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Case Report

Shovel Incisors, Dens Invaginatus and Multiple Protostylid in a Teenage Girl: A Case Report and Literature Review

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Abstract

Shovel-shaped incisors (SSIs) are teeth with thick marginal ridges adjacent a deep lingual fossa. Dens invaginatus (DI) is a malformation of teeth most likely causing from an infolding of the dental papilla during development of tooth. Protostylid is used for any additional cusps on the buccal surface of maxillary and mandibular premolars and molars. Morphological dental anomalies of the permanent teeth are seen to occur frequently; however, multiple anomalies in a single patient are relatively uncommon. A 19-year-old non-syndromic healthy Malay teenage girl who came for correction of misaligned teeth presented with an amalgamation of dental anomalies consists of SSIs and DI of the upper right lateral incisor, protostylid on bilateral upper maxillary molars, unilateral protostylid of right mandibular premolar, and Class III incisor relationship. The intraoral radiograph of tooth 12 revealed radiopaque invaginations from the palatal pits confined to the crowns of the teeth and there was no pulpal extension into the accessory cusp of both maxillary permanent second molars. The definitive treatment included sealing and build up the incisor DI tooth with composite, fissure sealant of deep fissure protostylid on bilateral maxillary second molars, close monitoring of the prostostylid on lower right premolar and fixed orthodontics appliance for the management of Class III incisor relationship. Early diagnosis, comprehensive treatment and regular follow-up are crucial to prevent any future consequences that may be associated with these dental anomalies.

Keywords: Dens invaginatus, dental anomalies, management, protostylid, shovel shaped

Introduction

Human teeth may display alterations and differences in morphological characteristics. Such variations may be observed on the crown either in the type of anomalous cusps or bizarre forms of tooth anatomy. Structural anomalies may just affect one tooth, a set of teeth, or the whole dentition.[1,2] The concurrent occurrence of abundant anomalies may be associated to certain syndromes, particularly in patients with chromosomal modifications blended with multi-systemic variations.[3]

Shovel-shaped incisors (SSIs) was first reported by Mühlreiter in 1870 (Mühlreiter 1870) whereby the number of cusps on maxillary molars was established in 1913

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by Keith.[4] SSIs are distinguished by a robust marginal ridge surrounding a deep lingual fossa described by a pronounced hollow area on the palatal surface of the teeth surrounded with a well-defined enamel boundary.[5]

SSIs is prevalent in Asians, Asian-derived peoples, and Indigenous Americans and is unusual or lacking in African and European inhabitants.[6] The condition may affect 9% of the incisors (4% of the central incisors and 5% of the lateral ones).[7] Hrdlicka[8] has classified four differences: (0) no shovel; (1) evidence of shovel; (2) semi-shovel shaped, and (3) noticeable shovel shape.

SSIs may lead to disturbances in occlusion, plaque accumulation, caries, periodontitis and trauma to the tongue.[7] Attrition may show the central pulp horn, so that conservative treatment, reduction, coverage and endodontic therapy may all perform a part. Any deep fissures or invaginations should be filled.[9] In orthodontics, in certain occasions, SSIs do not allow orthodontic reduction of increased overjet and therefore parts of the marginal edges need to be reshaped[7] and reconstruction of crown morphology for aesthetic purpose.[9]

Dens invaginatus (DI) is a developing abnormality rising from invagination/infolding of the crown and/or the root surface earlier to hard tissues mineralization occurs.[10] The invagination starts in the crown and may continue into the root.[10] The prevalence of DI on permanent dentition ranges from 0.3–10%[11] and most frequently found in the maxilla, which is the lateral incisors, followed by the central incisors, while it is rare in the canines, premolars, and molars.[12–14] The exact cause of DI remains unknown.[15] Oehlers in 1957, classified DI into three types corresponding to the depth of infiltration and interaction with periapical tissues or the periodontal ligament.[16] Type I is an enamel-lined minor invagination occurring within the coronal part of the crown without an extension outside the cemento-enamel junction. Type II is an enamel-lined invagination expanding into the root outside the cemento-enamel junction and continues as a blind sac. Type III is an invagination probing via the root to create an added apical or lateral foramen.[16] The most observed DI was Type I (69.8%–79%), followed by type II (15%–26.6%) and type III (3.4%–5%).[15]

