

## CASE REPORT

## Lobodontia: the unravelling of the wolf teeth

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### Abstract

Abnormalities of tooth shape and size generally coexist, as both are determined during the morphodifferentiation stage of odontogenesis. This report describes a case of lobodontia exhibiting multiple dental anomalies of which, “fang like” cuspids, multituberculism of molars, hypodontia along with severe generalized microdontia are distinctive. Radiographic features, diagnostic convolutions, differential diagnosis and clinical significance have also been highlighted.

**Keywords:** tooth abnormalities, hypercementosis, radiography.

### Introduction

Defective odontogenesis may result either from factors of environmental origin or from genetic abnormality. These defects may involve single tooth, groups of teeth, or an entire dentition [1]. However, involvement of the entire dentition is rare.

One such anomaly is lobodontia, which is defined as multiple anomalies resulting in a dentition resembling that of a carnivore (*lobos* = wolf) [2]. Although the term “lobodontia” was coined by Keene HJ and Dahlberg AA [2] in 1973, it was Robbins IM and Keene HJ [1] in 1964 who first reported such a case. This entity was later included in Witkop’s compilation of hereditary defects of dentin [3]. Gorlin RJ described lobodontia as a dental disorder, which should be of interest to the pediatric radiologist [4].

The distinctive features of this entity are pointed fang like cusps on canines and premolars [3], generalized reduction in crown dimensions and multitubercular molar crowns along with dens invaginatus, single conical molar roots with single canal, hypodontia and barrel or shovel shaped incisors [5]. A case report of an affected family in London [5] and a study in a large kindred residing in North America [2] suggested its mode of inheritance to be autosomal dominant. With a prevalence of 1:10<sup>6</sup> (one in a million), this condition seems to be extremely rare [2].

The aim of this report is to describe clinical and radiographic findings in a patient exhibiting multiple dental anomalies characteristic of lobodontia along with its clinical relevance.

### Patient, Methods and Results

A 32-year-old male reported to our clinic from a

rural dental camp with a chief complaint of multiple “small” teeth from childhood. As per the medical records, the patient was born after an uneventful full term pregnancy to non-consanguineous parents. His medical history was unremarkable in terms of any serious childhood illness or systemic abnormality. The patient revealed that his paternal side exhibited similarity to his dentition in terms of size and shape of teeth.

Systemic and extraoral examinations were normal. Intraoral examination revealed bilaterally missing permanent mandibular first molars that were extracted due to carious involvement. In addition, permanent mandibular right first premolar was missing, of which the patient was unaware. Crowns and root remnants of multiple retained deciduous teeth were easily distinguishable. Crowns of all the third molars were clinically visible. The patient had varying degrees of malocclusion ranging from cross bite to absence of interocclusal contact in the premolar region (Figure 1).



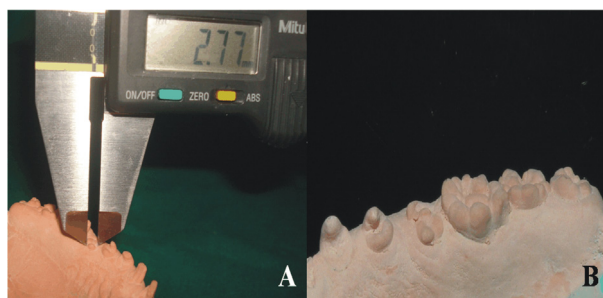
**Figure 1 – Generalized microdontia: frontal view.**

There was severe generalized microdontia (Table 1, Figure 2A) in conjunction with malformed crown structure (Figure 2B).

