

Kissing Molars and Hyperplastic Dental Follicles: Report of a Case and Literature Review

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'Kissing' molars are impacted permanent molars that have occlusal surfaces contacting each other in a single follicular space, with roots pointing in opposite directions. It is deemed to be appropriate to medically investigate mucopolysaccharidosis (MPS) in patients presenting with kissing molars as kissing molars have been linked with MPS. The case of bilateral occurrence of kissing molars in an 18-year-old woman is described. Pathological analysis of the follicular tissue suggested hyperplastic dental follicles. Therefore, this case report analysed the association of impacted permanent teeth with hyperplastic dental follicles, following the review of seven documented reports describing such association.

Key words: *Kissing molars, impactions, mucopolysaccharidosis (MPS)*

Van Hoof was the first to describe 'kissing' molars in an intellectually challenged, 31-year-old man in 1973, but almost 18 years later, in 1991, Robinson et al proposed the term kissing molars to describe a similar condition in a 25-year-old man^{1,2}. The same year, Nakamura et al suggested the possible association of kissing molars with mucopolysaccharidosis (MPS) following his radiographic study of three relatively mature cases of MPS; one case, however, had no other detectable radiographic abnormality but the dental anomaly. Hence, Nakamura et al concluded that kissing molars can occur as an isolated event, but the possibility of MPS being present is only suggestive in such cases³. Nakamura's associative finding was further corroborated by McIntyre after evaluating and treating a 19-year-old woman who had kissing molars affected with pericoronitis⁴.

By definition, kissing molars are "impacted permanent molars that have occlusal surfaces contacting each other in a single follicular space with roots pointing in

opposite directions"⁵. To date, the literature documents 12 reports on this unusual condition¹⁻¹². Of these, five discuss its bilateral occurrence in both the jaws^{1,2,3,6,10} and seven describe its unilateral occurrence in the lower jaw only^{4,5,7-9,11,12}. While the incidence of kissing between the third and fourth molars is quite rare, such involvement between the second and third molars is relatively more common. So far, all reports describe their diagnosis and management in brief; one, however, suggests a classification system, in addition, for them⁹. We describe here a case of bilateral kissing molars in the mandible of an 18-year-old woman. We also analyse the association of impacted permanent teeth with hyperplastic dental follicles, following our review of seven documented reports describing such association.

Case report

An 18-year-old woman reported to the Department of Orthodontic and Dentofacial Orthopedics, College of Dental Sciences and Hospital, Davangere, for orthodontic therapy, and as a preliminary measure, an orthopantomogram and a lateral cephalogram were advised before orthodontic work-up. Upon radiographic evaluation, it was observed that the lower second and third molars were impacted, with their occlusal surfaces in mutual contact within a normally appearing follicular space, bilaterally. Although orthodontic uprighting of

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Fig 1 Orthopantomogram revealing bilaterally impacted lower second and third molars with their crowns facing each other in single follicular space. Also note the extreme position of the inferior neurovascular bundle at the lower border of mandible.

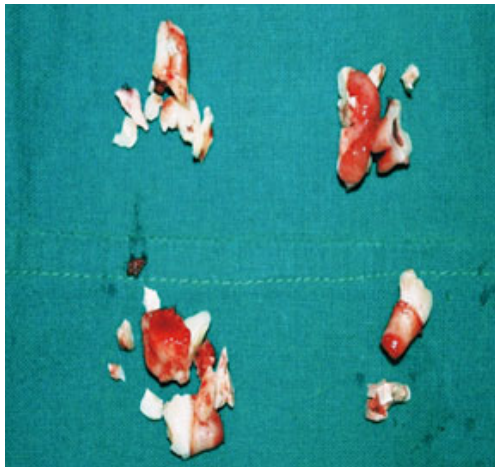


Fig 2 The four lower impacted teeth sectioned and removed.



Fig 3 Bone cavity (follicular space) seen following extraction of the impacted molars and enucleation of the follicular tissue.

both the second molars was possible and the same was suggested to the patient, the patient refused treatment, citing length of time and compliance to treatment as difficult to maintain. Therefore, the patient was referred to the Department of Oral and Maxillofacial Surgery for further management.

The patient had no associated medical problems. Clinically, a very diffuse swelling in the lower face on both sides was evident. The overlying skin was normal. There was no history of pain or other constitutional symptoms. Upon palpation, the swelling was bony hard and non-tender. Oral hygiene was fair, with mild generalised gingivitis present. The patient had permanent dentition and a class I molar relationship; however, the lower second and third molars were missing bilaterally with mild buccal cortical expansion evident. The alveolar ridge in the second molar region appeared swollen up to the retromolar trigone but was non-tender. The upper third molars were also missing.

