Concomitant Oligodontia and Supplemental Maxillary Lateral Incisor: A Case Report

Jinda Lertsirivorakul
Assistant Professor
Department of Pediatric Dentistry
Faculty of Dentistry, Khon Kaen University
Khon Kaen 40002
Tel: 043-202405
Fax: 043-244475
E-mail: jinda_le@kku.ac.th

Abstract

Simultaneous presence of hypodontia and hyperdontia, also called oligo-pleiodontia, is a rare condition. A case with the absence of all second premolars and mandibular first molars along with the presence of a supplemental lateral incisor has not previously been reported. This paper aimed to report the clinical and radiographic findings of a 9-year-old female who presented a supplemental left lateral incisor coexisting with oligodontia which involved the absence of the right maxillary first premolar, all permanent second premolars, both mandibular first molars and the right second molar. Comprehensive dental treatment plan was also discussed. The results suggested that concomitant oligodontia and hyperdontia can result in several clinical problems which may require multidisciplinary approach and long-term dental care.

Key words: hyperdontia; hypodontia; oligodontia; oligopleiodontia

Introduction

Oligodontia is defined as the congenital absence of six or more permanent teeth, excluding the third molars.[1] The prevalence in Caucasian populations in North America, Australia and Europe is very rare, approximately 0.14%, with a higher prevalence in female than in male.[2] Oligodontia can be found as an isolated nonsyndromic trait or as part of a syndrome, such as ectodermal dysplasia, incontinentia pigmentii, Down syndrome, and Rieger syndrome.[3] Isolated oligodontia is inherited in an autosomal dominant manner with reduced penetrance. Recently, mutations in the genes MSX1 and PAX 9 which encode transcription factors were demonstrated to be associated with isolated nonsyndromic oligodontia.[4] The dentition in oligodontia manifests in a wide diversity. Agenesis of premolars or maxillary lateral incisors, however, occur most frequently[5] and the most stable teeth are the maxillary central incisors and first molars.[1]

Supernumerary tooth, or hyperdontia, is a term used to define a presence of one or more extra teeth which develop in addition to the normal number found in a dentition.[6] A wide ranging prevalence of hyperdontia has been reported from 1.5 -3.5% with a male-to-female ratio of approximately 2:1.[7] Such variation may be attributed to study design and racial differences among the studied population. Classification of hyperdontia can be based on time of appearance (predeciduous, similar to permanent, postpermanent, and complementary), or their position in the dental arch (mesiodens, paramolars, postmolars, or impacted)[8], or on their morphology (conical, tuberculae, supplemental, or odontome).[9] Although 90% of all supernumerary teeth have been found in the premaxilla, only 7% were in the lateral incisor region.[10]

Concomitant hypodontia and hyperdontia,[11] or hypo-hyperodontia,[12] or oligo-pleiodontia,[13] is a very rare condition in which developmental missing teeth and supernumerary teeth occur in the same subject. It presents more often in the permanent dentition than in the primary or mixed dentitions.[14]

While several cases of this phenomenon have been reported in the literature, almost all of them were congenital absence of one or more second premolars or maxillary lateral incisor coexisting with hyperdontia, mostly maxillary mesiodens. Very few of them were oligodontia (Table 1). In addition, a case of oligodontia in which all second premolars, and mandibular first molars are congenitally absent has never been reported. This paper aims to describe a rare case of oligodontia coexisting with a supplemental maxillary lateral incisor.

Case Report

A healthy Thai female, aged 9 years, presented to the Pediatric Dentistry Clinic, Faculty of Dentistry, Khon Kaen University, because a dentist informed her mother that she had many missing teeth. The information collected from the parents and medical record from Srinagarind Hospital revealed that during pregnancy her mother underwent genetic amniocentesis due to elderly primigravida and the result showed normal chromosomes. The patient was born at 40 weeks gestation with a weight of 2,750 g and a length of 48 cm. Her father was 46 and her mother was 37 years old at her birth. Both parents and her 3-year-old brother were normal and healthy. There was no family history of hereditary tendencies to supernumerary or congenitally missing teeth. Her school performance was very good. The pediatrician who looked after her since birth was consulted and confirmed that apart from asthma, she had no other medical problems or signs of any syndrome. The dental history revealed regular routine dental care and no history of traumatic injury.

