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Journal of Dental Specialities

Journal homepage: https://www.jdsits.in/

Case Report

Cherubism: A rare hereditary skeletal dysplasia manifesting as bilateral maxillofacial swellings – A case report and review of the literature

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Abstract

Background: Cherubism is a rare, hereditary skeletal dysplasia, predominantly affecting children. It is characterized by painless, bilateral, symmetrical bony swellings localized to the maxillofacial region, often resulting in a distinctive "cherubic" appearance. The condition typically regresses spontaneously after puberty. Early diagnosis is essential for appropriate management and differentiation from other similar disorders.
Case Description: We report a case of an 11-year-old female child presenting with unilateral bony swelling in the maxillofacial region, which later evolved into bilateral lesions. Radiographic examination revealed bilateral multilocular cystic spaces, a hallmark feature of cherubism. Histopathology confirmed the diagnosis, showing perivascular eosinophilic cuffing around giant cells. The therapeutic approach for this patient was tailored to her specific functional and aesthetic needs, focusing on conservative management as spontaneous regression was expected.
Conclusion: Cherubism is a condition that requires a comprehensive approach for diagnosis, including clinical, radiological, and histopathological evaluation. Clinician awareness is crucial for distinguishing cherubism from other similar disorders. Early intervention can help manage functional and aesthetic concerns while monitoring for spontaneous regression during puberty.

Keywords: Cherubism, Skeletal dysplasia, Maxillofacial, Bilateral lesions, Radiology, Histopathology, Childhood.

Received: 01-03-2025; **Accepted:** 05-08-2025; **Available Online**: 29-09-2025

1. Introduction

Cherubism is a rare, inherited fibro-osseous disorder primarily affecting the maxillofacial skeleton, although it can rarely involve other bones, such as the ribs, humerus, and femur.[1] Characterized by the replacement of bone with fibrous tissue, cherubism presents a distinctive clinical appearance, including symmetrical facial swelling, particularly of the cheeks, and a characteristic "angelic" gaze[2] reminiscent of Renaissance art.[3] This condition is typically inherited as an autosomal dominant trait,[4] though variations in inheritance patterns and associated syndromes have also been reported.[5]

As a benign, self-limiting disease, cherubism often manifests during childhood, with the most notable clinical feature being progressive swelling of the jaws due to excessive bone resorption.[6] Early diagnosis is crucial, particularly for healthcare providers, including dentists, as the condition can be detected through routine panoramic radiographs (OPGs) that reveal characteristic skeletal changes. While cherubism generally resolves on its own with age, understanding its pathophysiology and the process of bone resorption and replacement is important for managing potential complications and ensuring appropriate clinical intervention.

This article highlights a rare case of cherubism presenting with initial unilateral swelling, accompanied by a review of 50 other reported cases, offering valuable insights into the clinical presentation, diagnostic approach, and

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https://doi.org/10.18231/j.jds.96348.1759140597

management of this rare disorder. Through this case study and literature review, we aim to enhance awareness of cherubism, particularly among dental and medical professionals, to facilitate early detection and improve patient outcomes.

2. Case Description

An 11-year-old female patient was referred from a private dental clinic with a chief complaint of a painless, gradually enlarging swelling on the right side of the face, present for five years. According to the patient's grandparents, the swelling was first noticed at the age of six and has progressively increased to its current size. The family history revealed that her parents were in a consanguineous marriage, and her father experienced a similar swelling during his childhood, which resolved spontaneously. Extraoral examination revealed a diffuse swelling measuring approximately 4 × 3 cm on the right lower third of the face. The swelling was roughly oval in shape, firm to palpation, and had ill-defined margins. There were no signs of secondary changes or discoloration of the overlying skin (**Figure 1**). Intraoral findings demonstrated obliteration of the buccal vestibule extending from the region of tooth 44 to the distal aspect of tooth 46. The affected region of the right posterior mandible was non-tender, firm to palpation, and exhibited significant expansion of both the buccal and lingual cortical plates. The dental examination identified Turner's hypoplasia affecting teeth 21 and 36, rotated teeth in regions 15, 25, 35, and 45, and the congenital absence of teeth 17, 27, 37, and 47. Relevant clinical images are provided (**Figure 2** and **3**). Based on the clinical presentation, including the absence of multiple molars and the bucco-lingual cortical plate expansion in the posterior mandibular region, a provisional diagnosis of ameloblastoma was established. Differential diagnoses considered include dentigerous cyst and fibrous dysplasia, given the overlapping clinical features.

