A MULTIDISCIPLINARY APPROACH TO MANAGING BIMAXILLARY HYPERHYPODONTIA: A CASE REPORT

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Abstract

Bimaxillary hyperhypodontia (BHH) is a very rare numeric anomaly with a prevalence of 0.002% to 3.1% described by the presence of a supernumerary tooth in the premaxilla region and a missing mandibular incisor tooth. This case highlights the multidisciplinary management of a child presenting with BHH who complies with the recommended protocol by surgically removing the supernumerary tooth and then proceeding with orthodontic treatment for function and aesthetics. A 9-year-old healthy Malay boy presented with a fully erupted tooth 21, a labially palpable bulge of unerupted tooth 11, a missing tooth 32, and a tendency for an anterior and posterior crossbite. The radiographs showed an inverted, unerupted, conical-shaped supernumerary tooth overlapping the unerupted tooth 11 and hypodontia of tooth 32. The management was surgical removal of the supernumerary tooth and the placement of an upper removable appliance with a palatal expansion screw followed by comprehensive fixed orthodontics.

Keywords: Case Report, Dental Anomalies, Hyperhypodontia, Multidisciplinary Management, Pediatric Patient

Introduction

Hyperhypodontia is a combination of dental anomalies, which encompasses hyperdontia or pleiodontia as well as hypodontia or oligodontia, that can affect deciduous and/or permanent dentition. This condition was first documented by Cameliari in 1967 and occurs in either one or both jaws (1).

Hyperdontia is defined as the excess in the number of teeth or tooth-like structures in either deciduous or permanent dentition. The associated etiology of hyperhypodontia includes atavism, embryological aberration, dichotomy, hyperactivity of the dental lamina, progress zone theory, unified theory, and hereditary (2). The prevalence of hyperdontia is at 0.1- 6% and is seen more in males than females (3).

Meanwhile, hypodontia refers to failure of tooth/teeth development in deciduous or permanent dentition. Some theories behind the etiology of this condition have been discussed such as the predisposition of certain areas of the dental lamina to environmental changes, inherited anomaly, genetic influence, and environmental insults such as trauma, infection, and toxins. Its prevalence is 1.6-36.5% and is seen more in females than males (4).

On the other hand, the prevalence of both hyperdontia and hypodontia concomitantly or hyperhypodontia is only 0.002% to 3.1% (2). It has also been associated with cleft palate and cervical vertebrae abnormalities, Down's syndrome, Marfan syndrome, Ellis-van Creveld syndrome, and Opitz G/BBB syndrome (5). In 1979, Gibson further categorized this condition into premaxillary, maxillary, mandibular, and bimaxillary depending on the location of the anomaly in the jaw with a prevalence of 8.82%, 12.18%, 14%, and 65%, respectively (6). In Malaysia, there was one reported case of mandibular hyperhypodontia dating back to 1977 by Low, T. The patient was treated with restorative and orthodontic treatment (7).

A multidisciplinary approach combining the expertise of oral surgeons, prosthodontists, restorative specialists, and orthodontists has been recommended to achieve more ideal dentition (8). There are previously reported cases of bimaxillary hyperhypodontia (BHH) managed by the removal of the supernumerary tooth and closure of space due to hypodontia, either by restorative or orthodontics management (5, 9).

Interdisciplinary cooperation in dentistry provides ample experience and knowledge to attain an optimal outcome

for the successful management of patients with BHH. Hence, the goal of this case report is to discuss a case of BHH in a 9-year-old boy and its management, which involved a pediatric dentist, an oral surgeon, and an orthodontist, differing from previously reported cases.

Case Report

A 9-year-old healthy Malay boy came to the Paediatric Specialist Dental Clinic and presented with delayed eruption of his upper right front tooth. The child's medical history was uneventful. History taking revealed that his upper right front deciduous tooth was extracted due to an infection during comprehensive dental treatment under general anesthesia 2 years ago. Further questioning unveiled a family history of hypodontia (mandibular incisor) on the maternal side.

