

Lobodontia and Multiple Dens Invaginatus: A Case Report

Khumpee Songkapol

Instructor

Department of Oral radiology

Faculty of Dentistry, Mahidol University

Preeyaporn Srimawong

Instructor

Department of Oral radiology

Faculty of Dentistry, Mahidol University

Warungkana Chidchuangchai

Assistant Professor

Department of Oral Medicine

Faculty of Dentistry, Mahidol University

Correspondence to:

Khumpee Songkapol

Instructor

Department of Oral Radiology,

Faculty of Dentistry, Mahidol University

Bangkok 10400

Tel: 02-2036490

E-mail: dtksk@mahidol.ac.th

Abstract

A rare case of multiple dental anomalies in a 19-year-old Thai male is reported. Intraoral examination presented a combination of anomalies which involves the entire dentition with no apparent systemic complications. These dental anomalies include shovel-shaped incisors, carnivorous-like crowns and multitubercular crowns. Radiographic findings revealed ten dens invaginatus associated with four periapical radiolucencies and abnormal root shaped molars.

Key words: dens invaginatus; dental morphology; lobodontia; multiple anomalies

Introduction

Developmental alterations of teeth can be classified according to the shape, size, number and structure of the tooth.¹ They may involve a single tooth or multiple teeth in a patient. Differences of dental anomalies may occur in the same individual, and even the same tooth, but multiple anomalies of tooth morphology in the same individual are extremely rare, especially in the entire dentition. Abnormalities of the tooth shape may occur in the crowns or in the roots. Crown morphology may entirely be distorted with the loss of the usual cusp and groove relationship. Examples of anomalies seen in anterior teeth are shovel-shaped incisors, peg-shaped incisors, deep lingual pit and fossa, bifid cingulum, and supernumerary cusp (talon cusp), while the posterior teeth may have the cusp of Carabelli, multitubercular crowns, and even central occlusal cusps. These anomalies may associate with dens evaginatus, dens invaginatus or abnormally shaped roots, which can be revealed by radiographic examination.²⁻⁵

The occurrence of multiple dental anomalies which involve the entire dentition is relatively rare. Few studies²⁻⁵ concerning odd-shaped and barrel-shaped teeth, deep lingual pit or very distinct mamelons in incisors, peak-shaped (fang-like) cusps with a prominent labial lobes in canines, pointed cusps or pyramidal cusps in premolars and multituberculate crowns in molars have been reported. Radiographs revealed dens invaginatus affected in multiple teeth, and single conical root form in molars.²⁻⁵ The clinical manifestation of the canines were quite conical in shape and the posterior teeth have sharp, cutting teeth similar to the carnivorous species. Witkop⁶ described the dentition that appeared in carnivores having fang-like cusp in canines and premolars, and anomalous cusps on molar teeth as "lobodontia". The teeth of this condition resemble multiple axial core defects, such as dens invaginatus or evaginatus defects.

The aim of this study was to report the clinical and radiographic findings in a new case of lobodontia condition and to review multiple dental anomalies.

Case Report

A 19-year-old Thai male visited the dental clinic at the Faculty of Dentistry, Mahidol University, Bangkok, Thailand for a routine dental examination. He had no significant medical and dental history. No familial history of dental anomalies could be ascertained. The extraoral examination showed a symmetrical straight facial profile. The intraoral examination revealed normal color and texture of oral mucosa, except fistula opening at the left buccal mucosa between the upper first and second premolars. The patient's teeth revealed 31 permanent teeth, except the lower right third molar. The intraoral photographs, occlusal model, periapical full mouth radiographs were obtained with the patient's informed consent. The upper left deciduous canine is still retained with a good clinical stability. (Fig. 1A) There were crowding at the upper left premolars, and spacing between the deciduous and permanent canines and also at the lower anterior teeth. (Fig. 1A, 1B) The interarch relationship was crossbite at both anterior and posterior teeth as shown in Fig. 2A and Fig.