An orthopantomogram revealed impacted second and third molars, whose occlusal surfaces were in intimate contact within the radiolucency (follicular space), but were limited to the cemento-enamel junction. The maxillary third molars were locked (impacted) beneath the second molar crowns (Fig 1). The inferior mandibular canal was considerably close to the lower border of the mandible on both sides, making postoperative neurosensory deficit a strong possibility, more so with the left. Also, the deep-seated impactions, along with the presence of the space, appeared to reduce the cross-sectional area of the jaw in that region, raising serious concerns of intraoperative jaw fracture. We also explained to the patient that a CBCT would be necessary to actually estimate such possible risks but the patient refused to comply.

Surgical removal of the impacted molars was performed under general anaesthetic (Figs 2 and 3). Following an odontectomy, the follicular tissue was enucleated out and sent for pathological analysis. Postoperative recovery was uneventful. Haematoxylin and eosin stained sections demonstrated a fibromyxoid background with areas of dense connective tissue capsule having numerous inactive-looking small odontogenic epithelial islands and areas of calcification in the stroma (Fig 4). Thus, the diagnosis of hyperplastic dental follicle was established. Also, the intracellular presence of MPS was ruled out using 2% toluidine blue.

Although the association of kissing molars with mucopolysaccharidosis is suggestive, the latter is known to predominantly affect the eye and the skeletal system. Therefore, to exclude such abnormalities, additional investigations such as a radiograph of the

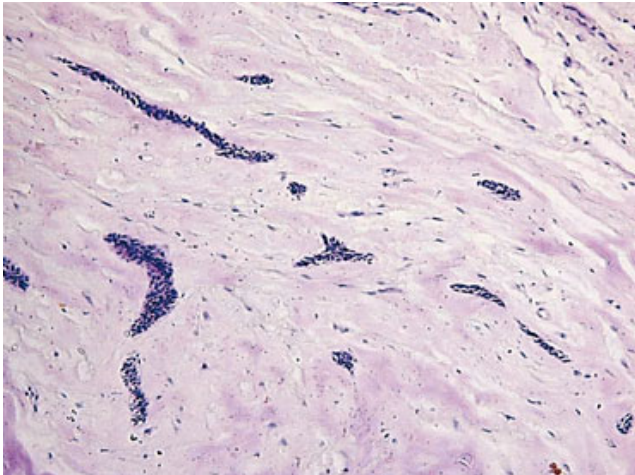


Fig 4a Photomicrograph showing numerous rests of odontogenic epithelium (indicated by black arrows) dispersed within the fibrous connective tissue (dental follicle), suggestive of hyperplastic dental follicle (10 x magnification).

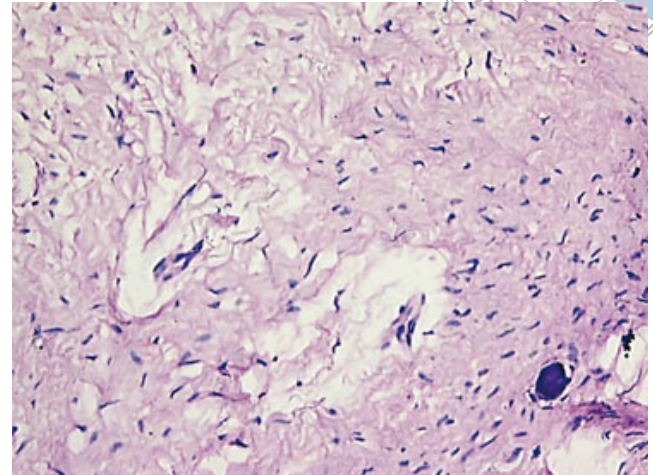


Fig 4b Photomicrograph showing areas of calcifications in the stroma (10 x magnification).

lateral skull, the elbow (anterioposterior and lateral), the pelvis (anterioposterior) and a chest radiograph were performed, which revealed no abnormality; an ophthalmology consult ruled out corneal clouding. Hence, the possibility of MPS was excluded in our case.

Discussion

Although Robinson et al coined the term of kissing molars in 1991, the credit goes to Van Hoof for having described this rare condition in 1973^{1,2}. To date, 12 reports have been described in the literature with regard to the incidence, associated pathologies, differing treatments and their respective outcomes (Table 1)¹⁻¹².