2.1. Biochemical investigations:

1. Serum calcium-9.1mg /dl
2. Serum phosphate -4.2mg /dl
3. Parathyroid hormone -13.7pg /ml

2.2. Radiographic findings

Bilateral, distinct multilocular radiolucencies involving the mandibular posterior areas are shown on the orthopantomogram.

2.3. Right side of OPG

A significant multilocular radiolucency is observed extending from the mesial side of tooth 45 posteriorly, involving the body, angle, and ascending ramus of the mandible. The lesion exhibits a classic multilocular "soap-bubble" or "honeycomb" appearance with well-corticated margins. Mainly, the condylar and coronoid processes are spared, and there is no evidence of cortical perforation or pathological fracture.

2.4. Left side of OPG

A multilocular radiolucent lesion is observed extending from the distal aspect of tooth 36, affecting the body and angle of the jaw, despite the lack of clinical swelling. The lesion displays internal septation, consistent with a multilocular architecture, and is similarly well demarcated. The mandibular canal is displaced inferiorly in association with the right-sided lesion, indicating possible cortical expansion. No evidence of periosteal reaction, resorption of adjacent roots, or pathologic tooth mobility is visualized on the radiograph (**Figure 4**).

Histopathological evaluation, along with clinical and biochemical correlation, is essential for a definitive diagnosis.

2.5. Histopathological findings

An incisional biopsy was performed under local anesthesia distal to tooth 46. Histopathological analysis revealed a cellular fibrovascular connective tissue stroma containing loosely arranged spindle-shaped and bipolar fibroblasts. Numerous multinucleated giant cells were observed throughout the lesion, characterized by smaller size and containing 5–8 nuclei. The stroma also exhibited small, thin-walled blood vessels, areas of hemorrhage, and perivascular eosinophilic cuffing in some regions. (**Figure 5** and **6**)

2.6. Grading and classification

The lesion was classified and graded based on the systems proposed by Motamedi (1998) and Raposo-Amaral (2007): (**Table 1**)

1. Grade I: Lesions confined to the mandible without evidence of root resorption.
2. Class 2: Lesions involving the body of the mandible (left side).
3. Class 5: Lesions involving the body and ramus of the mandible (right side).

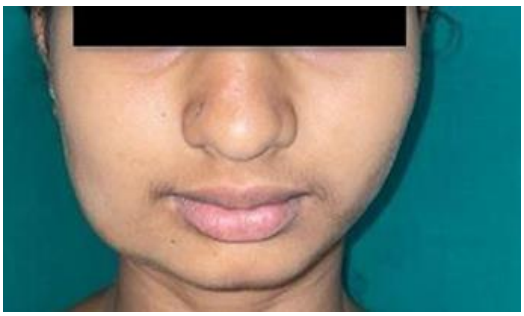

Figure 1: Extra oral-Prominent swelling on the right posterior one third of face

These findings provide critical insights into the extent and nature of the lesion, supporting further clinical and therapeutic considerations.

After correlating the clinical, radiographic, and histopathological findings, along with the grading system, a final diagnosis of cherubism was established.