Fig. 1 Intraoral photographs. A, Upper arch. B, lower arch. The various abnormality in shape and crown morphology in entire dentition is shown. The prolong retention of the upper left deciduous canine is also presented (arrow)



Fig. 2 The patient's occlusion. A, The anterior view. B, The right side. C, The left side. The anterior and posterior cross bite are observed. The peak-shaped (fang like) cusps in canines, prominent point cusp in premolars and lower first molars, and fistula opening (arrow) at the buccal mucosa of upper left premolars are also illustrated.

2B, 2C, respectively. From the intraoral photographs and occlusal model, the teeth were divided into 4 groups according to morphologic features, as follow:

1. Incisors group: The upper central incisors were shovel-shaped with very distinct three mamelons, while the lateral incisors showed the prominent cingulum with an abnormally deep lingual pits. (Fig. 3A) The lower central incisors were bulbous-shaped with a distinct notch at the incisal edge, while the lateral incisors showed very distinct three mamelons. (Fig. 3B)

2. Canines group: The crown of both upper and lower canines had a prominent convex labial ridge separated by pronounced developmental groove and also showed peak-shaped cusp, especially in the lower canines which slightly curve backward into the cusp tip. (Fig. 3B)

3. Premolars group: The crown of upper and lower premolars showed the reduction in size with non existent lingual cusps, as shown in Fig. 4A, 1A and Fig. 4B, 1B respectively. All of them exhibited the exaggeration of the middle labial lobes with a sharp prominent pointed cusp resembling carnivore teeth morphology.

4. Molars group: The crown of both upper and lower molars was multitubercular in its appearance, as shown in Fig. 4A, 1A and Fig. 4B, 1B respectively. The occlusal surface showed steep ridges separated by deep grooves system with marginal ridge tubercles. The upper third molars also exhibited central cusps.

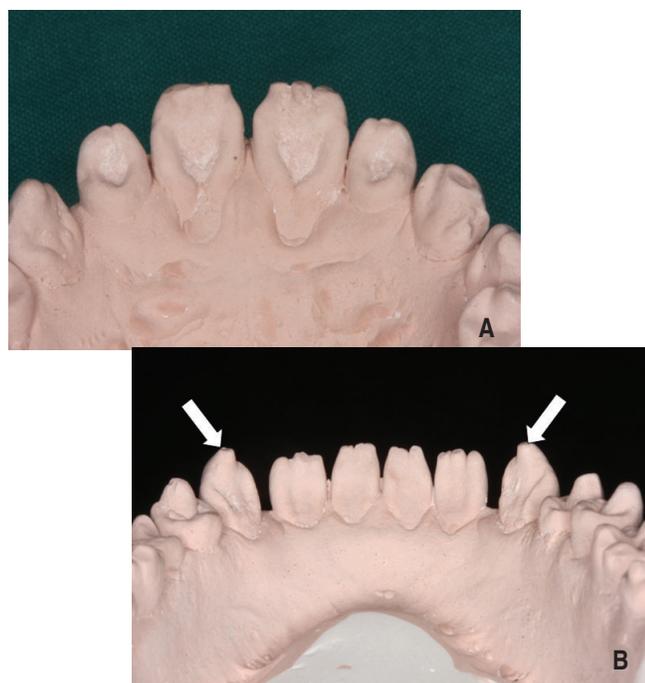


Fig. 3 Incisal and canine groups. A, The upper occlusal model shows shovel-shaped central incisors and deep lingual pit with a prominent cingulum in lateral incisors. B, The lower occlusal model shows bulbous-shaped crown with a notch in central incisors and very distinct three mamelons in lateral incisors. The cusp tip of the left and right canine also exhibited slightly curve and backwardness (arrows)

