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CASE REPORT

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Continual management for delayed teeth eruption following excision of an intraosseous compound odontoma: a case report

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Abstract

Background: Intraosseous compound odontoma is the most common benign odontogenic tumor formed by odontogenic tissue components differentiating into ameloblasts and odontoblasts, with subsequent deposits producing tooth-like structures (denticles), causing primary teeth retention and impeding permanent teeth eruption in children. Odontoma removal without orthodontic traction is performed to allow for spontaneous eruption of the succedaneous teeth. **Case**: This report was written to describe an 8-year-old girl with an intraosseous compound odontoma obstructing the development of permanent incisor teeth. Because spontaneous eruptions have not yet occurred, the treatment plan subsequently transitioned to manage space for eruption. While this strategy was not considered in the initial plan, we proposed this appliance be preferred treatment to maintain the space until the permanent incisor teeth erupted. **Conclusions**: Odontoma removal not followed by spontaneous eruption of the permanent tooth soon apparently requires further management for space managing.

Keywords

Compound odontoma; Delayed eruption; Impacted; Intraosseous; Space maintainer

1. Introduction

Odontoma is a common benign odontogenic tumor known to develop from epithelial and mesenchymal cells, generating approximately 22% of all cases affecting the jaw. This is an abnormal calcified mass comprising all components of odontogenic tissue, such as enamel, mature dentin, and pulp [1–3]. Furthermore, odontoma is assumed to be a developmental anomaly caused by the formation of completely differentiated epithelial and mesenchymal cells, which produce ameloblasts and odontoblasts [4].

According to World Health Organization (WHO) classification (2022), odontoma is categorized into compound and complex types [5]. Compound odontoma is characterized by the development of dental tissue in the form of small tooth-like structures (denticles) located in the upper incisor area, and commonly found in children aged 6 to 10 years. Meanwhile, complex odontoma consists of a disorganized mass of dental hard tissues without a definite tooth-like structure, primarily located in the area of the first or second molar of the mandible [6]. Odontoma is clinically classified into three types, namely central (intraosseous), peripheral (extraosseous or soft tissue), and erupted odontoma. The most frequent clinical variant is intraosseous (central) odontoma, which mainly affects the anterior maxilla and the mandibular molar area. Peripheral or extraosseous odontoma is a tumor with the same histological characteristics as intraosseous but only occurs in the soft tissues covering the bearing areas of the mandibular and maxillary teeth. However, it is rare and easily misdiagnosed due to being similar to other soft tissue tumors [7].

The etiology of odontoma is not fully understood, with suspected causative factors including infection, local trauma, odontoblast hyperactivity, genetic mutations disrupting tooth formation regulation, heredity, or syndromic abnormalities [8] such as Gardner syndrome [9]. Furthermore, this syndrome, often characterized by multiple intestinal polyposes, osteomas and epidermoid cysts, is associated with dental abnormalities including an increased frequency of multiple odontomas, as well as supernumerary and impacted teeth [10]. Thus, dentists might have an important role to play in the early diagnosis of Gardner syndrome.

Odontoma develops slowly and painlessly and is often associated with the retention of deciduous teeth or delayed eruption of primary and permanent teeth [11]. This tumor causes tooth inclusion, including the canine [12]. This phenomenon is due to the inability of periodontal mechanoreceptors around the unerupted tooth to respond directly to the occlusal load, thus interfering with periodontal ligament proliferation. The proliferation of the periodontal ligament occurs when the periodontal mechanoreceptors undergo structural changes and adaptations during tooth eruption, leading to increased cellular activity and extracellular matrix synthesis. This proliferation exerts a force

that pressures the teeth in an occlusal direction [13]. There is a significant relationship between inclusion of maxillary canines and malocclusions in the presence of skeletal Class III and dental Class II, even if the eruption time has not come [14].

Odontoma is discovered during routine radiographic examinations to evaluate the cause of tooth eruption failure [15], while the treatment includes surgical excision, with recurrence being rare [16]. The provided recommendation suggests waiting for a spontaneous eruption at certain developmental stages, specifically 6 months, or until the complete root formation of impacted or adjacent teeth before planning orthodontic treatment. Despite attempting to preserve the natural potential for tooth eruption, orthodontic treatment remains necessary to maintain or create space in the dental arch [17].