General physical and extra-oral examination of this patient was within normal range although she had slightly small stature. Her weight (22 kg) was at the 25th percentile of the standard weight and her height (121 cm) was at the 5th percentile of the standard height of Thai children. She had bilateral facial symmetry and straight profile. Intra-oral examination revealed healthy soft tissues in the presence of average oral hygiene practices. As shown in Fig 1, she had mixed dentition with the following teeth erupted:

#16, #55, #54, #53, #12, #11	#21, #22, #22, #63, #64, #65, #26
#85, #84, #83, #42, #41	#31, #32, #73, #74, #75

The patient had centered mandibular midline. However, the maxillary incisor center-line shifted approximately 1 mm to the right with slightly distal tipping of teeth #11 and #12, contributed to the presence of a supplemental maxillary left lateral incisor which was of normal shape and size (Fig 2). The overbite was 4 mm and overjet was 8 mm. The canine relationship was Class II on the right and Class I on the left. The primary molar relationship was slightly distal step both sides (Fig 3).

Radiographic examinations consisting of bitewings and orthopantomograph revealed dental caries on proximal surfaces of upper primary molars and several missing teeth suggestive of congenital absence of the following permanent teeth:

#15, #14	#25
#47,#46, #45	#35,#36

In addition, there was no sign of follicles or commencing calcification of third molars (Fig 4).

To ensure optimum function and esthetics, a multidisciplinary discussion regarding her treatment plan has been established among a pediatric dentist, an orthodontist, an oral and maxillofacial surgeon, and a prosthodontist. Preventive measures included oral hygiene instruction, diet counseling, sealant on teeth #16 and #26 and topical fluoride treatment. The maxillary lateral incisor which was lingually erupting, and teeth #53, #63 and #54 were extracted in order to allow the eruption of teeth #13 and #23. Teeth # 55 and #65 were restored using composite resin and will be retained as long as possible to obviate need for a partial denture retainer. Since the mother was concerned about the proclined incisors and the permanent canines nearly erupted, the orthodontist decided to place fixed upper and lower orthodontic appliances in order to prepare for the eruption of teeth #13 and #23 and correct the deep overbite.

Table 1 *Previous case reports of concomitant hypodontia and hyperdontia*

Authors	Hypodontia	Hyperdontia
Camilleri,1967[11]	#12, #22	Maxillary mesiodens
Munns,1967[15]	#12, #22	#15
Brook and Winter, 1970[16]	#12, #22	Maxillary mesiodens
Mercer, 1970[17]	#15, #35, #45	Maxillary mesiodens
Nathanail, 1970[13]	#35, #45	Maxillary mesiodens
Low, 1977 [18]	#31, #41	Mandibular mesiodens
Gibson, 1979[12]	#12	Maxillary mesiodens
	#22	Maxillary mesiodens
	#15, #22	Maxillary mesiodens
	#15, #25	Maxillary mesiodens
	#15	Maxillary mesiodens
	#31, #41	Mandibular mesiodens
	#42	Four denticles in #45, #46 region
	#45	#12
	#15, #25, #35	Maxillary mesiodens
	#15	Maxillary mesiodens
	#35, #45	Maxillary mesiodens
	#15, #25, #35, #45	Maxillary mesiodens
	#25	#17, #26
	#22, #23, #25, #35, #31, #41, #45	Maxillary mesiodens
	#18	Maxillary mesiodens
	#38, #48	Maxillary mesiodens
	#18, #38, #48	Maxillary mesiodens
	#18, #28, #38, #48	Maxillary mesiodens
	#18, #28, #38, #48	Maxillary mesiodens
	#18, #28, #38, #48	Maxillary mesiodens
Spyropoulos et al, 1979[8]	#15, #14, #12, #22, #24, #25, #35, #37, #44, #45	Mandibular incisor
	#13, #45	Mesiodens
	#31	#22
Segura et al,1998[19]	#22	Mesiodens
Zhu et al, 1999[20]	#12, #22	Region #46
Sharma, 2000[21]	#23	#15, #14, #11, #21, #22, #24, #25, #34, #35, #32, #42
Matsumoto et al, 2001[22]	#32, #25	#22
El-Bahannasawy et al, 2004[23]	# 53	#54, #14
Das et al, 2006[24]	#31, #41	Area #31, #41