The extra-oral examination (Figure 1) revealed non-syndromic facial features. He has a mild Class II skeletal relationship with increased facial proportions (Figure 2 and Table 1).

Figure 1: The extra-oral photographs of the patient show a non-syndromic Class I skeletal relationship with a normal facial ratio

Figure 2: The lateral cephalogram of the patient

Table 1: The lateral cephalometric analysis of the patient showing mild Class II skeletal relationship

	Value	Normal value (Eastman)
SNA	82°	*81±3°*
SNB	77°	*78±3°*
ANB	5°	*3±2°*
MMPA	32°	*27±4°*
UiMx	126°	*109±6°*
LiMd	97°	*93±6°*

Upon intraoral examination, mixed dentition was noted with the presence of a fully erupted tooth 21, a labially palpable unerupted tooth 11, marked missing tooth 32, mobile tooth 62 as well as intact stainless-steel crowns on teeth 64, 74, 75, 85 (Figure 3). He presented with bilateral class I molar relationships. The patient presented with cusp to cusp on bilateral posterior teeth. Tooth 14 was erupting palatally and may predispose the patient to a posterior crossbite.

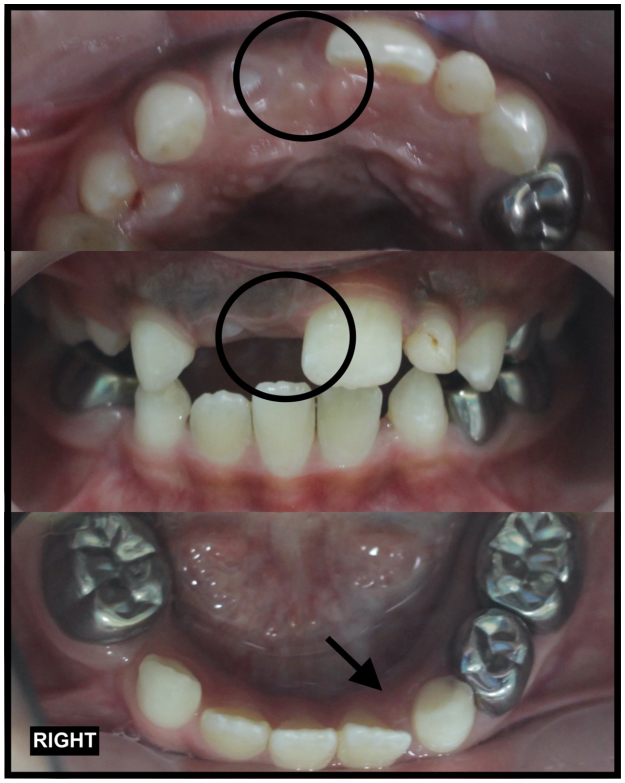

Figure 3: The intra-oral photographs of the upper occlusal and anterior view show clinically missing tooth 11 (marked by a black circle). The lower occlusal view shows missing tooth 32 (marked by a black arrow).

A panoramic radiograph was taken, and a conical-shaped supernumerary tooth was seen overlapping the unerupted tooth 11 and no tooth bud 32 was noted (Figure 4). All

permanent teeth were present except teeth 18, 28, 32, 38, and 48. A cone-beam CT scan (CBCT) was also taken to locate the position of the supernumerary tooth. The 14.3 mm length conical tooth was seen to be inverted and palatally placed with reference to the position of the unerupted tooth 11 (Figure 5). The root apices of the supernumerary and tooth 11 were open and at the R1/2 stage. The sagittal view exhibits thinning of the palatal bone plate with no lesions or adjacent structure displacement. The diagnosis of BHH was made based on the presence of the inverted supernumerary tooth in the premaxilla area and congenitally missing tooth 32.