There are few publications regarding delay in spontaneous eruption of permanent teeth after odontoma removal, including treatment in space maintaining. In a previous study, spontaneous eruption of both upper permanent lateral incisor and canines clinically occurred three years after the odontoma removal, while panoramic radiographs showed that the roots of both teeth were much longer but had not closed completely [18]. However, it was not explained how the eruption space could be maintained.

This manuscript aims to report a case of a girl who presented delayed teeth eruption associated to odontoma and its continual management following odontoma removal. Surgical excision was performed, and a 7-month postoperative follow-up showed no recurring tumors. Even though odontoma is a relatively common benign tumor, this case report has clinical significance due to the non-occurrence of spontaneous permanent teeth eruption, suggesting the need for space management treatment to facilitate eruption.

2. Case report

An 8-year-old girl accompanied by the mother was presented to the hospital with a cavity in tooth 64, recurrent pain, and a history of swelling. Her mother also complained that the primary upper right central incisor had not loosened despite the eruption of the permanent tooth next to it. The patient had an overall good health status, with dental class I malocclusion, no history of dental trauma, and no supernumerary teeth. Although teeth 51 and 52 had experienced carious lesions, both teeth had no history of periapical infection. Neither parent had a history of delayed eruption.

The facial profile appeared normal during extraoral examination, but carious lesions were observed on teeth 16, 55, 52, 51, 64, 75, 74 and 84 when assessed intraorally (Fig. 1A–C). The alveolar regions in teeth 51 and 52 became protruded and palpable, with captured panoramic images showing caries extension to the dentin in teeth 16, 55, 52, 51, 75, 74 and 84, while the pulp was affected in teeth 64 and 85. Additionally, a radiopaque lesion similar to the tooth anatomy, causing dental impaction, was detected in the periapical area of tooth 51 (Fig. 1D). A provisional diagnosis of delayed eruption related to odontoma with multiple caries was made based on clinical and radiographic exams. Treatment plans comprised Dental Health Education, tooth extractions (64, 51, 52), odontoma excision surgery, dental endodontic treatment (85), and dental restorations (16, 55, 75, 74, 84), which started with parental consent and adherence to the established schedule.

On the first day of consultation, tooth 64 was extracted to address the main complaint of the patient. During the second appointment, extractions of teeth 51 and 52 were performed, along with surgical odontoma removal, through flap formation and reduction of the bone near the labial (Fig. 2A–C). Local anesthetic was administered to the small-sized treatment area, while the patient remained cooperative and calm. Additionally, the number of odontoma was few and located closer to the labial area (which could be palpated around teeth 51 and 52). Postoperative instructions provided included controlling the open area for 1 week, and the suture wound became fully closed (Fig. 2D). Histopathological examinations of hematoxylin and eosin (H and E stain) sections of the extracted tooth-like structures using a microscope identified normal appearing enamel spacing, dentin, and pulp tissue, as well as the absence of cementum (Fig. 3). This led to the establishment of a definitive diagnosis of intraosseous compound odontoma.

At the third appointment, a vital pulpotomy was performed on tooth 85, followed by Stainless Steel Crown (SSC) placement. In the subsequent two sessions, the Glass Ionomer Cement restoration (GC Fuji IX Gold Label 9) was applied to teeth 16, 55, 75, 74 and 84. The patient was instructed to monitor the spontaneous eruption of tooth 11 for the next 7 months.

During the 7-month follow-up, the mother reported no eruption of the upper right central incisor thereby impacting the appearance of the child. The clinical examination did not reveal any symptoms of eruption for tooth 11 (Fig. 4A–C), confirmed by unchanged tooth positioning on the panoramic image (Fig. 4D). Cone Beam Computed Tomography (CBCT) scans were conducted to verify root position and morphology, presenting incomplete apex formation (Fig. 5).

The treatment plan subsequently transitioned to space management and observation, which was based on clinical examinations, panoramic radiography, and CBCT. The analysis conducted in this report used the Moyers method to measure the mesiodistal width of the four lower incisor teeth, which was found to be 22 mm. Additionally, the length of the upper arch was estimated as 25.5 mm on both the right and left sides. These measurements showed an extra space of 3.5 mm on both sides, prompting the design of a functional space maintainer (Fig. 6). Periodic evaluations were scheduled over the next 6 months to monitor tooth 11 eruption progress in patient.