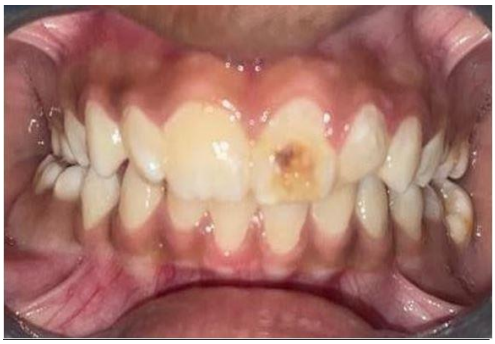

Figure 2: Intra orally–Turner's hypoplasia irt 21, 36

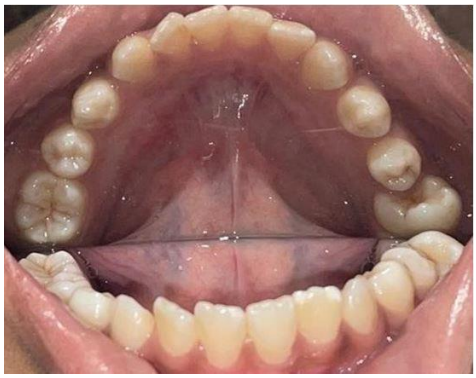

Figure 3: Intra orally-Mesioangular inclination irt 36, mesio-angular rotated tooth irt 35,45

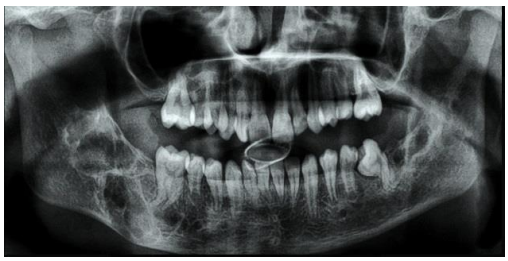

Figure 4: OPG – Bilateral, distinct multilocular radiolucencies involving the right & left mandibular posterior region

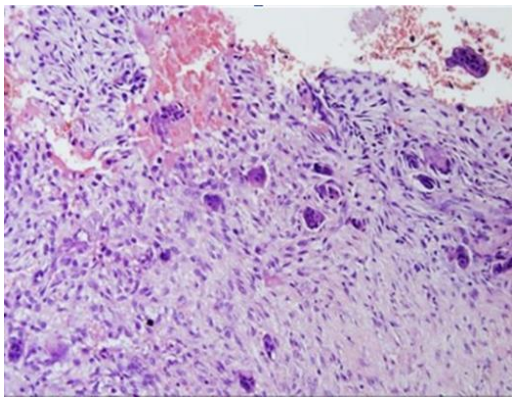

Figure 5: 20x view showing fibro-vascular stroma with giant cells and extravasated RBC's

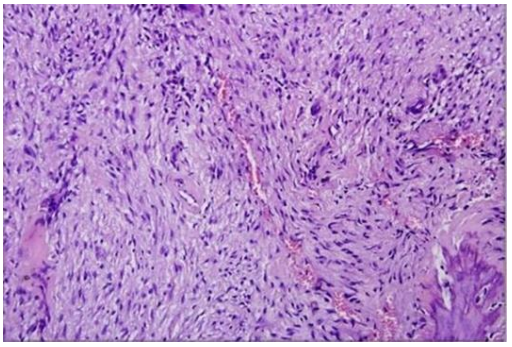

Figure 6: 20x view showing perivascular cuffing

Table 1: Grading system according to raposo-amaral (2007)[26]

Grade I	Mandibular ramus involvement on both sides without evidence of root resorption
Grade II	Maxillary tuberosities and mandibular rami are involved on both sides without evidence of root resorption
Grade III	Mandibular lesions that are aggressive and exhibit root resorption
Grade IV	Lesions exhibiting root resorption that affect the maxilla and mandible
Grade V	Lesions affecting the mandible and maxilla that are uncommon, rapidly expanding, aggressive, and severely deformative are typically seen in younger individuals
Grade VI	Rare adolescent lesions affecting the maxilla, mandible, and involving the orbits that are aggressive, rapidly growing, and severely deformative
Grade VII	Bilateral mandibular and maxillary involvement associated with other dental anomalies or syndromes