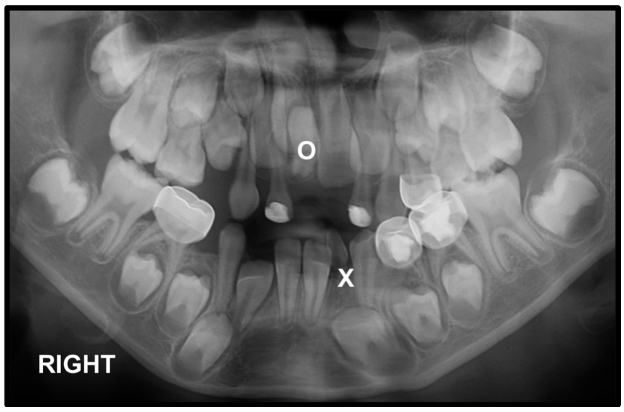

Figure 4: Panoramic radiograph showing the inverted supernumerary tooth overlapped with unerupted tooth 11 (marked by white circle) and missing 32 with no evidence of tooth bud (marked by green x). All permanent teeth are present except 32, 18, 28, 38, and 48.

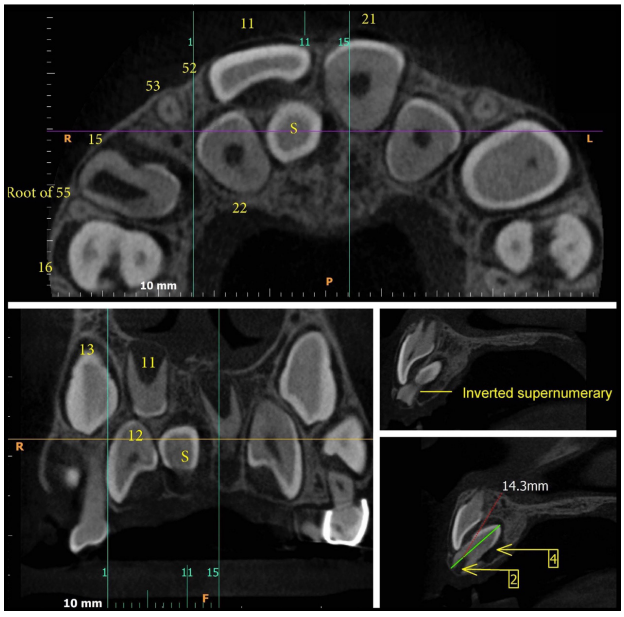

Figure 5: CBCT film of the anterior region of the maxilla demonstrating position of the inverted supernumerary (S) (marked by green line with length 14.3 mm) palatal impacted to tooth 11. Box 2 indicates the open apex of the supernumerary and Box 4 shows the thinning of the palatal wall.

Clinical management

Considering that tooth 11 is in a quite favorable position with incomplete root apex formation, coupled with the uncertainties of the COVID-19 pandemic, it was decided that surgical extraction of the supernumerary tooth was to be carried out without gold chain attachment. An upper removable appliance will be issued to the patient to maintain eruption space or expand the inter-canine distance if need be, whilst monitoring the spontaneous eruption tooth 11.

Removal of the inverted supernumerary tooth was done via palatal approach under general anesthesia by the oral and maxillofacial surgeon after local infiltration was administered using a 1.7 ml cartridge of 4% articaine hydrochloride and 1:100,000 epinephrine around the labial sulcus and the mid-palate. The bone on the palatal of tooth 11 was guttered after the full mucoperiosteal palatal flap was raised. After the supernumerary tooth and the remaining dental follicle were removed, the flap was sutured. (Figure 6(A-F)).

