CASE REPORT

Central Odontogenic Fibroma of the Mandible in a 9-year-old Child: A Case Report with 24-month Follow-up

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Abstract

Background: Central odontogenic fibroma (COF) is a rarely benign tumor with an incidence of only 0.1% among all odontogenic tumors, which commonly involve the mandible.

Case description: A 9-year-old child reported with asymptomatic bony expansion of the mandible, showing an unilocular radiolucency on an orthopantomogram. The tumor was managed surgically by enucleation, and the surgical stent was placed, which was later modified as a removable functional space maintainer. The patient was followed up for 24 months with the complete healing of periapical radiolucency and the eruption of displaced permanent tooth to the proper position.

Conclusion: This case report discusses the importance of the dentist in assessing and diagnosing COF, especially in the pediatric age group.

Keywords: Case report, Central odontogenic fibroma, Enucleation, Unilocular radiolucency.

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Background

Odontogenic fibromas represent a group of tumors that originate from odontogenic mesenchymal tissue and account for <5% of all odontogenic tumors.[1,2] Broadly, they may be central (intraosseous) or peripheral (extraosseous) in location. Central odontogenic fibroma (COF) is a rare entity that commonly involves the mandible, with an incidence of only 0.1% of all odontogenic tumors.[3,4] The World Health Organization (WHO) defines COF as "a benign fibroblastic neoplasm containing varying amounts of inactive odontogenic epithelium in a relatively mature fibrous stroma."[5] The lesion may evolve from the dental papilla, dental follicle, or the periodontal membrane and, therefore, is invariably related to the coronal or radicular portion of the teeth. Clinically, it presents as a slow-growing asymptomatic expansion of the buccal or lingual cortical bone associated with displacement and mobility of adjacent teeth.[3,6,7] Radiographically, the tumor may present as a unilocular or multilocular radiolucency with well-defined borders resembling other entities like unilocular ameloblastoma, odontogenic cysts, or desmoplastic fibroma.[8] Histologically, Gardner[9] had subdivided COF into a simple type and complex or WHO type, which shows connective tissue stroma with multiple strands or islands of odontogenic epithelium. This reported case is a rare occurrence of COF (WHO type) in the pediatric age group, which was managed by enucleation and the proper guidance of erupting displaced permanent teeth by using an acrylic surgical stent as a functional space maintainer through the follow-up period of 24 months.

Case Description

A 9-year-old healthy female child reported to our Outpatient Department with the chief complaint of swelling in the mandible for the past 6 months. As per the history, the swelling was painless and gradually increased to the present size. The patient underwent extraction of the left primary mandibular in the first molar for the same reason without any associated trauma, pain, or fever. Medical history was nonsignificant, with no history of systemic illness or long-term medication. On extraoral examination, there were no signs or symptoms. Intraorally, a diffuse swelling extending buccally from the distal of the primary mandibular left primary canine to the distal of the primary mandibular second primary molar. On palpation, a firm and nontender cortical expansion was noticed without any eggshell cracking sign. The mucosa overlying the swelling was normal in color and texture with respect to the primary mandibular left first and second molar region with no submandibular or sublingual lymphadenopathy (Fig. 1A). Orthopantomogram (OPG) showed a unilocular radiolucency with well-defined sclerotic borders extending from the left mandibular permanent canine to the mesial of the left mandibular permanent first molar. There was root resorption associated with the left mandibular primary second molar with displaced developing tooth buds of the left mandibular permanent canine, first and second premolars (Fig. 1B). The differential diagnosis based on the clinical and radiological examination were dentigerous cyst,[9,10] radicular cyst,[10,11] odontogenic keratocyst,[12,13] ameloblastoma,[14] odontogenic myxoma,[15] and odontogenic fibroma. The clinical and radiographical features of these conditions presenting as bony expansion of the mandible are described in Table 1.