Many theories explaining altered tooth position (delayed/noneruption/impaction) have been suggested, but the exact etiology is yet to be determined¹³. More importantly, however, Nakamura et al, who, after observing multiple 'rosetting' of molars in 3 of 4 patients with MPS, concluded that the isolated event of rosetting may not rule out the possible presence of MPS in such patients and, therefore, such cases call for appropriate medical investigation.³ Furthermore, Cawson et al suggested MPS as a possible etiological factor in multiple tooth impactions that he observed in patients affected with MPS¹⁴.

MPS results from a quantitative or qualitative deficiency of lysosomal enzymes required to break down glycosaminoglycans. Over time, these molecules accumulate within the cells, blood and connective tissue, resulting in permanent deleterious effects¹⁵. Widespread common radiographic abnormalities include J-shaped sella turcica, defective anterior vertebral body devel-

opment, costal widening, an underdeveloped superior acetabular region, coxa valga and proximal tapering of the long bones. Although the combination of these abnormalities is highly suggestive of the condition, none are diagnostic.¹⁵ We did not consider the possibility of MPS in the present case as the phenomenon of kissing molars was a lone radiological finding without any evidence of follicular enlargement (Fig 1) or widespread radiographic abnormalities, and tissue microscopy suggested hyperplastic dental follicles without the intracellular presence of MPS (Fig 4).

The normal pericoronal radiolucency is considered to be in the range of 2 to 3 mm; however an increase in the space should be viewed with suspicion^{16,17}. Recent reports suggest that unerupted/impacted teeth including kissing molars may have the propensity to form developmental odontogenic cysts, most commonly the dentigerous cyst that manifests as an asymptomatic unilocular radiolucency^{8,9,10}. However, the radiographic differential diagnosis may also include other pathologic entities such as odontogenic keratocyst, unicystic ameloblastoma, dental follicle (DF) with no disease, hyperplastic dental follicle (HDF) and adenomatoid odontogenic tumour^{18,24}.

Of relevance is the HDF (synonym: peri-follicle fibrosis), a rare lesion that is often confused with odontogenic fibroma, but distinct clarifications between the apparent histological similarity of hyperplastic dental follicle with that of simple and central odontogenic fibromas were given by Gardner in 1980¹⁹. Sandler et al also suggested the possible presence of calcifications in hyperplastic dental follicles, which he termed calcified (CHDF).²⁰ Although calcification has been reported

Table 1 Cases of kissing molars as reported in the English literature

Author and year of reporting	Gender/age	Symptoms	Radiographic presentation	Medical problems	Treatment	Postoperative complications and follow-up	Histopathological findings
Van Hoof, 1973 ¹	M/31	None	B/L mandibular impacted	Intellectually challenged	-	-	-
Robinson et al, 1991 ²	M/25	None	B/L mandibular impacted	None	-	-	-
Nakamura et al, 1992 ³	M/25	None	B/L 'rosette' formation in both jaws	MPS	-	-	-
	M/1½	None	-	MPS	-	-	-
	M/17	None	B/L mandibular 'rosette' formation	MPS	-	-	-
	M/21	None	B/L 'rosette' formation in both jaws	-	-	-	-
Manani A, 1996 ⁷	M/29	None	U/L mandibular impacted	None	-	-	-
McIntyre, 1997 ⁴	F/19	Pericoronitis	U/L mandibular impacted	None	Ex under GA	Trismus and dry socket	-
Bakeen and Baqain, 2005 ⁶	M/23	Facial pain	B/L mandibular impacted	None	Ex under GA	-	No evidence of disease
Koerner, 2006 ¹²	M/39	Pericoronitis	U/L mandibular impacted	-	Ex + Iliac grafting + MMF under GA	-	-
Krishnan, 2008 ⁸	F/36	Left-sided mandibular swelling including alveolar ridge	U/L mandibular impacted	None	Ex under LA	None	Dentigerous cyst
Buffano and Gallesio, 2009 ⁵	M/42	Pain in the right retro-molar region	U/L mandibular impacted	None	Ex under LA; socket filled with granular calcium sulphate, antibiotic, corticosteroid & NSAID	None	No evidence of disease
Giraldi Neto et al, 2012 ¹¹	F/30	None	U/L mandibular impacted	None	Ex under LA	None	No evidence of disease
	F/22	None	U/L mandibular impacted	None	Ex under LA	None	No evidence of disease
Sà Fortes et al, 2012 ¹⁰	M/33	None	B/L mandibular impacted	None	Ex under LA; preoperative corticosteroid, antibiotic & NSAID	None	Dentigerous cyst
Aydin Gulses et al, 2012 ⁹	F/26	-	U/L mandibular impacted	-	Surgical removal	None	-
	F/32	-	U/L mandibular impacted	-		None	Dentigerous cyst
	F/37	-	U/L mandibular impacted	-		None	-
	F/22	-	U/L mandibular impacted	-		None	Granulomatous changes of the follicle
	F/20	-	U/L mandibular impacted	-		None	-
	M/44	-	U/L mandibular impacted	-		None	-
	M/23	-	U/L mandibular impacted	-		Paresthesia of IDN (4 months)	Granulomatous changes of the follicle
	M/16	-	U/L mandibular impacted	-		Paresthesia of IDN (6 months)	Dentigerous cyst
	M/27	-	U/L mandibular impacted	-		Paresthesia of IDN (3 months)	Dentigerous cyst
Present case study	F/18	None	B/L mandibular impacted	-	Preoperative corticosteroid, antibiotic and NSAID; surgical removal under GA; socket filled with a cube of gel foam	None	Hyperplastic dental follicle

