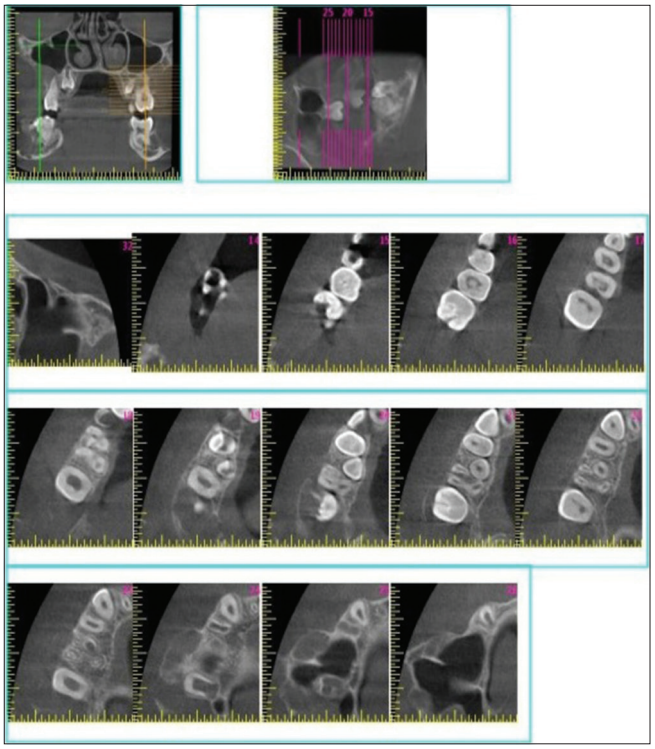

Figure 15: Left maxillary posterior - reconstructed sagittal and 1 mm axial sections at 2 mm interval

Characterization of jaw lesion with right and left maxillary posterior region

- A large roughly oval, osteolytic and expansile lesion with regular outline is evident in the posterior maxilla
- Superiorly the lesion extends into the floor of maxillary sinus

- The lesion has grown predominantly buccally with complete resorption of lateral cortex at few sites (black arrows)
- Lesion is circumscribed by thin radiodense bone. Multiple loculation seen [Figures 14 and 15].

Impression

- Maxillary and mandibular posterior region: Findings are suggestive of a fibro-osseous lesion, cherubism may be considered in the differential diagnosis.

DISCUSSION

Cherubism is a rare hereditary autosomal dominant benign lesion of childhood.[7]

According to the World Health Organization, cherubism belongs to a group of nonneoplastic bone lesions that affect only the jaws. It is also considered a member of the family of fibrous-osseous diseases and some authors refer to this disorder as familial fibrous dysplasia.[7]

Cherubism or multilocular cystic disease of jaws was first recognized as a separate entity in 1933 by William A. Jones in a family with several affected members. He designated the descriptive name "cherubism" because "the full round cheeks and the upward cast of the eyes give the children a peculiarly cherubic appearance." As this name so accurately captured the clinical features of the disease, it became the standard nomenclature.

Cherubism is defined by the appearance of symmetrical, multilocular expansile, radiolucent lesions of the mandible and/or maxilla that typically appears at the age of 2–7 years. Swelling of the submandibular lymph nodes in the early stages contribute to the fullness of the face as is seen in our case also. As the soft fibrous dysplastic tissue in the lesions expands, the protuberant masses can infiltrate the orbital floor and cause the characteristic upward tilting of the eyes exposing the sclera below the iris. Cherubism lesions are limited to the jaws, and in most cases, the dysplastic expansile masses begin to regress with the onset of puberty.[8]

The prevalence in male is 100% when compared with female 50%–70%, i.e., 2:1 ratio.[5] Two forms exist hereditary (familial) and nonhereditary (nonfamilial).[9]

Epidemiology

Cherubism is a very rare disorder with only an estimated 300 cases reported in the literature. Because of its rarity, it is difficult to determine a disease frequency for this disorder. Cherubism affects males and females with equal frequency and has been reported in patients of all racial and ethnic backgrounds.[8]

A molecular pathogenesis of cherubism has been proposed, with the detection of a mutation in the gene encoding SH3 - binding protein 2 (SH3BP2) and possible degradation of the Msx-1 gene which is involved in the regulation of mesenchymal interaction during craniofacial morphogenesis. It is believed that the different clinical manifestations of cherubism are due to the changes secondary to mutations or incomplete penetrance.[10]

According to Hyckel *et al*., cherubism is a location-stable phenomenon found only in the jaws with multiple occurrences. Furthermore, structure-associated process is a very likely link to the pathogenic mechanism. The authors defined cherubism as a genetically determined alteration of tooth germ development. They proposed the molecular model of cherubism pathogenesis which is based on interaction between a disturbed (due to mutation in SH3BP2) parathyroid hormone-related protein (PTHrP) receptor with the Hox gene Msx-1 activity. Thus, the temporal and spatial termination of the clinical symptoms is explained by SH3BP2-dependent signal transduction pathways interfering with jaw morphogenesis. In the cap stage of the second and third molars, a spatial compartmentation does not take place, being necessary for normal dental development. This leads to the dysregulation of mesenchymal bone formation and thus to the development of giant cell granulomas containing osteoclasts.[11]