**Table 1 – Comparison of mesiodistal crown dimensions with an anatomic average\***

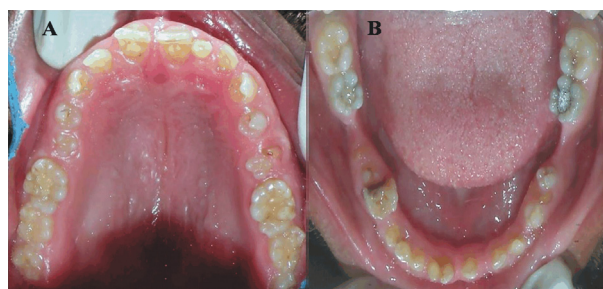
	Central	Lateral	Canine	First premolar	Second premolar	First molar	Second molar
<b>Maxillary</b>							
Average	8.5	6.5	7.5	7	7	10	9
Patient	5.73	3.37	4.05	3.5	2.77	7.84	6.86
<b>Mandibular</b>							
Average	5	5.5	7	7	7	11	10.5
Patient	2.85	3.23	3.25	4.09	3.02	NA	6.53

Measurements (in mm) taken at widest portion of clinical crown on diagnostic casts. \*Anatomic average taken from Ash MM Jr [6].



**Figure 2 – (A) Measurement of clinical crown on diagnostic cast using digital vernier caliper. (B) Palatal view of maxillary diagnostic cast showing abnormal dental morphology.**

The incisors, apart from their small size, displayed no other striking feature. The maxillary canines demonstrated marked tapering with its cusp tip pointing lingually giving it a “fang like” appearance. Maxillary first premolars exhibited a characteristic conical shape of the pronounced buccal cusp and a diminutive lingual cusp, whereas maxillary second premolars were severely dwarfed and had a trituberculate appearance. The mandibular premolars exhibited a prominent middle labial lobe with distinct developmental grooves separating the three buccal lobes. The maxillary and mandibular molars exhibited a characteristic “cup shape” appearance when viewed from the lateral aspect, whereas from the occlusal aspect they had a “rosette like” multitubercular appearance with numerous peripheral and interstitial cusps (Figure 3, A and B).

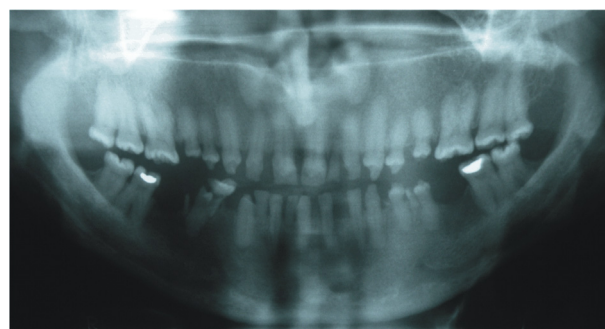


**Figure 3 – (A) Intraoral view of maxillary arch. (B) Intraoral view of mandibular arch.**

Vitality testing with ice and electric pulp tester (Digitest™, Parkell Inc., Farmingdale, NY, USA) elicited no response from mandibular left first and second premolars indicating loss of vitality in an apparently intact tooth.

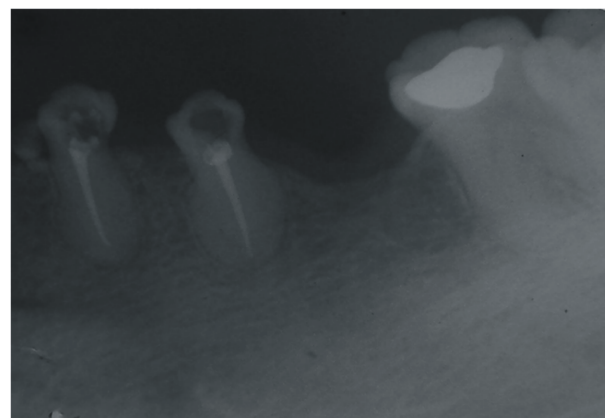
Panoramic radiograph confirmed the presence of multiple retained deciduous teeth and root stumps. Severe generalized microdontia could be appreciated with virtual absence of contact in between the teeth.