3. Discussion

Cherubism is a rare, inherited, fibro-osseous condition primarily affecting the maxillofacial region, with characteristic bilateral, painless, and progressive swelling of the jaws.[7] Described initially as a "familial multilocular cystic disease of the jaws" by Jones in 1933, it was later renamed "cherubism" in 1938[8] due to the distinct "cherubic" appearance it imparts to affected individuals.[9] The condition typically manifests between the ages of 2 and 9 years, with spontaneous regression occurring after puberty. It is more common in males (2:1 ratio) and predominantly affects the mandible, followed by the maxilla.[10] Although the disease generally resolves with age, it requires careful management, particularly in patients with aggressive manifestations. This case report presents an 11-year-old female patient diagnosed with cherubism, emphasizing the clinical, radiographic, and histopathological features that led to the diagnosis, and compares them with similar cases in the literature.

Cherubism is a genetic condition, typically inherited in an autosomal dominant pattern, though rare autosomal

recessive occurrences have also been documented.[11] A key etiological factor is mutations in the SH3BP2 gene, leading to overstimulation of osteoclasts and subsequent bone resorption, contributing to the development of the characteristic cystic lesions seen in cherubism.[12] In the present case, the patient exhibited bilateral maxillofacial swelling, initially unilateral, which later became bilateral. This progression aligns with the typical pattern of cherubism, as seen in similar report.[13]

Family history played an essential role in the diagnosis, with the patient's father also experiencing a similar condition in childhood, which regressed over time. Such familial associations, particularly in consanguineous marriages, support the hereditary nature of the disease.[14] Interestingly, this case demonstrates the rarity of unilateral presentation in cherubism, which has been noted in previous studies but is not as common as bilateral involvement.[15]

Radiographic imaging is a critical diagnostic tool in cherubism. The hallmark feature of cherubism is bilateral, multilocular radiolucencies of the jaws, with a characteristic "floating tooth" appearance due to the resorption of supporting bone.[16] The panoramic radiograph of this patient showed multilocular radiolucency involving the right side of the mandible, extending from the mesial aspect of tooth 45, and similarly, the left side exhibited multilocular radiolucency involving tooth 36. This presentation is consistent with other reports, where multilocular radiolucencies are the most common radiographic finding in cherubism, typically sparing the condyles and coronoid processes.[17]

The floating tooth phenomenon observed in this patient, where teeth appeared to be suspended within the alveolar bone without any supporting structures, is a classic radiological sign of cherubism. This finding aligns with similar cases documented in the literature, where radiographs reveal a lack of normal bony support for the teeth.[18] The absence of secondary changes, such as cortical thinning or radiographic signs of systemic involvement, supports a benign progression in this case, similar to other reported cases of cherubism that demonstrate regressive patterns over time.[11]

Histopathologically, cherubism shares features with other giant cell lesions, such as central giant cell granuloma (CGCG), but exhibits distinctive findings that are critical for diagnosis. The presence of multinucleated giant cells, loose fibrovascular stroma, small blood vessels, and focal perivascular eosinophilic cuffing are classic features of cherubism.[9] In this case, the biopsy revealed a cellular fibrovascular connective tissue with multinucleated giant cells, some containing 5–8 nuclei, consistent with previous reports of cherubism.[19]

The perivascular eosinophilic cuffing, described as a hallmark of cherubism, was also observed in this patient, confirming the diagnosis. This distinctive histopathological feature was first reported by Sarda et al.[20] and has since been cited in numerous studies as a pathognomonic sign of cherubism.[3] Furthermore, the small size of the giant cells in cherubism, compared to those seen in CGCG, further distinguishes cherubism from other giant cell lesions.[12]

Treatment of cherubism is largely conservative due to its self-limiting nature. As the condition typically regresses with age, especially after puberty, no treatment is usually necessary unless functional or aesthetic concerns arise. In this case, the patient's lesion was managed conservatively, with close monitoring of progression. Similar approaches have been reported in the literature,[21] where most cases of cherubism are managed with periodic radiographic follow-up and clinical observation.[22]