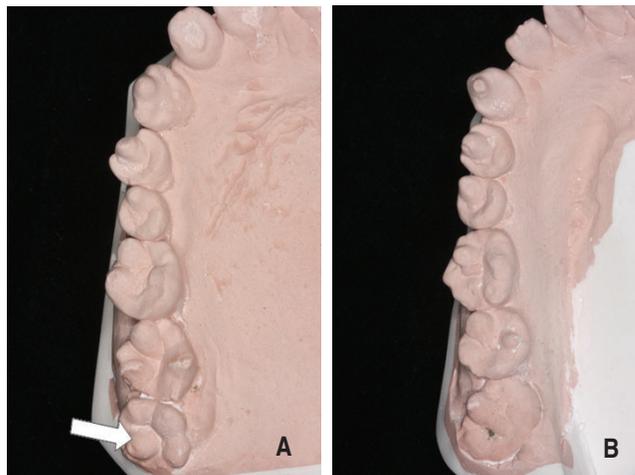


Fig. 4 Premolar and molar groups. A, The right upper model shows the reduction in crown size of premolars, multitubercular crown in all molars, and also demonstrates central cusp in upper third molar (arrow). B, The left lower model shows the prominent pointed cusp in premolars and multitubercular crown in all molars

Radiographic examination revealed normal development of maxilla, mandible and temporomandibular joint, except tooth morphology in most dentition. (Fig. 5, 6) The radiographs of upper incisors showed minor form of the invagination with enamel infolding confined to the crown. The premolar radiographs showed the invagination at the coronal region as a blind sac at the upper right premolars, the upper left second premolar, the lower left and right second premolars, while the invagination penetrating and perforating the root apex of the upper left first premolar. Four periapical radiolucencies were found in the upper right premolars, the upper left first premolar and the lower right second premolars. Furthermore, single conical-shaped roots of all upper and lower molars, except the lower left and right first molars, were observed. Moreover, the extensive severe vertical bone loss at the upper left and right first molars was noted.

Due to painless and clinically asymptomatic in affected teeth, the initial diagnosis were pulp necrosis with chronic apical periodontitis for the upper right premolars and lower right second premolar, and chronic apical abscess for the upper left first premolar. The patient was referred to endodontic clinic for root canal treatment of these teeth.



Fig. 5 Panoramic radiograph showed generalized abnormalities of dental morphology

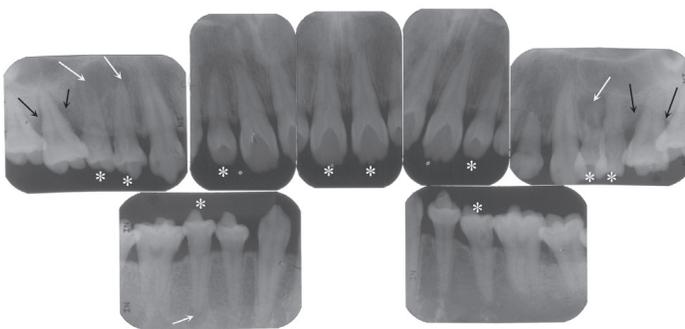


Fig. 6 Periapical radiographs demonstrated ten dens invaginatus (*), four apical radiolucencies (white arrows) and severe vertical bone loss at the upper right and left first molars (black arrows)

Discussion

The clinical finding of multiple dental anomalies and radiographic appearances in our case were similar to those described by previous authors. (Table 1) The dentition resembles the early mammalian or Triconodont stage which exhibit three buccal cusps in line in the development of posterior teeth. The largest cusp is centered buccally with smaller cusps located anteriorly and posteriorly. The lower canine occludes into a space between the upper canine and outermost incisor.⁷ Some of the teeth can be seen in the carnivorous animals such as *Canis familiaris*. (domestic dog). The term “lobodontia” have been used to describe this characteristic in human teeth.⁶ Gorlin⁸ supported this unusual dental anomalies may be hypodontia or general reduction in crown size. The incisors tend to be invaginated or to be shovel-shaped. The cingulum of incisors and premolars is accentuated.