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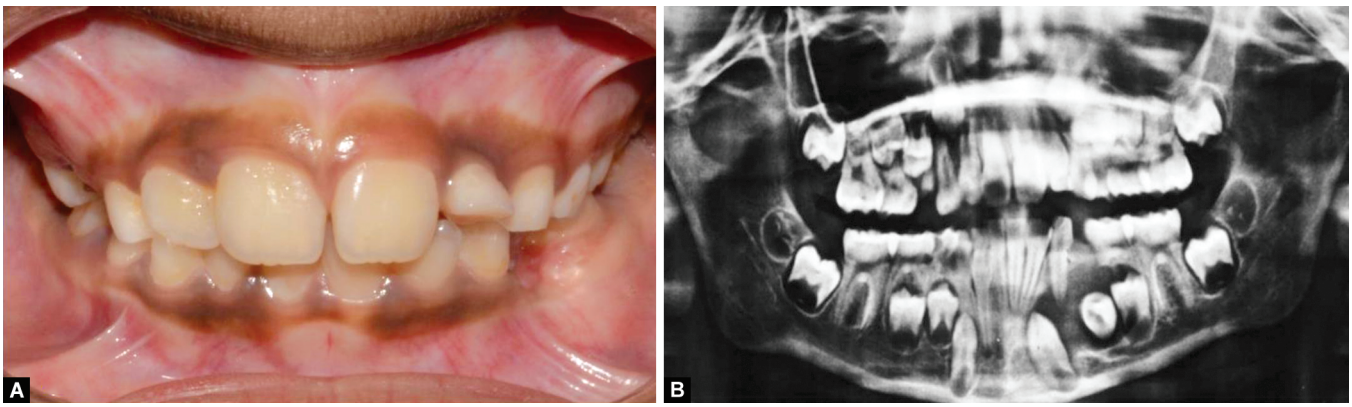


Figs 1A and B: Preoperative photographs: (A) Frontal view showing buccal bone expansion on left mandibular posterior teeth region; (B) Orthopantomogram showing unilocular radiolucency with well-defined borders

Table 1: Differential diagnosis of bony expansion of mandible and their characteristic clinical and radiological features