In cases where functional or aesthetic impairments occur, more aggressive interventions, such as curettage and surgical contouring of the affected bone, are considered.[23] However, this approach can lead to recurrence or further expansion of lesions, as has been reported in some studies.[24] Calcitonin therapy has been explored in aggressive cases to reduce cystic expansion, although its use remains controversial and not universally adopted.[11]

Surgical management, including resection of large lesions or corrective surgery for cosmetic concerns, may be required in severe cases. However, due to the potential for spontaneous regression, interventions are generally reserved for cases with significant deformities or functional impairments.[25]

Cherubism can be classified based on the extent of involvement, and grading systems, such as those proposed by Raposo-Amaral (2007)[26] and modified in 2014, help in determining the severity and management approach. In the present case, the grading system was applied, with the lesion involving both the body and ramus of the mandible, indicating a higher severity. Such grading systems are valuable for monitoring disease progression and planning interventions, if necessary.

Table 2: A literature review of the PubMed/MEDLINE database was performed from 1990 to 2023, with the keywords cherubism/ childhood disease. The search yielded 50 cases occurring in the mandible, maxilla extending into the maxillary sinus, orbital floor, frontal bone, and the temporal bone of which the lesion affected predominantly males than females with dental abnormalities in 41 cases.

S.No	Year	Author	Age/Sex	Site	F/NF	Dental abnormalities	Provisional diagnosis	Biochemical	Grading	Treatment
1	1991	Fair Cloth et al[27]	23/F	M	F	Agenesis of 3rd molars	NA	N	Fs– Grade I	Genioplasty
			F	M & Mx	F	Agenesis of 2nd & 3rd molars	NA	↑P &ALP	Fs-Grade III	SC &C
2	1993	Ashraf F et al[28]	18/F	M & Mx	NF	ML, deformed, IT	NA	↑ALP	Fs-Grade IV	SC &C
3	1996	Gonjiro Hitomi et al[29]	15/M	M & Mx	F	UT –37&47	AB	↑ALP	Fs-Grade II	Monitored
4	1999	T.Yamaguchi et al[30]	6/M	M & Mx	NF	DT	NA	N	NA	NT
6	2001	M Kozakiewicz et al[31]	6/M	M, Mx &OI	F	RT, DT with RR	Ch,CGCG,GCT	NA	NA	NT
			5/M	M	F	RT, DT, hypodontia	Ch,CGCG,GCT	NA	NA	NT
			5/F	M	F	Impacted teeth	Ch,CGCG,GCT	NA	NA	Osteoplasty
			5/F	M	F	NA	Ch,CGCG,GCT	NA	NA	NT
			6/M	M	F	MT irt31,32,36,37, 41, 42, 46 & 47	Ch,CGCG,GCT	NA	NA	NT
7	2003	Yasar Ozkan et al[32]	9/F	M & Mx	NF	MT irt 11,12,21,22	NA	↑ALP	NA	NT
8	2003	Ravi kiran Ongole et al[33]	7/F	M & Mx	F	RT,DT	NA	N	Mo-grade II, class 1	NT
			14/M	M	F	TT irt 15,16,IT irt 17,47,37	NA	N	Mo-gradeI, class5	NT
9	2004	Luciana Cardoso Fonseca et al[34]	10/F	Mx, M, TB	NF	Absent	NA	N	R&E-Grade-III	NT
10	2005	MF Gomes et al[35]	13/F	M	NF	DT irt 47, 48incomplete formed roots 37 was located Below the 36 that presented R irt 38	NA	N	NA	NT
11	2006	Miguel Peñarrocha et al[36]	20/F	M	NA	DT, Agenesis of 3rd molars	NA	NA	A-Grade I, subtypeII	NT
			14/M	M & Mx	NA	DT, Agenesis of 3rdmolars	NA	NA	A-Grade III, subtype III	NT
			12/F	M & Mx	NA	DT, Agenesis of 3rdmolars	NA	NA	A-Grade II, subtype II	NT