3. Discussion

This case presents the use of space maintainer as a follow-up management after removal of intraosseous compound odontoma which is not followed by spontaneous eruption of succedaneous teeth shortly.

Intraosseous compound odontoma is a benign tumor often found in close proximity to teeth, causing eruptive disorders and malocclusion. Additionally, the structure resembles the teeth [19] and is commonly found in the anterior maxillary area (81.8%) [20]. The most common location is at the maxillary incisors and canines, while the right side of the mouth has

FIGURE 1. Intra oral and radiographic examination. (A–C) Carious lesions in teeth 16, 55, 52, 51, 64, 75, 74 and 84, retention of teeth 52 and 51. (D) Panoramic figure showed multiple radiopaque lesions with well-defined border surrounded by radiolucent area on the anterior right upper jaw.

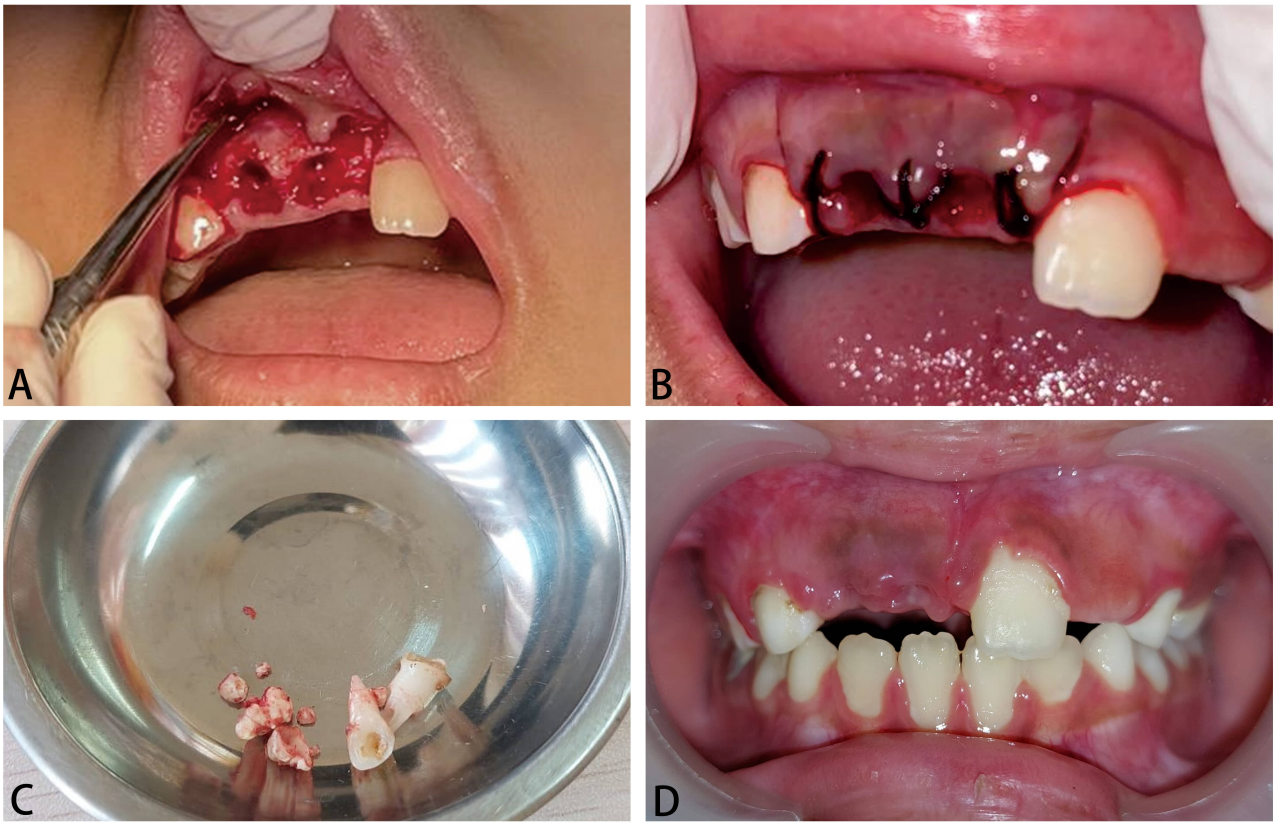

FIGURE 2. The odontoma removal and post odontoma removal control. (A,B) Surgical odontoma removal processing. (C) The specimen collected from the operation site showing numerous miniature tooth-like structures representing compound type of odontoma. (D) The intra oral examination on the 7th day follow-up post-odontoma removal.