In 1978, Arnott suggested a grading system for the lesions of cherubism. Cherubism is divided into Grades 1, 2, 3 and 4 depending on location and the severity of involvement of jaws. These classifications are based on the extent of lesion at the time of evaluation. The grade often increases on follow-up examination.[12]

On the basis of extent of involvement, Ramon and Engelberg proposed a grading system for cherubism:

- Grade 1 - Involvement of both mandibular ascending rami
- Grade 2 - Same as Grade 1 plus involvement of both maxillary tuberosities
- Grade 3 - Massive involvement of whole maxilla and mandible, except the condylar processes
- Grade 4 - Same as Grade 3 with the involvement of the floor of the orbits causing orbital compression.[13]

Although extragnathic skeletal involvement is rare, Davis *et al*. reported some rare occurrences in other bones, i.e., ribs, humerus and femur.[14]

The first signs of manifestation of the disease are generally observed at about 2 years of age, followed by accelerated growth from 8 to 9 years and spontaneous interruption after puberty.[8] The patient reported to us was 9 years of age during her first visit and 11 years during her second visit which showed similar accelerated growth. The phenotype ranges from no clinical manifestation to severe mandibular and maxillary overgrowth with respiratory, vision, speech and swallowing problems. Intraorally, it presents as a hard, nontender swelling palpable in the affected area. Submandibular and upper cervical lymphadenopathy are common although reactive regional lymphadenopathy, particularly of the submandibular lymph nodes, usually subsides after 5 years of age.[15]

Clinical or radiographic findings of cherubism are not evident until the age of 14 months to 3 years. Typically, the earlier the lesion appears, the more rapidly it progresses. The progressive swelling of the face, with a marked increase in fullness of cheeks and jaws, is common to all cases and is due to enlargement and expansion of the underlying bony structures, the skin and subcutaneous tissue being normal. The bilateral enlargement of maxilla when present, contributes to cherubic analogy by causing stretching of skin of the cheeks, thus exposing a thin line of sclera causing "eyes raised to heaven" look. This was not reported in our case and is rarely encountered in other case reports also. Frequently, cherubism is accompanied by abnormalities in the configuration of dental arch and dental eruption. In severe cases, tooth resorption occurs. The signs and symptoms of disease depend on the severity of the condition, range from clinically, radiologically undetectable features to grossly deformed jaws, upright palate, respiratory obstruction and impairment of vision and hearing. In few cases, cherubism has been described as being connected with other diseases and conditions such as Noonan's syndrome.[7]

The syndromes associated with cherubism are neurofibromatosis type I, Noonan-like/multiple giant cell lesion syndrome, Ramon syndrome and Jaffe–Campanacci syndrome.[9]

Radiographic features

Cherubism has been described as a subtype of fibrous dysplasia, specifically a hereditary craniofacial fibrous dysplasia because of the radiographic similarities between the conditions.[16]

Radiographically, cherubism is characterized by bilateral, expansive, multilocular, radiolucent lesions, clearly delimited by cortical bone in the mandible. Bone alterations generally start at the angle and ascending ramus of the mandible. The changes may extend to involve the mandibular body, displace the mandibular canal, and in some cases, involve the coronoid process. Maxillary involvement is less frequent and less extensive. In severe cases, infiltration of the orbital bone can occur leading to exacerbated exopthalmos which limits ocular movements.

Hanging and floating teeth along with multiple retained deciduous teeth are also commonly seen. Different types of dental abnormalities ranging from delayed eruption, displacement of teeth to root resorption are known to occur.

Teeth show floating tooth appearance.[17]

Cherubism has been classified according to the severity grades, from the Seward and Hankey system as follows:

- Grade 1: Involvement of the bilateral mandibular molar region and ascending rami, mandible body, or mentis
- Grade 2: Involvement of bilateral maxillary tuberosities as well as the lesion of Grade 1, diffused throughout the mandible
- Grade 3: Massive involvement of the entire maxilla and mandible except the condyles
- Grade 4: Involvement of both jaws with condyles.[2]

The magnetic resonance imaging (MRI) findings of cherubism were first described by Beaman *et al*., who described cherubic lesions as nonspecific, homogeneous, isointensity to skeletal muscle on T1-weighted images and heterogeneous isointensity on fat-suppressed T2-weighted images.[18]

Atalar *et al*. reported that MRI is helpful for determining soft tissue involvement in patients with aggressive cherubism and for assessing the vascular structures preoperatively.[19]

Histopathological features

Microscopically, the lesions showed numerous multinucleated giant cells and vascular spaces which are randomly distributed against a background of highly cellular connective tissue. Histochemical and immunohistochemical characterization of the multinucleated giant cells reveals that these are osteoclasts since they are positive for tartrate-resistant acid phosphatase and express vitronectin receptor.[3] The giant cells are foreign body type with 5–20 nuclei.[2] The cellular stroma contains focal deposits of hemosiderin pigments. Eosinophilic collagen perivascular cuffing can be seen in some cases, and this perivascular hyalinosis is considered pathognomonic for cherubism.[3]

Neville also described the microscopic findings of cherubism as essentially similar to those of isolated giant cell granuloma which seldom permit a specific diagnosis of cherubism in the absence of clinical and radiologic information. The lesional tissue consists of vascular fibrous tissue containing a variable number of multinucleated giant cells which are small and usually aggregated focally. The stroma in cherubism often tends to be more loosely arranged than that seen in giant cell granuloma.