			8/M	M & Mx	NA	DT, Agenesis of 3rdmolars	NA	NA	A-Grade II , subtype III	NT
			10/F	M & Mx	NA	DT, Agenesis of 3rdmolars	NA	NA	A-Grade II, subtype III	NT
			12/M	M	NA	DT, Agenesis of 3rdmolars	NA	NA	A-Grade I, sub type III	NT
			14/F	Mxrysinus	NA	DT, Agenesis of 3rdmolars	NA	NA	A-Grade IV, sub type I	C-3times
12	2007	Flavia S.C. Pontes et al[37]	15/M	Mxrysinus	NF	Tooth agenesis, retained teeth, ectopia	NA	N	NA	Tracheotomy
13	2009	Carmen Mortellaro et al[38]	3/M	M & Mx	F	Ectopic eruption irt 46	NA	NA	A-Grade III	C -M, Wait &see-Mx
			14/M	M & Mx	F	Right side–E, left side-PE	NA	NA	Cannotgrade	C
14	9	Virinder Goyal et al[39]	8/M	M & Mx, Mxry sinus	F	Floating tooth	NA	↑ALP	A-Grade II	NT
15	2011	Divya Mehrotra et al[40]	27/F	M & Mx	NA	Edentulous in both the arches	NA	NA	S&H–Grade III	C-mandibular lesions
			32/M	M & Mx	NA	IT irt Canine	NA	NA	S&H-Grade II	C-mandibular lesions
16	2012	Justyna Wagel et al[41]	5/M	M & Mx	NF	Loss & medial displacement of the deciduous teeth	Ch	N	Marck & Kudryk-Grade III	CT
17	2013	Sudhaa Mani et al[13]	9/M	M	NF	DT irt 21,31, Lingually erupted 41, retained 81, MT irt 16, 46.	Dentigerous cyst	↑ALP,P&↓ Ca	Grade I	NT
18	2013	Virendra Kumar Prajapati et al[3]	8/M	M & Mx	NF	DT,MT	NA	N	NA	NT
19	2014	Manpreet Kaur et al[2]	8/F	M	NA	ACB, DDC irt mandibular primary 2ndmolars	NA	↑ALP	NA	NT
20	2014	Satya Ranjan Misra et al[42]	14/M	M	NA	T irt 16, 26, 36,46, DR irt 12, 22, TC irt 11,21, DI irt 24, RR irt 25,45, complex odontome lingual to 47.	Ch	N	R &A- VII	NT
21	2014	Babita Niranjan et	13/F	M	NF	36-Floating tooth	Ch	↑ALP	A-Grade I	NT