Recently, Metgud et al⁹ have reported a bizarre combination of anomalies which involves single conical unbifurcated molar root forms, taurodontism, dens invaginatus, prominent labial lobes of the canines, pyramidal cusps of the premolars, dens evaginatus of the molars, and localized reduction in tooth size involving the entire dentition without any other apparent systemic complications. We believed that this report is another case of lobodontia condition.

The etiology of multiple dental anomalies remains unclear. However, the genetic and environmental factors may influence on early tooth development.¹⁰ The defective gene may critically be expressed by transmission from X-linked dominant, autosomal dominant or autosomal recessive.¹¹ Robbins and Keene² and Brook and Winder⁴ described that the inheritance pattern of multiple dental anomalies is compatible with autosomal dominant trait. The mutation of a single gene can influence the process of odontogenesis during morphodifferentiation which result in the shape of the entire dentition. This phenomenon is supported by Nguyen et al⁵ who found such a phenomenon in two successive generations, although they cannot draw a conclusion. However, our report and Metgud et al⁹ are doubtful because the familial history were not carried out.

In the present case, the different morphologic characteristic found in the incisors is shovel-shaped. This unusual crown morphology which is often considered an anatomical variant rather than a morphological defect because this trait usually occur almost universally with a relatively high frequency rate in the Chinese, Mongoloid, Japanese, Eskimo, and American Indians.¹² This trait is a combination of a prominent marginal ridge enclosing a deep lingual fossa. Oehlers¹³ stated that this morphology usually occurred in the minor form of enamel invagination, as seen in the deep fissuring in the upper central incisors of the our case. The presence of distinct mamelons also remained in the upper central incisors and the lower lateral incisors as well as to the previous reports.^{2,4} Normally, the evidence of distinct mamelons in adult may be found with an anterior open bite relationship by which the teeth do not contact in functions.

Dens invaginatus is a developmental anomaly that dentists should be familiar with. Usually this affects maxillary incisor teeth, particularly lateral incisors. It is uncommon in the posterior teeth and very rare to observe this anomaly in multiple teeth or all quadrants. In earlier studies, many cases of multiple dens invaginatus without other multiple dental anomalies and

Table 1 Clinical and radiographic appearances of the patients with multiple dental anomalies as lobodontia

Authors (Year)	Robbins & Keene ² (1964)	Shuff ³ (1972)	Brook & Winder ⁴ (1979)	Nguyen et al. ⁵ (1996)	Metgud et al. ⁹ (2009)	Present authors (2010)
Clinical appearances						
Incisors						
barrel-shaped incisors	+	-	-	+	-	-
shovel-shaped incisors	-	-	-	-	-	+
very distinct mamelons	+	-	-	+	-	+
deep lingual pit	+	-	+	+	-	+
labioincisal groove	-	-	-	-	+	-
round incisor corner	-	+	-	-	-	-
Canines						
peak-shaped (fang-liked cusp)	+	+	+	+	+	+
exaggeration of middle lobe	+	+	+	+	+	+
Premolars						
reduction in size with non existent lingual cusp	+	+	+	+	+	+
pyramidal (pointed) cusps or canine-like appearance	+	+	+	+	+	+
Molars						
multitubercular appearance	+	+	+	+	+	+
Abnormal Tooth Numbers						
Hypodontia / missing tooth	-	+	+	+	-	-
Supernumerary tooth	-	+	-	-	-	-
Radiographic appearances						
multiple dens invaginatus (number of affected teeth)	+(5)	-	+(4)	-	+(2)	+(10)
single conical / non bifurcated roots	+	+	+	+	+	+
taurodontism	-	-	-	-	+	-
Genetic transmission (autosomal dominant)						
	+	+	+	+	?	?