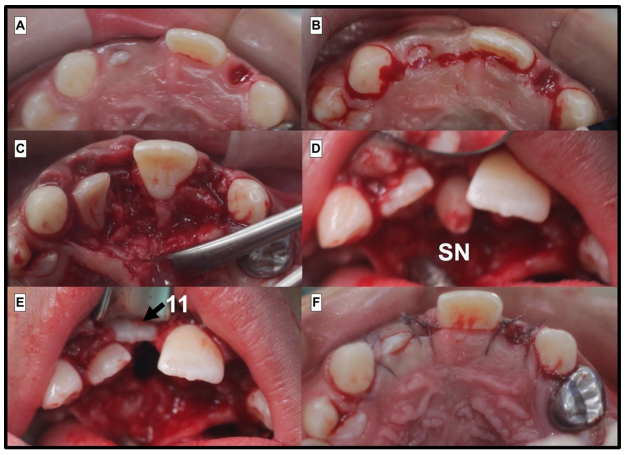

Figure 6: The intra-oral photos showing the upper occlusal view of the surgical removal of the unerupted supernumerary tooth through the palatal approach. (A), The pre-operative photo. (B), The crestal incision from mesial tooth 53 to tooth 63. (C), The full mucoperiosteal palatal flap was raised. (D), The unerupted everted conical supernumerary tooth (marked by SN) in relation to erupted tooth 21 and erupting tooth 12. (E), The impacted tooth 11 (marked by a black arrow) was located in between tooth 21 and tooth 12 ,and exposed post-extraction of the supernumerary tooth. (F), The post-operative photo shows the primary surgical closure using Vicryl 3/0 resorbable suture.

The patient was reviewed two weeks later and there was an evident lack of space for the eruption of tooth 11 due to tooth 12 and tooth 21 drifting into tooth 11 space [Figure 7(A)]. An upper removable appliance [Figure 8(A-B)]

with a midline expansion screw with activation once per week was issued to expand the intercanine distance and manage arch-tooth discrepancy to facilitate the eruption of tooth 11. Palatal finger springs were incorporated for retention and to provide further distal movement and space if necessary.

The post-operative three-month review showed the spontaneous eruption of tooth 11 [Figure 7(B)] as well as the palatal displacement of both upper lateral incisors. At this point, the removable appliance was ill-fitting, and the patient was instructed to stop using the appliance.

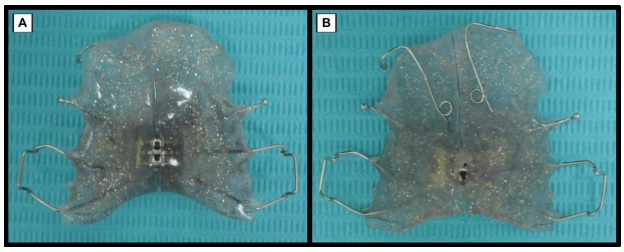

Figure 7: (A) The upper occlusal view shows a good surgical wound healing two-week post-surgery. (B) The anterior view demonstrates the erupting tooth 11 three-month post-operatively.

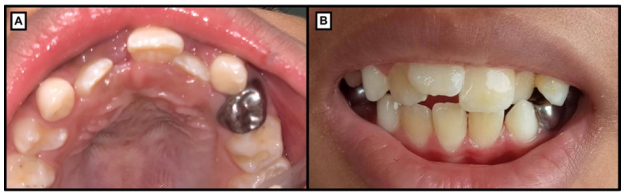

Figure 8: (A)The left photo shows the occlusal surface of the removable appliance. (B) The right photo shows the fitting surface of the removable appliance.

12-month dental follow-up after the surgery (Figure 9A & B) revealed that tooth 11 has erupted fully and proclined. There was mild crowding in the upper anterior segment with tooth 22 being palatally displaced. Overjet was 4mm on tooth 11 while overbite was 10% and incomplete. The permanent canines have yet to erupt.

The patient had mild spacing for the lower arch because of congenital missing tooth 32. Further definitive orthodontic treatment was required, and the options included a 2 x 4 appliance on the upper arch, or a full-arch fixed appliance once more permanent teeth erupts, most likely on an extraction basis of the upper premolars. The malocclusion will be reassessed prior to treatment with fixed orthodontic appliance.

Discussion

The introduction of BHH by Gibson in 1979 brought to our attention the occurrence of two dental anomalies with

Figure 9A: Extraoral photos of the patient 12 months post-surgery.