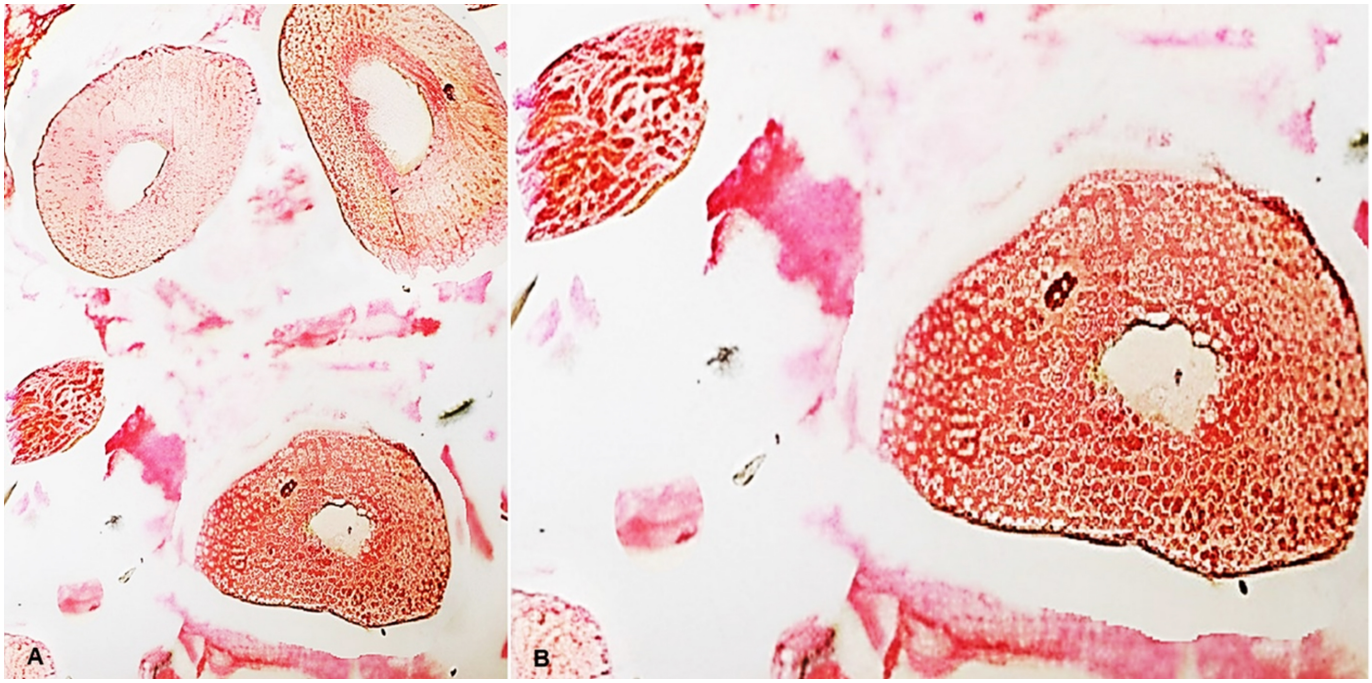

FIGURE 3. The histological examination of compound odontoma showed enamel, dentin and pulp tissue in organized form Hematoxylin Eosin Staining with magnification. (A) 10× and (B) 40×.