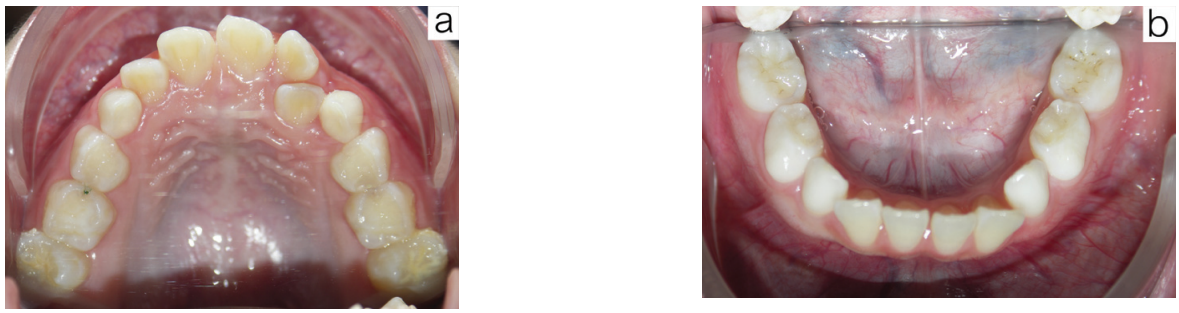

Fig. 1 *Occlusal views of maxilla (a) and mandible (b) show all erupted teeth.*

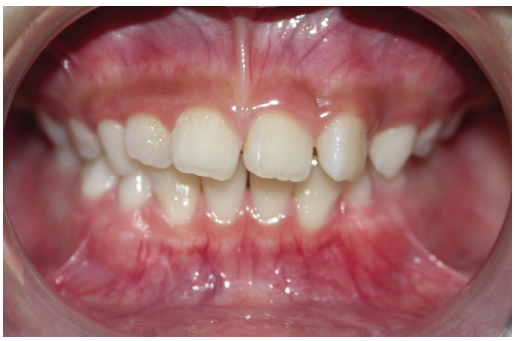

Fig. 2 *Frontal view shows the alignment of anterior teeth.*

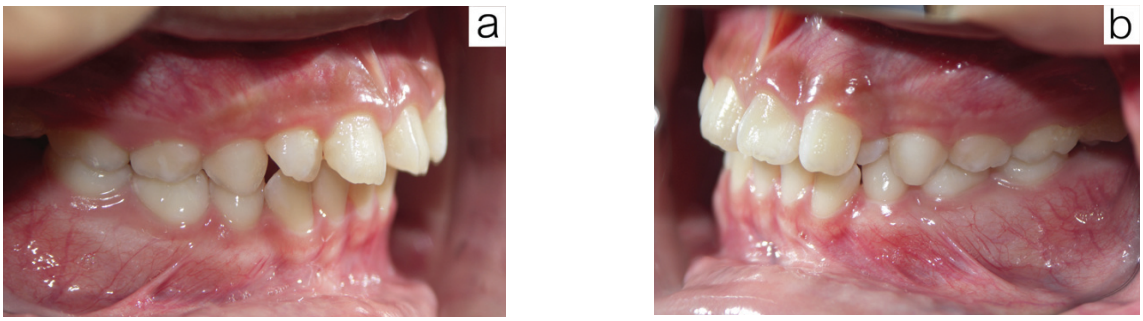

Fig. 3 *Intra-oral views show the relationships of the teeth on the right (a) and the left (b) sides.*

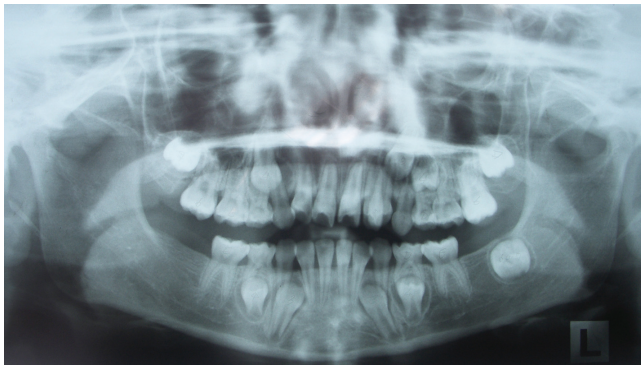

Fig. 4 *Panoramic radiograph shows a supplemental left maxillary lateral incisor and congenital absence of the maxillary first premolar, all second premolars, both mandibular first molars and the right second molar.*