		al[5]								
22	2014	Spyridon Tsodoulos et al[43]	12/F	M&Mx	F	Hypoplastic teeth, ectopic teeth	Ch	↑ALP	NA	NT
23	2014	Rasha Elsha Fey et al[44]	5/M	M & Mx	NA	DT in to the Floor of the orbit.	NA	`NA	S&H-Grade- II.	
24	2015	Sai krishna Degala et al[45]	10/M	M	NF	MT irt 45,46,47	Ch,CGCG,BT	↑ALP&AP	Fs-Grade II	C
25	2015	Arash Mirmohammad sadeghi et al[19]	27/F	M & Mx	NA	NA	NA	N	NA	NT
			26/F	M & Mx	NA	NA	NA	N	NA	NT
			21/M	M, Mx & OI	NA	NA	NA	N	NA	Debulkingand C
26	2015	Mohammad Shakeel et al[46]	10/M	M & Mx	NF	NA	NA	N	R&E -Grade II	NT
27	2015	AhmedF. Al-Omara et al[1]	20/F	M & Mx	NA	Numerous UT & DT	NA	NA	R&E -Grade III	NT
28	2015	Naveen Kumar et al[47]	5/F	M & Mx	F	M–permanent teeth	NA	↑ALP	A-Grade II	NT
29	2016	Abbas Shokr et al[48]	16/F	M & Mx	NF	DT	Ch,CGCG,BT	NA	NA	NT
30	2018	Tyler J Holley et al[49]	5/M	M	F	Absent	CGCG	N	Mo-Grade 1 subclass 5 A	NT
31	2018	W. Sidorowicz et al[50]	11/M	M & Mx	F	Agenesis irt 18, 17, 28, 27, 37, 38, 47,48, DT irt 31, 32, 41, 42	NA	N	S&H-Grade II	NT
32	2020	Sachin G.Ram et al[51]	6/F	M	NF	Agenesis irt 37,47	Ch	NA	Grade I	NT
33	2021	Sabah Nuri Mizel et al[52]	4/M	M, Mx, condyle	NF	MLT	Ch	N	R&E-Grade IV	NT
34	2022	A. Lah Fidi, MD et al[53]	12	M & Mx	NF	NA	Ch	NA	NA	Adviced EX With OS & I
35	2023	Mc Arthur MA et al[54]	6/M	OI	NF	Absent	Ch	NA	NA	SC irt right ramus region

Maxilla-MX, Mandible-M, Zygomatic bone–Zb, Frontal bone-Fb, Orbital involvement–OI, Familial–F, Non-Familial–NF, Ameloblastoma-AB, Cherubism-Ch, Central giant cell granuloma– CGCG, Brown's tumor–BT, Rotated tooth–RT, Displaced tooth -DT, Resorption–R, Dilacerated tooth– DiT, Unerupted tooth– UT, Malaligned/malformed-MLT, Dens invaginatus-DI, Edentulous-E, Partial Edentulous-PE, Taurodontism-T, Hypoplastic teeth-HT, Transposition tooth- TT, Impacted tooth -IT, Talon's cusp -TC, Root resorption- RR, Deep dental caries – DDC, Anterior cross bite– ACB, Not available – NA, Curettage -C, Surgical contouring- SC, Calcitonin therapy – CT, Orthodontic surgery – OS, Implants- I, Fordyce'- F, Ramon and Engelberg's- R&E, Raposo-Amaral –R&A, Arnott-A, Motamedi – Mo, Seward and Hankey- S&H. All the cases exhibited multilocular radiolucencies

4. Conclusion

Cherubism is a rare, hereditary condition that presents significant diagnostic challenges due to its similarity to other giant cell lesions and odontogenic conditions. This case report highlights the importance of a comprehensive diagnostic approach, including clinical examination, radiographic imaging, and histopathological analysis, in establishing an accurate diagnosis. The progressive, self-limiting nature of cherubism typically leads to spontaneous regression, making aggressive treatment unnecessary in most cases. However, ongoing monitoring and careful management are essential for addressing potential functional and aesthetic concerns. This case, along with similar reports in the literature, reinforces the importance of early diagnosis and timely intervention in managing cherubism, ensuring optimal outcomes for affected patients.

5. Clinical Significance

Clinically, cherubism leads to facial deformities, dental malocclusion, and potential functional impairments like difficulty with chewing or speaking. Diagnosis is based on clinical presentation, imaging (panoramic radiographs, CT), and genetic testing. Although often self-limiting, severe cases may require surgical intervention to correct deformities. The condition has psychosocial implications, particularly for children and adolescents, due to aesthetic concerns. A multidisciplinary approach, including genetic counseling, orthodontics, and psychological support, is essential for optimal management and quality of life.

6. Source of Funding

None.

7. Conflict of Interest

None.

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Cite this article: Alapati NS, Mannava C, Manyam R, Pasupuleti S, Sangineedy J, Moturi K. Cherubism: A rare hereditary skeletal dysplasia manifesting as bilateral maxillofacial swellings – A case report and review of the literature. *J Dent Spec*. 2025;13(2):303-311.