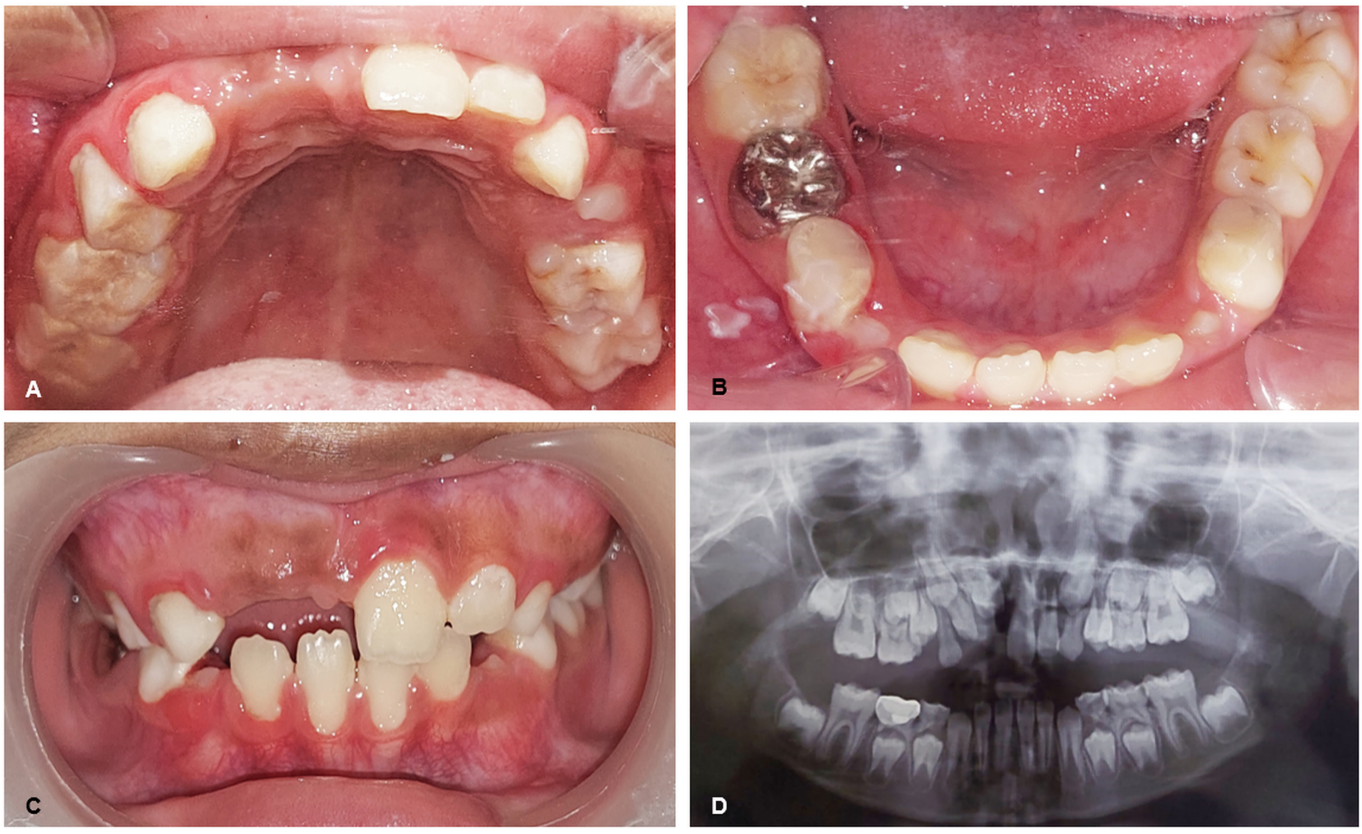

FIGURE 4. The 7 months post-odontoma removal. (A–C) Intraoral examination showed teeth 11 and 12 had not erupted 7 months post operation. (D) Panoramic figure showed all odontoma were removed and the apex formation remained incompleted after 7 months.