Regezi *et al.* suggest that it resembles central giant cell granuloma and that mature lesions exhibit a large amount of fibrous tissue and fewer giant cells.

Chomette *et al*. described three histologically, immunohistochemically and ultrastructurally distinct stages in cherubism lesions. In the first, osteolytic stage, the authors found numerous round, fusiform and multinucleated giant cells. The giant osteoclast-like cells are tartrate-resistant acid phosphatase positive.

The tissue of the lesions is well vascularized. Fibroblastic cells with fewer giant cells can be found in the periphery of the lesions. Hemosiderin, a breakdown product of hemoglobin and a sign of hemorrhage, is observed in endothelial cells and some surrounding fibroblasts.[8]

The second stage is characterized by proliferative spindle cells, which the authors associated with a reparative stage. Fibroblastic nodules with central vessels dominate the lesion while some osteogenesis can be observed near the cortex of the bone. Newly formed bone matrix and osteoid should be seen.[8]

The third stage is attributed to bone formation with cells staining positive for alkaline phosphatase (presumably differentiating osteoblasts) and high levels of ATPase (presumably associated with mineralizing matrix). The tissue contains more collagen fibers and fewer cells.[8]

With regard to biochemical parameters, serum calcium and phosphorus concentrations and thyroid-stimulating hormone, follicle-stimulating hormone, luteinizing hormone and T4 and T3 hormone levels are usually within normal limits, but alkaline phosphatase levels might be elevated.[19]

Diagnosis

The diagnosis of cherubism is based on patient age, family history, clinical examination, radiographic findings, biochemical analyses and molecular analysis.[8]

Prenatal diagnosis

For pregnancies at increased risk of developing cherubism can be identified by analysis of DNA extracted from fetal cells obtained by amniocentesis usually performed at about 15–18 weeks gestation or chorionic villus sampling at about 10–12 weeks gestation.

SH3BP2 – disease-causing mutation must be identified in the family before prenatal testing can be performed. Preimplantation genetic diagnosis may be available for families, in which the disease-causing mutation has been identified.[20]

Treatment

Treatment of cherubism has not been standardized. Surgical treatment appears to be unnecessary for Grades 1 and 2 cases, in the absence of secondary disturbances. Curettage appears to be necessary in more aggressive cases (Grade 3), to reduce maxillofacial deformity that occurs after puberty. Dukart *et al.* found that surgical curettage and recontouring performed during a period of rapid growth of cherubism lesions not only offer a favorable immediate result but also arrests the active growth of remnant lesions while stimulating bone regeneration. Calcitonin therapy seemed to be effective and resulted in remission of the lesion. The administration of calcitonin was done with nasal spray instead of by subcutaneous injections. The rationale of calcitonin administration is that it inhibits the osteoclastic activity of the giant cells. Radiation therapy is ineffective and contraindicated in view of the risk of osteoradionecrosis, interference with dentofacial growth and development and the effect on future surgical procedure.[20]

Curettage is the surgery of choice. Simple countering of lesions produces good cosmetic appearance. Liposuction has also been used to achieve good contour. Surgery showed that – good immediate results arrested the active growth of remnant. Cherubic lesions and even stimulated bone regeneration. Radiotherapy is contraindicated because of fear of retardation of jaw growth radio osteonecrosis and chances of malignant degeneration. Medical therapy such as calcitonin is theoretically appropriate but without proven result. The recent advancement in the treatment of cherubism is the genetic therapy.[21]

Calcitonin was tried as it inhibits the osteoclastic activity of the giant cells, but with varying results.

Based on the genetic mutations related to the disease, gene therapy is expected to play a role in future treatment.[22]

Gene testing for known mutations in SH3BP2 gene is offered by several commercial reference laboratories and testing on a research basis is available (see GeneTests: http://www.ncbi.nlm.nih.gov/sites/GeneTests/?db=GeneTests website for updated information). Testing for cherubism mutation may help to confirm the diagnosis.

Counseling by a medical geneticist or genetic counselor is recommended if family members are concerned that they may have cherubism. A gene test may resolve the concern if a mutation has been identified in the proband. Siblings of patients should be evaluated by physical examination, panoramic radiographs and genetic testing. Updated information about prenatal testing and preimplantation testing is available at GeneTests (http://www.ncbi.nlm.nih.gov/sites/GeneTests/?db=GeneTests website).[8]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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