+ present , - absent , ? unknown

medical syndromes have been reported.¹⁴⁻¹⁸ Sedano et al¹⁹ reported a case in which multiple dens invaginatus with various dental anomalies in shape and no familial history is detected. In contrast, Casamassimo et al²⁰ described multiple dens invaginatus combined with microdontia, taurodontia and suggested an X-linked recessive mode of transmission. From the previous cases of lobodontia condition, multiple dens invaginatus were also described.^{2,4} Robbins and Keene² detected dental invagination in all four maxillary incisors and the mandibular right first premolar. Brook and Winder⁴ found the invagination in only four maxillary incisors whereas the reports of Nguyen et al⁵ and Metgud et al⁹ were absent. In the present case, radiographic examination revealed ten invaginations which affected all quadrants in varying degree. According to Oehlers' classification,¹² type I was observed in the upper central incisors and the lateral incisors which was seen as a deep fissuring confined to the crown. Type II was observed in the upper right first premolar, the upper second premolars, and the lower second premolars which was seen as a blind sac or tear drop and not grossly dilated to the roots and crowns. Type III was observed in the upper left first premolar which was seen as the invagination penetrated the root and bursts apically. Multiple apical radiolucencies were also found in four teeth with clinically asymptomatic history due to the slowly necrosis of the pulpal tissue. The great number of invaginated teeth with all types of the degrees of invagination has not ever been reported in the literature.

Molar teeth which were irregular tooth form and present supernumerary cusps are called multituberculism or multituberculate crown. The small accessory cusps or interstitial cusps when viewed from the occlusal look like multiple dens evaginatus. This condition usually affect all of the molar teeth,^{2-5,9} which is the same as our report. Moreover, we also found the prominent central cusps in the maxillary third molars. This morphology was situated in the center of the occlusal surface of a molar and has also been classified as dens evaginatus. However, it is well-recognized among clinicians as it occurs primarily in premolars and is very common to expose to the fine pulpal extension. Studies on the central cusp in molar are relatively rare. Kocsis et al²¹ defined central cusp as a supernumerary macrostructure of the tooth surface, representing a cusp formation on the occlusal surface of premolars and molars. The surrounding cusp enamel may sometimes be separated from the cusp by a mild groove. This report showed the large prominent

central cusp with normal state of apical region from radiographic examination. However, multituberculate crown or pyramidal cusp which is predisposed towards periapical inflammatory response similar to dens evaginatus. Nonetheless there is no radiographic evidence of the pulp having extended into the protrusion of those cusps. Metgud et al⁹ reported that an asymptomatic periapical lesions affected in two premolars associated with pyramidal cusps resulting from occlusal trauma.

An abnormal root form is affected by all of the molars including single conical root or non-bifurcated root forms. This morphology has a single root tapering from the cervix to the apex. Nguyen et al⁵ used the term "pyramidal root" when the teeth showing complete fusion of the roots with a solitary enlarged root canal and the term "fused root" to refer to teeth with the root surfaces connected but still maintaining at least two distinct canals. Anywhere single conical roots were rarely found in the first molars.²² Some authors believe that this root abnormality results from failure of invagination of Hertwig's epithelial root sheath. The root surface area for periodontal attachment is diminished. Hou et al²³ found that a majority of the periodontal disease in molars with fused root exhibited deeper developmental grooves than normal molars. These grooves lead to accumulation of dental plaque, development of deep probing depth and rapid progression of attachment loss. Moreover, an unfavorable crown-to-root ratio and taper-shaped root offer low resistance to heavy occlusal load. This phenomenon may cause an increased risk of localized periodontal disease as seen in the upper first molars of our case.

A patient who has multiple dental anomalies can be part of various syndromes or may relate to other system⁸ such as Otodontal syndrome, and Oculo-facio-cardio-dental (OFCD) syndrome. These syndromes are genetically transmitted, and may affect many organs leading to poor quality of life. Otodontal syndrome - an autosomal dominant syndrome which showing the abnormalities of deciduous and permanent dentition, including large bulbous teeth with malformed occlusal surfaces (globodontia) and high incidence of sensorineural hearing loss. OFCD syndrome - an X-linked dominant syndrome consists of 4 following features (1) dental anomalies i.e. canine radiculomegaly, delayed dentition, oligodontia, persistent primary teeth, variable root length (2) eye anomalies i.e. congenital cataracts, microphthalmia, microcornea (3) facial abnormalities i.e. long narrow face, high nasal bridge, short nose with broad tip, long philtrum (4) cardiac anomalies i.e. atrial septal defect (ASD),

ventricular septal defect (VSD) including lethal in male. However, the abnormalities of lobodontia occurred only in dentition, not related to other system.