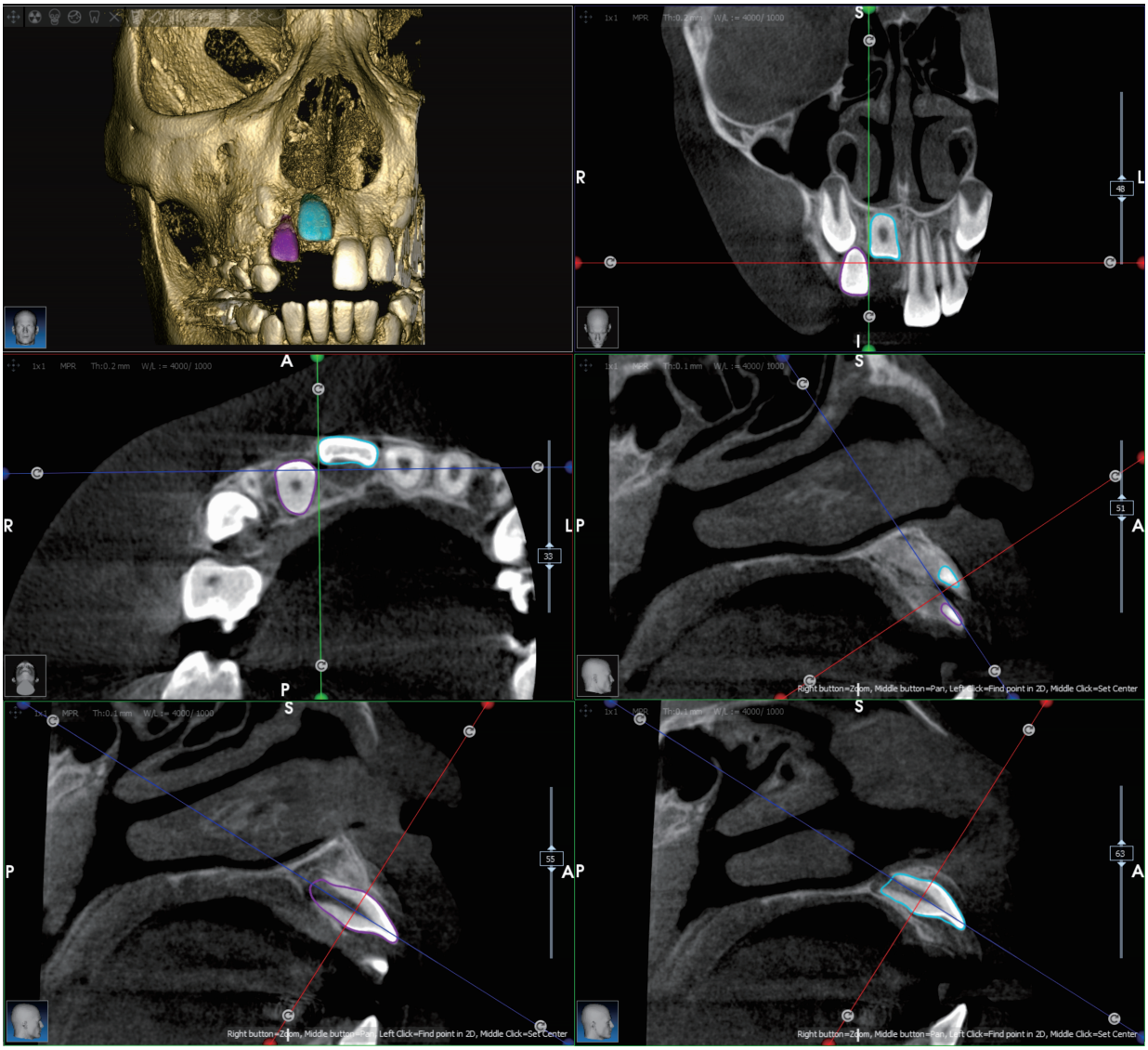

FIGURE 5. Cone Beam Computed Tomography (CBCT) showed tooth 11 with opened apex, which was adjacent to the inferior of dextral nasal base.

FIGURE 6. Functional space maintainer.

the propensity for odontoma development [21]. Furthermore, it is asymptomatic and majorly discovered during routine radiographic examinations, with the tendency to cause tooth eruption difficulties, delayed eruption, malocclusion, or localized swelling [22]. The number of denticles excised in this tumor varies from 4 to 37 [23, 24] and is formed due to local stimulation of the dental lamina [25].

The 8-year-old patient in this report was diagnosed with intraosseous compound odontoma based on clinical, radiographic and histopathological examinations. Intraoral examinations showed signs such as tooth 51 persistence and failure of tooth 11 to erupt at the normal chronological age, while 21 had already erupted. Moreover, the anamnesis presented no history of infection or trauma in the dental area, and there was no family history of the anomaly. Panoramic radiography showed images of several denticles in the periapical area of tooth 51. This supports the theory of compound odontoma being more common in younger people due to higher hyperactivity of the dental lamina [25], but in the current case, the cause of lamina hyperactivity is unknown. Previous investigation presented no family history, trauma, or infection in this case, besides odontoma was detected in the right anterior maxilla, known

as the most common location. Odontoma that obstructs tooth eruption probably causes tooth inclusion. Tooth inclusion, particularly canine inclusion, is at risk of causing malocclusion [26], but in this case, the patient is in a class 1 dental malocclusion. The canine germ, which serves as the occlusion key, is believed to be in a normal position and not affected by the odontoma.