The management of congenital absence of teeth #36 and #46 in this girl can be a transplantation of teeth #17 and #37 or #27 to take the place of teeth #36 and #46, carried out by an oral and maxillofacial surgeon when the permanent second molars have between one-half to two-thirds completed root development [25] or at the ages of 11-12 years.[26] Another option is a transplantation of tooth #27 to take place of tooth #46 plus orthodontic movement of tooth #37 to take place of tooth #36. The patient, therefore, will be radiographically assessed annually for the formation of those molars. If space is expected, a removable partial denture will be placed with possible replacement with a permanent prosthesis. The appropriate prosthesis with one or more implants, or without implant, will then be designed for her when she achieves dental and skeletal maturation[27] or approximately 20 years old. [28] The presence or absence of third molar has to be taken into account when designing the prosthesis. Third molar agenesis may occur up to the age of 16 years, although the possibility of their appearance after the age of 12 years is reduced.[29]

Discussion

Concomitant hypodontia and hyperdontia is a very rare phenomenon. Mercer[17] estimated that the probability of both anomalies coexisting lay between 8 and 15 per 10,000. Rose[30] reported a frequency of 13 in 10,000 patients. Gibson[12] reported a prevalence of 0.4% in 4,598 orthodontic patients. The etiology is unknown. Disturbances in migration, proliferation and differentiation of the neural crest cells and interactions between the epithelial and mesenchymal cells during the initiation of odontogenesis have been suggested.[14]

To the best of literature searching, this is the first case reported of oligo-pleiodontia in which the maxillary first premolar, all second premolars, both mandibular first molars and the right second molar are congenitally absent, along with a supplemental left maxillary lateral incisor.

The coexistent hypodontia and hyperdontia may result in several clinical complications. Supernumerary teeth may be impacted and associated with interference of normal eruption, impaction and displacement of the adjacent teeth.[31] On the other hand, hypodontia causes many treatment problems. In this case, the supplemental lateral incisor resulted in labioversion of tooth #12. In addition, the oligodontia which involved both mandibular first molars, the most stable teeth, and all premolars brought many complicated problems. Treatment planning of this case required multidisplinary approaches which included several specialists. Orthodontic treatment has been commencing early to prepare proper dental arrangement for future surgical and prosthodontic treatment. Regular clinical and radiographic reviews are of paramount importance in order to facilitate appropriate timing of interventions for long-term dental management.

Acknowledgement

The author wishes to express her gratitude to Prof. Keith Godfrey for his advice on the manuscript preparation.

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บทวิทยาการ

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จินดา เลิศศิริวรกุล
ผู้ช่วยศาสตราจารย์ ภาควิชาทันตกรรมสำหรับเด็ก
คณะทันตแพทยศาสตร์ มหาวิทยาลัยขอนแก่น
อ.เมือง จ.ขอนแก่น 40002
โทรศัพท์: 043-202405
โทรสาร: 043-244475
อีเมล: jinda_le@kku.ac.th

บทคัดย่อ

ภาวะฟันน้อยเกินและฟันเกินที่เกิดพร้อมกันหรือที่เรียกอีกชื่อหนึ่งว่า โอลิโกไพลโอดอนเทียนั้นเป็นภาวะที่พบได้ยาก กรณีที่มีการหายไปของฟันกรามน้อยซี่ที่สองทุกซี่และฟันกรามล่างซี่ที่หนึ่งทั้งคู่พร้อมกับการมีฟันตัดข้างเกินยังไม่เคยถูกรายงานมาก่อน บทความนี้มีวัตถุประสงค์เพื่อรายงานผลการตรวจทางคลินิกและภาพรังสีของผู้ป่วยหญิงอายุ 9 ปี ที่มีฟันตัดข้างบนซ้ายเกินพร้อมกับภาวะโอลิโกดอนเทีย ซึ่งมีการหายไปของฟันกรามน้อยบนขวาซี่ที่หนึ่ง ฟันกรามน้อยซี่ที่สองทุกซี่ ฟันกรามล่างซี่ที่หนึ่งทั้งคู่และฟันกรามล่างขวาซี่ที่สอง นอกจากนี้ยังกล่าวถึงแผนการรักษาแบบพร้อมมูลด้วย บทความนี้ชี้ให้เห็นว่าโอลิโกดอนเทียพร้อมกับฟันเกิน สามารถส่งผลให้เกิดปัญหามากมายทางคลินิก ซึ่งอาจต้องการการจัดการแบบสหวิทยาการและการดูแลทางทันตกรรมเป็นระยะเวลายาวนาน