A clinical management or treatment consideration for a patient with multiple anomalies varies among cases. Consultation with Endodontic, Periodontic, Prosthodontic, and Surgery clinics are required as needed. To identify the crown and root morphology, a thorough radiographic evaluation should be examined, including periapical and periodontal status. When the abnormal tooth morphology is observed (such as shovel-shaped crown, deep groove or fissure, exaggerated cingulum pit), the enamel invagination is possibly presented.²⁴ The clinicians should be aware of this condition, because the high risk of dens invaginatus affected multiple teeth and may have asymptomatic apical inflammatory disease. Prophylactic restoration of the pit and fissure of those teeth is important to avoid possible pulpal injury and related inflammation. In addition, selective grinding or cusp reducing in the pointed cusps may be performed to prevent traumatic occlusion.

Conclusion

The case described in this report is a rare entity of lobodontia condition. The major characteristics of this condition include anomaly of crown shape in almost or entire dentition, multiple dens invaginatus which affects a varying number of teeth, single conical root or non bifurcated root forms in all molars and autosomal dominant trait. A marked variant are hypodontia, missing teeth, and taurodontism. The diagnosis should be based on the clinical and radiographic evaluation, and familial history should also be clarified.

Acknowledgements

The authors gratefully acknowledge Assistant Professor Suchaya Pornprasertsuk-Damrongsri, Department of Oral Radiology, Faculty of Dentistry, Mahidol University, for her advice, and Assistant Professor Boonjeera Chiravate, Department of English, Faculty of Arts, Silpakorn University, for her assistance in language correction.

References

1. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and maxillofacial pathology. Philadelphia: WB Saunders; 2002. p. 50-100.
2. Robbins IM, Keene HJ. Multiple morphologic dental anomalies. Report of a case. *Oral Surg Oral Med Oral Pathol* 1964; 17:683-690.
3. Shuff RY. A patient with multiple conical teeth. *Dent Pract Dent Rec* 1972;22:414-7.
4. Brook AH, Winder M. Lobodontia - a rare inherited dental anomaly. Report of an affected family. *Br Dent J* 1979;147: 213-5.
5. Nguyen AM, Tiffée JC, Arnold RM. Pyramidal molar roots and canine-like dental morphologic features in multiple family members: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1996;82:411-6.
6. Witkop CJ Jr. Hereditary defects of dentin. *Dent Clin North Am* 1975;19:25-45.
7. Ash MM, Jr. Wheeler's dental anatomy, physiology and occlusion. Philadelphia: WB Saunders; 1993. p. 89-94.
8. Gorlin RJ. Otodental syndrome, oculo-facio-cardio-dental (OFCD) syndrome, and lobodontia: dental disorders of interest to the pediatric radiologist. *Pediatr Radiol* 1998;28:802-4.
9. Metgud S, Metgud R, Rani K. Management of a patient with a taurodont, single-rooted molars associated with multiple dental anomalies: a spiral computerized tomography evaluation. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009;108:e81-6.
10. Brook AH. A unifying aetiological explanation for anomalies of human tooth number and size. *Arch Oral Biol* 1984;29:373-8.
11. Hu JC, Simmer JP. Developmental biology and genetics of dental malformations. *Orthod Craniofac Res* 2007;10:45-52.
12. Pindborg JJ. Pathology of the dental hard tissues. Philadelphia: WB Saunders; 1970. p. 39.
13. Oehlers FA. Dens invaginatus (dilated composite odontome). I. Variations of the invagination process and associated anterior crown forms. *Oral Surg Oral Med Oral Pathol* 1957;10:1204-18 contd.
14. Krolls SO. A dentition with multiple dens in dente. *Oral Surg Oral Med Oral Pathol* 1969;27:648.