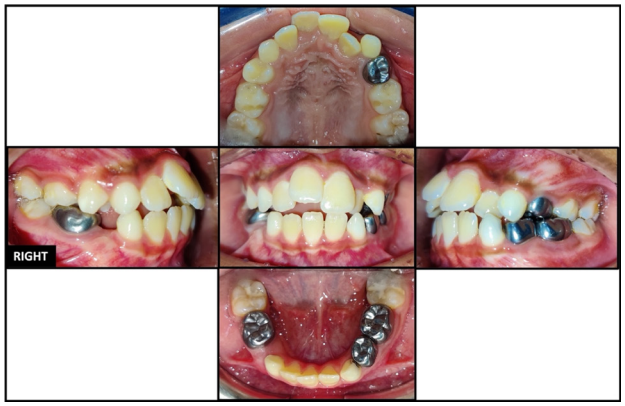

Figure 9B: Intraoral photos of the patient 12 months post-surgery.

opposing characteristics (6). It was described as having supernumerary teeth and missing teeth in the mandible or maxilla on the same patient at the same time be it on one or both jaws. The etiologies of the hyperhypodontia itself are still theoretical and this includes the probability of disturbances in neural crest migration (3). The case presented above could be due to embryologic aberration in which, the tooth germ of tooth 32 had migrated to the upper arch. It has been documented that the gender prevalence ratio of BHH in males: and females are 2:1 while the prevalence of oligodontia of the lower lateral incisor is 48%, pleiodontia in the premaxilla is 54% and an inverted supernumerary tooth is 47.7% (10). This shows that the case observed above is a relatively classic case of BHH as the patient fulfills most of the predictive factors.

The most significant effects of BHH itself are due to problems caused by hyperdontia and hypodontia itself. Hyperdontia over the anterior maxillary segment can cause deviated or prevention of teeth eruption, displacement of other teeth, and crowding (11). Additionally, hypodontia affects aesthetics by causing spacing or creating excess space between teeth whereas the functional issues that

could occur are the supra eruption of opposing teeth and varied gingival contours (4).

In such cases, to further understand the impact of this condition and predict possible complications, an orthopantomograph is used as the gold standard investigative radiograph to assess dentition and tooth development. Assessment done using the CBCT is only required for the localization of the supernumerary tooth to determine the most appropriate surgical approach (12).

In the present case, a palatal approach was used for the surgical removal of the supernumerary tooth as planned after considering the findings of the CBCT. The palatal mucosa consists of masticatory mucosa and was difficult to access in surgery for direct vision. The premaxilla area has the incisive foramen that contains the nasopalatine neurovascular bundle from the nasopalatine canal. There was a risk of vagal stimulation upon reflection of the palatal flap thus infiltration with local anesthesia containing vagolytic agents such as epinephrine over the bundle may block the pathway and prevent the reflex (13).

The probability for tooth 11 to erupt spontaneously is high as it is labially placed, with incomplete root formation and an open apex (14). For the upper arch, the expansion via slow maxillary expansion that was done using an upper removable appliance facilitated the formation of the space needed anteriorly for the eruption of the impacted tooth 11. Even though the patient did not exhibit outright posterior crossbite, the slow maxillary expansion was reported to offer more stable post-expansion, which could help correct that possible tendency (15).

The upper removable appliance was advantageous in this case in terms of evaluating patient compliance, good oral hygiene, and appliance maintenance (2). This phase of treatment began at the height of the COVID-19 pandemic outbreak; hence the removable appliance was used to limit the need for aerosol-generating procedures to prevent possible cross-infection. Adversely, the upper removable appliance can cause molar tipping, as well as buccal plate and root resorption of the anchoring teeth due to excessive stress at the apical and crestal region (16).

Conclusion

This case report highlights the different approaches to the management of BHH through surgical and orthodontics. A multidisciplinary team is key to comprehensively manage BHH, which includes expertise from pediatric dentists, oral and maxillofacial surgeons, orthodontists, and prosthodontists. The other vital component that contributes to the success of BHH treatments is the parent's and patient's compliance with the plan.

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Competing Interest

The authors declare that they have no competing interests.

Ethical Clearance

The parents were informed, and written consent was taken.

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