ABSTRACT

Hypodontia is quite uncommon in the primary dentition in which lateral incisors are the most commonly missing primary teeth in the patients affected. Studies have shown that hypodontia in the primary dentition involving only maxillary canine is rare. One case of hypodontia in the primary dentition involving only maxillary canines is presented here with impacted permanent canines. In children with congenitally missing maxillary canines, the permanent dentitions may show diverse anomalies in tooth numbers, ranging from hypodontia to hyperdontia. Impaction of maxillary canines is a frequently encountered clinical problem which usually requires an interdisciplinary approach. Hence, it seems worthwhile to focus on the means of early diagnosis and treatment of this clinical situation.

Keywords: Hypodontia, Agenesis, Primary maxillary canine, Impacted canines.

CASE REPORT

An 11-year-old Indian male patient was brought to the faculty practice clinic with a complaint of missing primary upper canines and malaligned lower teeth with no previous dental experience. His medical history was unremarkable in that there was no history of supernumerary or congenitally missing teeth in his family. On intraoral examination, it was found that the patient was in the late mixed dentition period. The teeth present were permanent maxillary and mandibular incisors, first molars, lower canines, primary upper and lower molars. Both maxillary primary canines were found to be congenitally missing as confirmed by the parents. Both the parents and the kid were asked repeatedly regarding any extraction of deciduous maxillary canine or exfoliation during permanent lateral eruption. The clinical picture clearly showed no crowding in upper arch; however, there was crowding in lower arch but there was canine erupted in place—not of much concern. Mandibular primary first molars were in exfoliating stage (Fig. 2). An orthopantomograph (OPG) of permanent canine displacement include space deficiency, disturbances in tooth eruption sequence, trauma, retention of primary canine, premature root closure, rotation of tooth buds as well as localized pathological lesions (cysts, odontomas). Here, we are reporting a case of congenitally missing two maxillary primary canines with impacted successors.

INTRODUCTION

Hypodontia is the congenital absence of one or more teeth in the dentition. It is a common anomaly that affects 20% of the population. Several studies have been reported with occurrence rate of 0.5% or less for agenesis, thus found to be rare anomaly in the primary dentition. Lateral incisors are the most commonly missing teeth in the primary dentition. It is seen that maxillary lateral incisors were more commonly found missing than mandibular lateral incisors. Primary etiological causes of permanent canine displacement include space deficiency, disturbances in tooth eruption sequence, trauma, retention of primary canine, premature root closure, rotation of tooth buds as well as localized pathological lesions (cysts, odontomas). Here, we are reporting a case of congenitally missing two maxillary primary canines with impacted successors.
taken for checking dental anomalies showed a mixed dentition with developing permanent teeth (third molars excluded). Orthopantomograph showed the presence of impacted maxillary permanent canines, overlapping the root apices of lateral incisors and first premolars (Fig. 3).

We could have expected exfoliation of deciduous maxillary canines if premolars were erupted but, in this case, deciduous molars were still in place and there was no crowding. Crowding was seen in lower arch but not upper and our case report was related to upper arch only. If deciduous maxillary canines were exfoliated during eruption of permanent lateral incisors there would have been some space distal to permanent lateral incisors.

The patient was advised orthodontic treatment after extraction of primary upper and lower molars which is an effective management option for children with impacted maxillary canines.

**DISCUSSION**

Recent reports have shown that the prevalence of hypodontia in the permanent dentition (third molars excluded) is about 4.5 to 7.4% in Caucasians, and the most commonly missing tooth is the mandibular second premolar. Compared with the situation in the permanent dentition, hypodontia in the primary dentition is rare and its prevalence has been found to be 0.5% or less in several studies.\(^5\) The incidence of hypodontia in the primary dentition varies ranging from 0.5% among the Swedish children to 1.0% among the Caucasians. However, a higher incidence of 5.0% in the primary dentition of Japanese children has been reported.\(^6\)

Most studies showed a higher prevalence of hypodontia in females as was evident in the meta-analysis, which demonstrated a male to female prevalence ratio of 1:1.4 but the present case reported is of a male patient.\(^1,7\) The maxillary lateral incisor was the most frequently missing primary tooth, followed by the maxillary central incisor and first primary molar. This case is interesting for several reasons: firstly, there are very few cases reported in the literature regarding congenital absence of only maxillary primary canines, which in itself is very rare. Secondly, radiographic evidence of this case showed the presence of the maxillary permanent canines which are impacted.

Studies have also shown that agenesis of a primary tooth was often followed by agenesis of the permanent successor.\(^5\) The current case showed the radiographic evidence of maxillary permanent canines which were impacted. Daugaard-Jensen et al had found that agenesis of a primary incisor was followed by agenesis of the permanent successor in 86% of cases.\(^6,8\) Witkop, in 1962, reported that 11 of 273 individuals with congenitally missing primary teeth also had congenitally missing permanent teeth. Thus, our case is not simulating to the literature studies. Early detection of ectopic maxillary canine teeth is important for carrying out an optimum interceptive treatment. However, late recognition and referral is a common problem.\(^7,9\)

Both environmental and genetic factors can cause a failure of tooth development. Environmental causes may be due to the failure of tooth bud cell proliferation from the dental lamina which may be due to an infection (e.g. rubella, osteomyelitis), trauma, drugs (e.g. thalidomide), chemotherapy or radiotherapy at a younger age.\(^10\) In addition, Kjaer et al suggested that the pathogenesis of mandibular tooth agenesis was related to disturbances in nerve tissue, oral mucosa and supporting tissues; all of which interact during odontogenesis.\(^11\) Genetic causes may be associated with mutation of MSX1 and PAX9 genes. Tooth agenesis is probably caused by several independent defective genes, acting alone or in combination with others, which eventually lead to specific phenotypes. Although tooth agenesis is associated with more than 49 syndromes, several case reports describe nonsyndromic forms that are either sporadic or familial in nature. Hypodontia is also often seen in syndromes associated with lip/alveolus with or without cleft palate.\(^12,13\)
The use of panoramic radiography along with a thorough clinical examination is recommended in detecting or confirming dental development. Methods of diagnosis that may allow for early detection and prevention should include a family history, visual and tactile clinical examinations and radiographic assessment. When the condition is identified early, extraction of the maxillary deciduous molars may allow the impacted canines to correct their paths of eruption and erupt into the mouth in relatively good alignment. This interceptive treatment may further reduce complications associated with palatally impacted canines including root resorption of the lateral incisors and the need for more complex surgical and orthodontic intervention.14

The success of early interceptive treatment for impacted maxillary cuspids is influenced by the degree of impaction and age at diagnosis. As a general rule, when the degree of overlap between the permanent maxillary cusp and the neighboring lateral incisor exceeds half the width of the incisor root, the chances for complete recovery are poor. Clinical studies purport resolution of palatal impaction in 91% of cases in which the crown of the canine is distal to the midline of the lateral incisor when treatment is initiated. The success rate drops to 64% if the cusp crown is positioned mesial to the midline of the lateral incisor before interceptive treatment. Other factors influencing prognosis include canine angulation and crowding. Ericson and Kurol found that more mesially positioned canine cusp tips are associated with greater resorption of lateral incisor roots. Arch crowding can also have a significant influence; moderate to severe crowding indicates the need for complex orthodontic treatment to resolve the impaction and the malocclusion.15,16

CONCLUSION
Congenitally missing primary teeth are a rare entity, when found should be thoroughly investigated and treated. Simultaneously, the parents should be made aware of this condition and various modalities of treatment.

REFERENCES