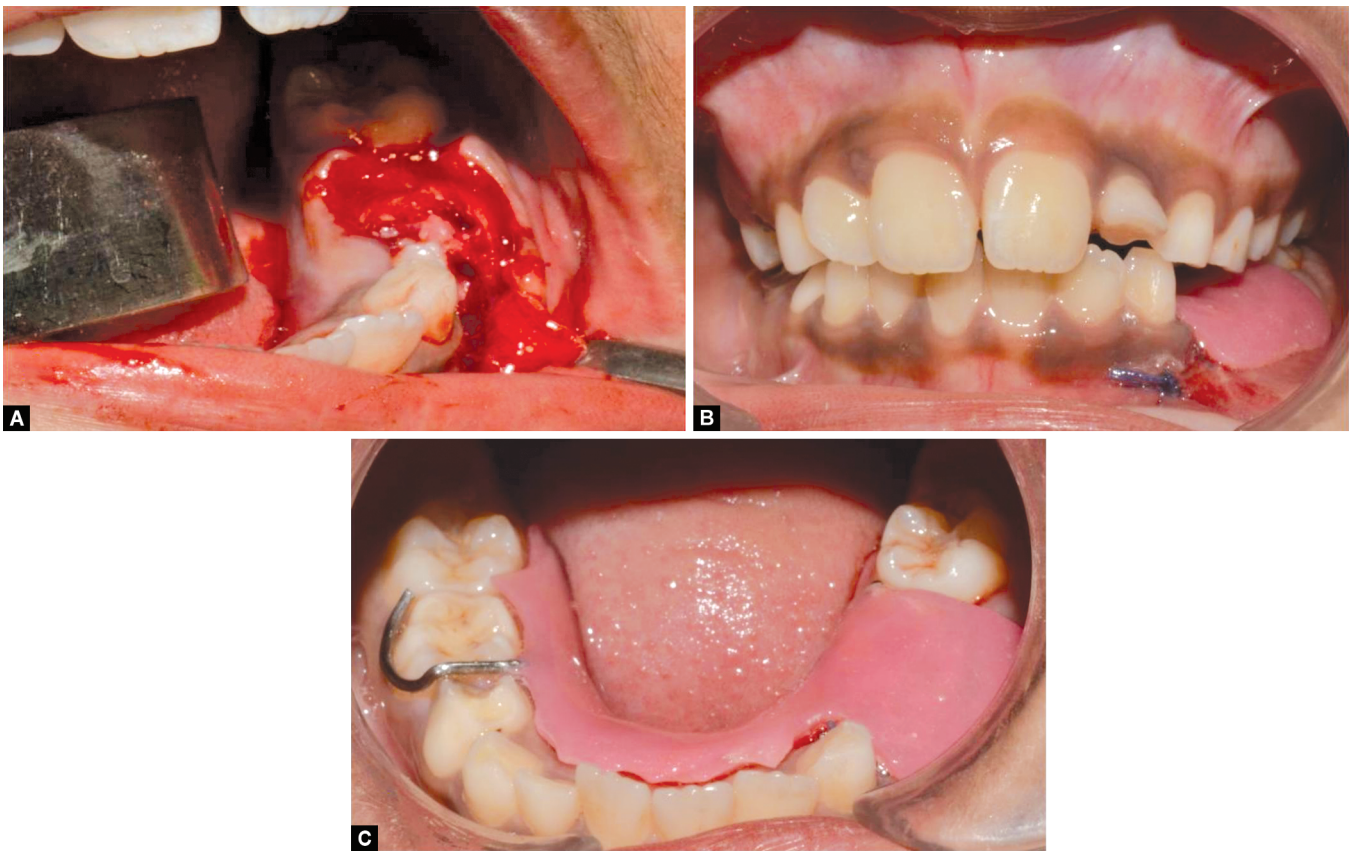
Differential diagnosis	*Clinical presentation*	*Radiological features*
Dentigerous cyst[9,10]	Associated with an unerupted tooth Mostly asymptomatic unless secondarily inflamed Common in the 20s and 30s	Shows unilocular or multilocular well-defined radiolucency with smooth borders Commonly associated with unerupted teeth
Radicular cyst/ periapical cyst[10,11]	Common sequelae of dental caries or inflammatory process of periradicular tissues Sometimes shows sinus formation on overlying mucosa Rare in primary dentition	Presents as pear or round-shaped well-demarcated radiolucency associated with root apex which is <1 cm in size
Odontogenic keratocyst[12,13]	Shows firm and tender swelling with eggshell crackling over the most prominent area Common in younger age groups	Appears as well-demarcated unilocular or multilocular radiolucency Commonly associated with unerupted teeth
Ameloblastoma[14]	Seen as a hard, painless lesion with eggshell cracking in the posterior mandible region Commonly occurs during the 40s and 50s	Shows unilocular or multilocular radiolucent cystic lesion Commonly associated with unerupted tooth and resorption of adjacent tooth
Odontogenic myxoma[15]	Presents as a slow-growing, painless cortical expansion of the mandible with mobile teeth and facial distortion Usually in the age group of 20s or 30s	Appears as well-defined radiolucent multilocules with fine trabecule arranged at right angles

The involved deciduous teeth were extracted, and enucleation followed by curettage of the lesion cavity was done under local anesthesia (lignocaine with 2% adrenaline). A surgical stent made up of self-cure acrylic resin was fabricated to serve as a space maintainer and to maintain its patency (Figs 2A to C). The histopathological examination of the excised tissue revealed predominant fibrocollagenous tissue fragments and mild lymphomononuclear inflammatory infiltrate with focal atypical cells arranged in clusters and small strands (Fig. 3). The atypical cells resembled inactive-looking odontogenic epithelial cells with round to oval nuclear contour, coarse irregularly clumped chromatin, conspicuous nucleoli and a moderate amount of eosinophilic cytoplasm. The pathology report confirmed the diagnosis of a COF.

The patient was followed up every 3 months. At the 6-month follow-up, healing of the operated site was found to be adequate with no clinical or radiographic signs of recurrence. At this time, the surgical stent was modified to act as a removable functional space maintainer (Figs 4A to C). The patient was followed up at regular 6-month intervals to monitor the status of the developing dentition. At the 24-month follow-up visit, the permanent mandibular left first and second premolar had fully erupted into a normal position with no recurrence evident clinically or radiologically (Figs 5A to D).