Depending on odontoma size and proximity to the unerupted tooth, it may be removed using conventional intraoral surgery [27], while orthodontic treatment to realign the impacted tooth is not performed regularly [28]. In certain cases, managing space is necessary when waiting for the impacted incisor tooth to develop [29]. A follow-up period of 7 months to 3 years, averaging 20 months is recommended to evaluate odontoma-impacted incisors for spontaneous eruption, without orthodontic treatment. The degree of apical displacement, maintenance of appropriate curvature space, chronological age, root maturation rate, impaction tooth inclination, and impaction tooth root curvature are all factors influencing spontaneous eruption rate after odontoma excision [30]. Therefore, a space maintainer should be used in the first 6 months post-operation during space closure occurrence. A tooth requires 4–5 months to erupt through 1 mm of bone, while space reduction may occur due to adjacent teeth movement from both mesial and distal directions [31].

The treatment options implemented in this case for odontoma removal were surgical excision and extraction of the remaining primary tooth. Eight denticles smaller than the patient teeth were obtained during the excision, and the surgery aimed to facilitate the spontaneous eruption of the impacted incisor while minimizing the need for orthodontic treatment. Seven months after surgery, clinical examinations showed that the upper right central incisor had not erupted and the post-extraction space remained preserved, while CBCT confirmed the apex was still open. According to CBCT examination, the incisal surface of the impacted incisor was at the same level as the Cemento Enamel Junction of the left upper central incisor tooth, and the apical part was marked by the inferior medial part of the dextra nasal base. The impacted teeth had the tendency of spontaneous eruption, but it might take longer, leading to the use of a maintainer, serving as are partial dental prosthesis for space management. Orthodontic traction was not considered a viable treatment option due to incomplete (one-half) root formation of tooth 11, since the traction produced forces elicited an exaggerated response from pulp tissues. Particularly, pulp tissues often experienced vacuolization and circulatory disturbances during orthodontic treatment, which could initiate a series of biological reactions resulting in root resorption [32]. According to Wan *et al*. [32] (2023), tooth roots that developed in the mixed dentition period showed no impaired increases in length and volume after orthodontic treatment, when performed on immature teeth with two-thirds root formation.

The removable space maintainer design often covers a large area of oral tissue which occasionally causes soft tissue irritation and discomfort. However, the advantages of this removable appliance include ease of cleaning and cost-effectiveness, as well as the capability to improve speech, swallowing, and physical appearance [31, 33]. The transmission of acrylic base pressure from the device can influence the process of alveolar bone resorption. Production of greater pressure transmission value, leads to faster occurrence of the alveolar bone resorption [34], thereby facilitating permanent teeth eruption due to alveolar bone resorption caused by the transmission of base plate pressure in the edentulous area.

The use of a removable space maintainer tends to be a viable treatment option provided there is proper counseling and strong motivation from both the patient and parents. Additionally, a follow-up is planned for the next 6 months to monitor the eruption progress of the impacted incisor. Pending the completion of this case report, the upper incisor will remain under observation for spontaneous eruption. This may limit the generalizability of the result. A longer follow-up period may elucidate other eruption issues with dentition as the child grow.

4. Conclusions

The presence of primary teeth persistence and delayed permanent teeth eruption due to odontoma must be promptly followed up with removal surgery. This removal can be performed without continued orthodontic traction aimed at the spontaneous eruption of permanent teeth along with complete root formation. Odontoma removal not followed by spontaneous eruption of the permanent teeth soon apparently requires further management to maintain eruption space.

AVAILABILITY OF DATA AND MATERIALS

All data generated or analyzed during this report are included in the submitted article.

AUTHOR CONTRIBUTIONS

UW and ZNZ—designed the treatment plan expertly. UW and YM—conducted the treatment. SS—acted as supervisor and provided valuable advices. EN and ND—performed the data analysis. UW and TDU—composed the manuscript. All authors thoroughly read and approved the final manuscript.

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This research was conducted under ethical approval recommendation from Health Research Ethics Committee, Dental Hospital of Hasanuddin University with the reference number is 0177/PL.09/KEPK FKG-RSGM UNHAS/2023. We have obtained parental consent to treat this case and parental consent was documented in ethical record.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

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