15. Burton DJ, Saffos RO, Scheffer RB. Multiple bilateral dens in dente as a factor in the etiology of multiple periapical lesions. **Oral Surg Oral Med Oral Pathol** 1980;49:496-9.
16. Karaca I, Toller MO. Multiple bilateral dens in dente involving all the premolars. Case report. **Aust Dent J** 1992;37:449-52.
17. Altinbulak H, Ergul N. Multiple dens invaginatus. A case report. **Oral Surg Oral Med Oral Pathol** 1993;76:620-2.
18. Hosey MT, Bedi R. Multiple dens invaginatus in two brothers. **Endod Dent Traumatol** 1996;12:44-7.
19. Sedano HO, Ocampo-Acosta F, Naranjo-Corona RI, Torres-Arellano ME. Multiple dens invaginatus, mulberry molar and conical teeth. Case report and genetic considerations. **Med Oral Patol Oral Cir Bucal** 2009;14:e69-72.
20. Casamassimo PS, Nowak AJ, Ettinger RL, Schlenker DJ. An unusual triad: microdontia, taurodontia, and dens invaginatus. **Oral Surg Oral Med Oral Pathol** 1978;45:107-12.
21. Kocsis GS, Marcsik A, Kukai EL, Kocsis KS. Supernumerary occlusal cusps on permanent human teeth. **Acta Biol Szeged** 2002;46:71-82.
22. Cleghorn BM, Christie WH, Dong CC. Root and root canal morphology of the human permanent maxillary first molar: a literature review. **J Endod** 2006;32:813-21.
23. Hou GL, Tsai CC, Huang JS. Relationship between molar root fusion and localized periodontitis. **J Periodontol** 1997;68:313-9.
24. Bishop K, Alani A. Dens invaginatus. Part 2: clinical, radiographic features and management options. **Int Endod J** 2008;41:1137-54.

โลโบดอนเทีย และเด็นส์อินแวกจินต์สหลายตำแหน่ง: รายงานผู้ป่วย

คัมภีร์ ทรงกำพล

อาจารย์ ภาควิชารังสีวิทยา
คณะทันตแพทยศาสตร์ มหาวิทยาลัยมหิดล

ปรียพร ศรีมาวงษ์

อาจารย์ ภาควิชารังสีวิทยา
คณะทันตแพทยศาสตร์ มหาวิทยาลัยมหิดล

วราภรณ์ ชิตช่วงชัย

ผู้ช่วยศาสตราจารย์ ภาควิชาเวชศาสตร์ช่องปาก
คณะทันตแพทยศาสตร์ มหาวิทยาลัยมหิดล

ติดต่อเกี่ยวกับบทความ:

อาจารย์ ทันตแพทย์คัมภีร์ ทรงกำพล
ภาควิชารังสีวิทยา
คณะทันตแพทยศาสตร์ มหาวิทยาลัยมหิดล
ถนนโยธี ราชเทวี กรุงเทพฯ 10400
โทร 0-2203-6490
อีเมล: dtksk@mahidol.ac.th

บทคัดย่อ

ผู้ป่วยชายไทยอายุ 19 ปี จากการตรวจในช่องปากพบวิกลสภาพหลายอย่างด้านรูปร่างของฟันซึ่งพบได้น้อย และไม่มีโรคทางระบบอื่น วิกลสภาพของฟันเหล่านี้ประกอบด้วย ฟันรูปเสียมของฟันตัด ตัวฟันที่มีรูปร่างคล้ายฟันในกลุ่มสัตว์กินเนื้อตัวฟันที่มีหลายปุ่มภาพรังสีแสดงให้เห็นเด็นส์อินแวกจินต์ส จำนวน 10 ซี่ ซึ่งมีเงาโปร่งรังสีที่ปลายรากร่วมด้วย จำนวน 4 ซี่ และรูปร่างของรากฟันผิดปกติในฟันกราม