Discussion

Central odontogenic fibroma (COF) is an extremely rare tumor that has been reported in the literature with considerable ambiguity in terms of epidemiology. Most reports are of individuals ranging from 11 to 66 years of age with a mean of 40 years and a female predominance of 2.8:1.[7] This lesion had been reported to have a mandibular predilection with maxilla:mandible ratio of 1:6.5 particularly for the epithelium rich type.[7] For the proper management of COF, it is essential to differentiate it from other benign odontogenic tumors based on distinct clinical and histological features. In this case, the tumor involved was localized in the mandible, but the age of the patient was 9 years, which is unlike the reported literature suggesting that the odontogenic mesenchymal tissue (dental papilla/dental follicle or the periodontal membrane) around the deciduous teeth might be the etiological factor for odontogenic fibroma in children. To the best of our knowledge, only a handful of case reports exist describing the occurrence of COF in the pediatric age group (Table 2). There were no case reports discussing the importance of surgical stents as functional removable space maintainers, which helped in the proper guidance of erupting permanent teeth.



Figs 2A to C: Intraoperative photographs: (A) Enucleation of the lesion done under LA; (B) A frontal view showing the surgical stent made of self-cure acrylic resin was placed to maintain the patency; (C) Occlusal view

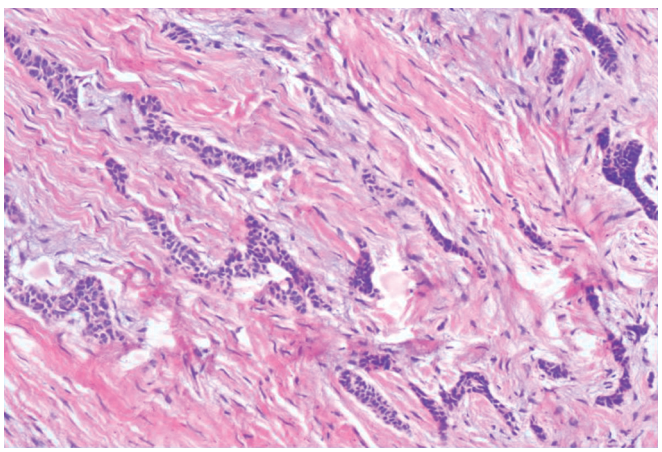
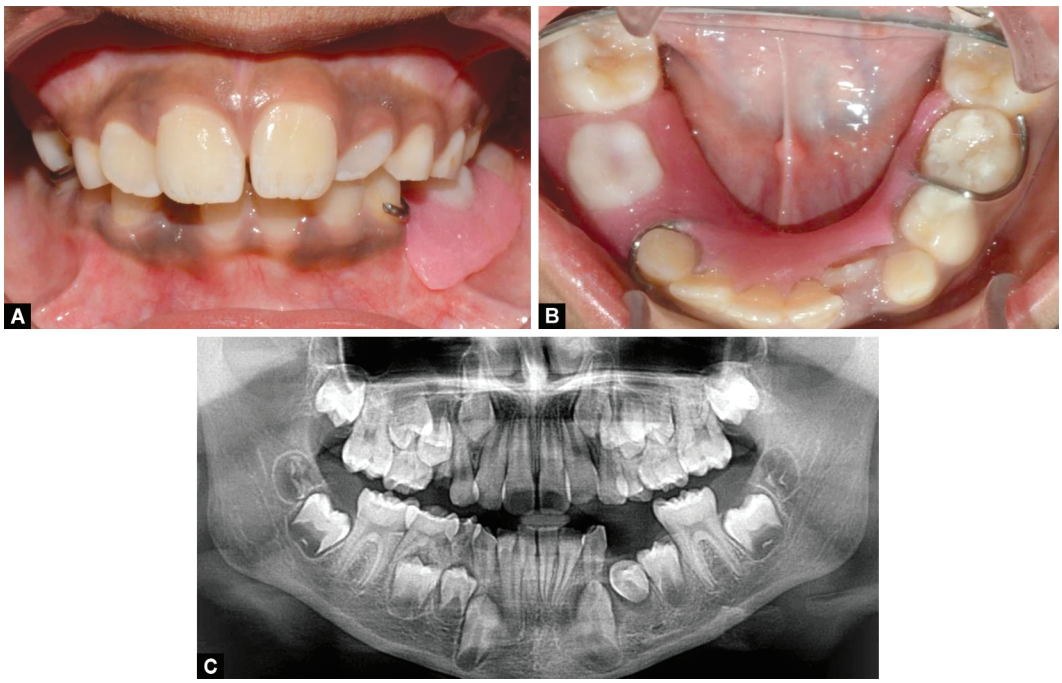