Exaggerated interdental spaces were evident especially in the anterior and premolar region. A large number of crown morphology aberrations in the form of loss of proximal contour of maxillary incisors, tapering maxillary canines, and dwarfed maxillary second premolars could be appreciated. Maxillary first premolars and mandibular premolars exhibited a conical crown giving an “ice cream cone” radiographic appearance. Maxillary second molars exhibited a pyramidal shaped single root. Presence of severe hypercementosis of the dentition in the premolar-molar region and radiopaque areas in the pulp chamber, suggestive of pulp stones could also be appreciated (Figure 4). Routine hematologic and endocrinologic evaluations were within normal limits.



**Figure 4 – Orthopantomograph showing abnormal crown morphology and hypercementosis in addition to generalized microdontia.**

Patient was made aware of a comprehensive treatment plan but unfortunately, due to financial constraints, only endodontic treatment for the premolars (Figure 5) and careful removal of the occlusal interferences was carried out so as to reduce trauma from occlusion and fracture of evaginated defects and the pointed cusps.



**Figure 5 – Periapical radiograph showing endodontic treatment in mandibular left premolars exhibiting microdontia and hypercementosis.**

## Discussion

The clinical and the radiographic findings along with the history in this case are most consistent with the diagnosis of lobodontia.

True generalized microdontia of the magnitude as was found in our case is uncommon, with sporadic cases reported worldwide. The maxillary second premolars were the most severely affected. The proband's dentition was categorized as having true generalized microdontia based on the Magnusson's criterion which defines a tooth as malformed if its mesiodistal dimension differs by two mm or more from its normal size [7]. Endocrinological investigations and medical records in conjunction with history excluded the possibility of hypopituitarism, radiation exposure or chemotherapy, which are commonly associated with true generalized microdontia. Other syndromes associated with microdontia were ruled out based on absence of extraoral and systemic findings in our case.

The molars exhibited multiple tubercular excrescences whereas maxillary second premolars had a trituberculate appearance. These aberrant structures most probably represent dens evaginatus or odontomes of the axial core type as described by Levitan ME and Himel VT [8]. Multituberculum along with peak shaped cuspids have also been reported in Ekman–Westborg–Julin syndrome [9], but the peculiar feature of macrodontia in the same differentiates it from lobodontia.

In addition to the above findings, presence of single rooted maxillary second molars, hypodontia of mandibular right first premolar and the familial occurrence of the trait helped us identify it as a case of lobodontia. The unique combination of dental findings in this case demonstrates the profound influence that morphogenetic disturbances in odontogenesis may have, on dental morphology and growth. Karyotyping and genetic counseling, although not done in our case, can be carried out to gain an insight into the genetic disturbance in order to help patients and their relatives better understand the probability of developing or transmitting it.

This condition apart from its diagnostic complexities has clinical implications as the evaginated defects and the pointed cusps might contain pulpal extension to various extents [10]. Traumatic occlusion, attrition or fracture of these evaginated defects, may lead to pulpal irritation and loss of pulp vitality as was found in our case. Therefore, early recognition of these defects and their appropriate management, depending on the extent of pulpal injury, should be carried out. Isolation during

endodontic therapy is also compromised due to the anomalous morphology and size of the teeth, which demands modification in rubber dam clamp selection and also the placement technique either by selecting a smaller clamp size or by stabilizing the clamp with visible light cured resin. Access opening becomes much more challenging owing to decrease in size of the tooth and also loss of occlusal anatomic details. As molars tend to present with single root and single canal, careful reading along with interpretation of the radiographs is essential in order to prevent mishaps during access opening. These teeth may also present with dens invaginatus type of defects, which may act as a pathway for pulpal invasion [5], therefore sealing of the same should be carried out prophylactically.

## Conclusions

Owing to the sporadic occurrence of lobodontia, there is a lack of awareness of this particular entity. The unique clinical and radiographic features of lobodontia as described above should help the dentist to diagnose it more consistently and also consider it in the differential diagnosis of cases presenting with non-syndromic generalized microdontia.

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