Table 2

Author and year of reporting	Gender/age	Symptoms	Radiographic presentation	Medical problems	Treatment
Sandler et al, 1988 ¹⁹	M/18	Asymptomatic, firm, tuberous expansions in the mandibular vestibule bilaterally in the premolar regions; well-localized and 1-2 cms in diameter; similar observation in maxillary left 2nd premolar region	1, 2, 13, 15, 16, 17, 18, 20, 21, 28, 29, 31 and 32	Unremarkable	Abundant calcified material in the whorled area of connective tissue and in the hyalinised tissue surrounding the odontogenic epithelium
Lukinmaa et al, 1990 ²⁵	M/24	Unerupted left lower 2nd premolar and 2nd molar; congenitally missing lower left 1st premolar and 3rd molar; H/o extraction of lower 1st molar 12 years before; retarded eruption of upper left 2nd premolar	18 and 20	Enamel hypoplasia with defective crown & root development	Presence of intensely basophilic, concentric calcified bodies arranged in groups and large numbers (type A calcification); presence of epithelial islands, some resembling odontogenic epithelium and others demonstrating squamous differentiation and tendency toward keratinisation
Gardner et al, 1995 ²⁴	M/26	Unerupted left lower 2nd and 3rd molars	17 and 18	Not mentioned	Connective tissue: dense, moderately cellular and fibrous with scattered, numerous odontogenic epithelial rests and whorled structures; Type A calcifications, dominant; type B calcifications elsewhere;
	M/40	Seven deeply embedded teeth	7 teeth		Similar to case 1 with type B calcifications, dominant; type A calcifications interspersed in type B
Gomez et al, 1998 ²²	M/15	Unerupted 1, 2, 5, 6, 7, 11, 12, 15-22, 27-32; agenesis of 4 and 13	1, 2, 5, 6, 7, 11, 12, 15-22 and 27-32	Unremarkable	Connective tissue: hyperplastic, dense and fibrous with increased deposits of type A and B calcifications surrounding cords and islands of clear odontogenic epithelium; dystrophic calcifications in the pulp and dentin of the removed teeth
Walker et al, 2004 ²³	F/6	Missing right mandibular primary 2nd molar (T)	T (hyperplastic dental follicle with an incipient adenomatoid odontogenic tumor)	Not mentioned	Connective tissue: well-circumscribed, lightly-to-heavily collagenized with hypocellular fibroblastic proliferation and moderate vascularity; duct-like epithelial structures and several spindle-shaped epithelial islands associated with several foci of irregular calcifications
Sun et al, 2010 ²⁰	F/12	H/o extraction of 13, 6 months earlier; buccal bulges in 22 and 27 regions, depressible; congenitally missing 17 and 32; impacted 6, 11, 22 and 27;	6, 11, 22 and 27	Unremarkable	Considerable ground substance and multiple odontogenic epithelial rests (juxtaepithelial hyalinization)
	M/15	Similar findings; impacted 6, 11, 22 and 27	6, 11, 22 and 27	Unremarkable	Same as in case 1
Cho et al, 2011 ²⁶	M/11	7 impacted teeth; paramolars, congenitally missing mandibular central incisors	2, 6, 11, 15, 18, 19 and 31	Not mentioned	Connective tissue: myxoid to loose fibrous with type I and II calcification
	M/14	6 impacted teeth; H/o of extraction of two supernumerary teeth at 8 years of age	2, 6, 11, 15, 18 and 31	Not mentioned	Connective tissue: myxoid to loose fibrous with type I and II calcification
	M/11	4 impacted teeth; buccolingual expansion of the jaws; paramolars	2, 15, 18 and 31	Not mentioned	Connective tissue: myxoid to loose fibrous with type I calcification
	M/15	7 impacted teeth; a supernumerary tooth observed between the left mandibular 1st and 2nd premolars	2, 4, 13, 15, 18, 20 and 31	Not mentioned	Connective tissue: dense fibrous having type 1 calcification with Liesegang ring
	M/17	3 impacted teeth only	2, 15 and 19	Not mentioned	Connective tissue: dense fibrous with type I and II calcification
Present case study	F/18	Clinically missing 8s and lower right and left 2nd molars; mild buccal bulge in the lower right and left 2nd molar regions	17, 18, 31 and 32	Unremarkable	Fibromyxoid background and areas of dense connective tissue having numerous inactive-looking small odontogenic islands with areas of calcification in the stroma present