Fig. 3: Histopathological figure

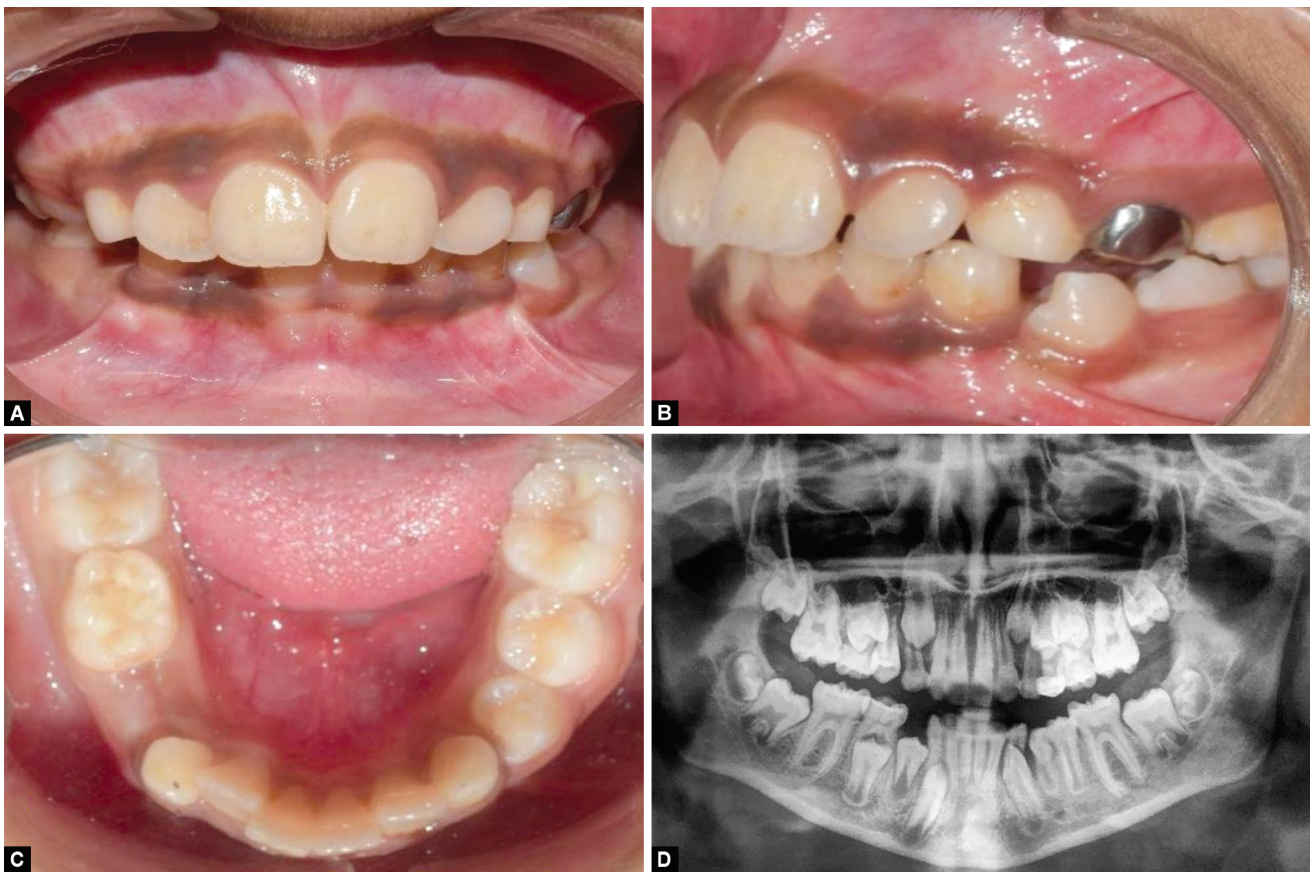
This tumor commonly appears as a slow-growing asymptomatic cortical expansion of the buccal and lingual bone of the maxilla or mandible. These findings were consistent with this case, where the patient complained of slowly developed painless swelling. Radiographically, the lesion presents more commonly as a unilocular radiolucency causing resorption of the adjacent teeth and less frequently as a more aggressive multilocular radiolucent lesion.[8] In this case, the tumor showed a unilocular radiolucency with well-defined borders, which caused root resorption of the involved primary tooth and displacement of the developing permanent teeth.

Histologically, Gardner DG[9] had classified COF into the simple type (epithelium-poor) and the complex or WHO type (epithelium-rich). The simple type is characterized by interspersed mature collagen fibers with many uniformly arranged fibroblasts and small inactive islands of odontogenic epithelium. The complex type exhibits islands of odontogenic epithelium with collagenous fibrous tissue, which are relatively immature, along with the presence of osteoid, dysplastic dentin, or cementum-like material. This case had a histological appearance similar to the epithelial-rich type as it showed fibrocollagenous tissue fragments with inactive odontogenic epithelial cells that had round to oval nuclear contour with irregularly clumped chromatin and conspicuous nucleoli with moderate amount of eosinophilic cytoplasm.

The recommended treatment is conservative management by enucleation followed by curettage of the lesion. Correa Pontes et al.[20] reported in a systematic review of case reports of COF that recurrence is common in lesions involving the anterior maxilla with cortical perforation and uncommon if the lesion occurs in the posterior mandible region. These conclusions were consistent with this case, where the tumor was located in the posterior mandible and had no recurrence at the 24-month follow-up. Further, the use of a surgical stent that acted as a removable functional space maintainer guided the erupting displaced permanent teeth, and it also helped prevent the entry of foreign material into the lesion cavity. This present case report stresses the importance of a surgical stent made of self-cure acrylic, which aided in the successful holistic management of this case beyond the surgical treatment phase.



Figs 4A to C: The 6-month follow-up photograph: (A) Frontal view showing surgical stent modified as a removable functional space maintainer; (B) Occlusal view; (C) OPG showing bone regeneration and normal path of eruption of left permanent canine and premolars



Figs 5A to D: A 24-month follow-up: (A) Frontal view showing left permanent mandibular premolars erupting in the proper position; (B) Lateral view; (C) Occlusal view; (D) OPG shows complete healing with no signs of recurrence and displaced permanent teeth are erupting in the normal position

Table 2: Comprehensive literature review of COF in the pediatric age group

Author/year/country	*Age (in years)/ sex at diagnosis*	*Clinical manifestations*	*Radiological features*	*Histological features*	*Treatment done, and the recurrence rate*
Anbiaee et al., 2015, Iran[16]	4/male	Firm, painless bony swelling at the lower border of the mandible	Unilocular radiolucency with a poorly defined margin in the posterior border of the mandible	The proliferation of ectomesenchymal tissue with some myxoid changes in the stromal cells and osteocyte lacunae in the bony trabecule	Surgical resection with immediate reconstruction with bone graft No recurrence at 6-month follow-up
Bodner 1993, Israel[17]	16/male	Painless expansion of the left maxilla in the canine and premolar region	Well-circumscribed, pear-shaped radiolucency in canine and premolar region	Cellular areas of fibroblasts with small spindle-shaped nuclei, collagen fiber, island, and strands of odontogenic epithelium	Enucleation done Recurrence was not reported
Doyle et al., 1985, United States[18]	16/female	Asymptomatic bony expansion in the left side of the mandible	Unilocular radiolucent lesion between two mandibular premolars	Islands and strands of inactive odontogenic epithelial cells in well-circumscribed fibrous connective tissue	Curettage done No recurrence after 3 years
Rahman et al., 2010, India[19]	13/male	Painless, soft tissue swelling on the right side of the face	Impacted mandibular canine with radiolucency surrounding the tooth	Islands of fibrous to myxoid type of odontogenic epithelium in the cellular connective tissue stroma	Enucleation done Recurrence not reported

Conclusion

Central odontogenic fibroma (COF) is a rare benign odontogenic tumor that commonly occurs in adults, and it is difficult to diagnose because of nonspecific clinical symptoms and radiographic features, especially when it occurs in children. The occurrence of painless swelling for a long period in children might indicate a developing odontogenic fibroma and may be confirmed radiographically. Enucleation of the lesion followed by the curettage of the involved bone is currently recommended treatment with no or less frequent recurrence.

Clinical Significance

The asymptomatic swelling for a long period in children might be due to odontogenic fibroma. Additional considerations for management in a pediatric age group are warranted, such as the placement of a surgical splint following the surgery that would act as a space maintainer and help in guiding the erupting permanent teeth into their proper position.

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