as a common microscopic finding, the condition of multiple unerupted/impacted teeth and enlarged dental follicles (calcified) within the jaws is extremely rare, with exclusive male predilection and premature calcifications, different to HDF^{21,27}. Kim et al have suggested possible mechanisms responsible for the development of HDF. Yet, there is no definite explanation as to why they are caused^{21,22}. However, a more recent report suggested a genetic tendency as a possible cause²¹.

To date, seven reports on HDF including multiple calcifying (MCHDF) are documented in the literature (Table 2); eleven males and two females presented with impacted teeth and the posteriors were more frequently involved i.e. 67 of the 85 teeth^{20,21,23-27}. In six reports, calcified material was noted in the connective tissue capsule of the follicle, besides discussing and correlating other microscopic findings with histologically-similar odontogenic tumours^{20,23-27}. However, the remaining report noted the unusual absence of calcifications in the follicle in association with a familial tendency. Also, another report for the first observed dystrophic calcifications in the dentin and pulp of extracted (unerupted) teeth^{21,23}.

Teeth failing to erupt or that get impacted due to a HDF may appear similar to those in regional odontodysplasia and other odontodysplasias such as amelogenesis imperfecta, rough hypoplastic type, dental follicular haematomas and gingival hyperplasia^{28,29}. This may be of practical importance to dentists as they frequently come across and surgically treat impacted teeth in day-to-day practice. Also of diagnostic importance is the precise histological distinction between HDF and other odontogenic tumours (odontogenic fibroma, simple and central types), especially when calcifications are present. Studies have estimated their presence in one-third of HDF, but more recent reports, describing possible intra-follicular calcification processes, observe higher incidence rates of calcification in the follicle^{27,30-33}.

Thus far, none of the available literature on kissing molars describes orthodontic guidance of the permanent mandibular second molars into ideal occlusal position, even though such treatment could be considered an alternative in such patients¹⁻¹². While, on the one hand, time and compliance to such treatment may appear crucial, on the other, the literature advocates odontectomy as a decisive form of treatment, even for these unusual impactions. However, treatment acceptance is always a matter of personal choice.

Of significance is the accurate assessment of surgical difficulty plays a critical role in the management of impacted permanent teeth including kissing molars^{34,35}. Moreover, other factors such as an accompanying medical condition – either related or unrelated, the presence of an associated ‘local’ pathology (not always), the choice of anaesthetic technique and finally the knowledge and expertise of the surgeon also merit equal consideration, if not greater.

Conclusion

We have comprehensively reviewed the literature on kissing molars. In addition, the clinical and radiological features of MPS have been reviewed in brief and their relevance to kissing molars has been analysed. Despite recent advances in imaging, a conventional radiograph, even today, has significance in contemporary clinical practice from a diagnostic standpoint, as such anomalies may pose as an incidental finding. Additionally, patients presenting with this rare condition seek appropriate medical investigation before embarking on surgery.

As for any impacted tooth, surgical removal remains the treatment of choice, and it may be suggested to have the enucleated follicular tissue microscopically analysed to determine the probable cause of delayed tooth eruption or impaction. In a way, the dentist or the treating surgeon would be wary of the possible presence of disturbances affecting tooth formation and/or development and eruption.

Acknowledgement

The authors would like to thank Dr Rajeshwari G Annigeri, Professor and Head, Department of Oral Medicine and Radiology, and Dr Mandana D Donoghue, Professor and Head, Department of Oral and Maxillofacial Pathology, for having interpreted the findings of the case of kissing molars in an 18-year-old woman, and for having referred the same to our department for definitive management. Dr Reshma Hammannavar Babu, Dr Anubhav A Jannu and Dr Mayur M Shingade are also thanked for having helped prepare and astutely assess this manuscript.

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