General features of teeth with DI include peg shape, conical morphology, incisal notching, increased mesiodistal and labiolingual diameter, and the presence of an enlarged palatal cingulum or cusp.[15] Coexistence of DI and other developmental dental defects such as double teeth, SSI, multi-rooted teeth and talon cusp were also reported.[13] DI is an anomaly of great clinical significance, sometimes causing aesthetic issues especially on the maxillary teeth, occlusal interference compromised overjet and occlusal stability.[12] The objective of treatment is to preserve pulp vitality as much as possible.[12] Sequelae of undiagnosed and untreated invaginated teeth include pulp pathosis, eruption delay, cysts and internal resorption.[12] Depending to the connection of the infolding and root canal system, the infolding can either be managed conservatively or in arrangement with the root canal system if both are affected.[14] The treatment of choice is rather oriented towards preventive measures; sealing the invagination with composite or fissure sealing resin when the pulp is not infected, endodontic treatment or extraction.[14] Extraction is indicated in certain situation such as the occurrence of mesiodens, non-restorable tooth, very wide open apex, inaccessibility of conservative or surgical endodontic treatment.[14]

A protostylid is an extra cusp found on the mesial half of the buccal surface on the maxillary and mandibular molars.[5] The earliest explanation was presented by Dahlberg in 1950[17] who described it as an anomalous cusp on the buccal surface of both the upper and lower premolars and molars. The prevalence of protostylid varies in different ethnic groups, like 18.5% in Japanese people, 37.5% in Chinese population, while 5.4 % in Colombian population.[18] Asian population are reported to have a prevalence rate of 2%.[19] The protostylid has a higher prevalence in the second deciduous lower molars, followed by the first permanent lower molars.[5,20] If the protostylid is expressed in the deciduous dentition, it will be expressed in the permanent dentition.[20] The etiology of supplemental cusp structure or unusual appearance is undetermined.[19] It was said that these characteristics are possibly due to dental lamina overactivity, but at current, the atypical form of the teeth was thought to be caused by MSX and PAX genes.[21] There were five portions of the protostylid arrangement which was suggested by Snyder et al(1969)[22]; (0) no cusp; (1) pit; (2) eminence; (3) elevation; (4) cusp.

There are few problems that may arise due to protostylid such as potential sites for plaque retention and caries, sensitivity and devitalization due to fracture or attrition of protruded portion of cusp leading to pulpal exposure.[19] Besides, these anomalies will pose difficulty in the placement of crowns, interference with cementation of brackets, bandings, and alignment of wires during orthodontic treatment.[23] As a result, protostylid may be removed by ameloplasty after the pulpal extension into the protostylid is ruled out.[23]

The aim of this case report is to highlight the importance of appropriate diagnosis and a multidisciplinary approach in the management of SSIs associated with DI and multiple protostylid in a teenage female patient requiring orthodontic treatment. The concurrent occurrence of SSIs, DI, and multiple protostylid in a single non-syndromic patient is undoubtedly unique and probably rare.

Case Presentation

A healthy 19-year-old Malay female teenage came to the comprehensive dental clinic at Faculty of Dentistry Universiti Teknologi MARA for the correction of misaligned teeth. The family and medical histories were non-contributory. The extraoral examination revealed a concave facial profile and a shallow mentolabial sulcus. Upon intraoral examination, no soft tissue abnormalities were observed, and she presented with fair oral hygiene and low caries risk. The patient demonstrated with Class III incisor relationship, ICDAS 34 on tooth 22, severe crowding of the maxilla, rotated tooth 15, 24, and 25, bilateral posterior crossbite, diastema, and lower dental midline shifted to the right 2mm (Figs. 1a-c). The Index of Treatment Need (IOTN) was 4d with aesthetic component 8.

A bilateral protostylid was present on the buccal area of the bilateral upper right and left second permanent molars (17 and 27) observed in the occlusal plane (Fig. 1b). It was a triangular structure with the apex towards occlusal surface measuring about 4 mm cervico-occlusally on buccal surface and 3 mm mesiodistally on both tooth 17 and 27 (Figs. 2a, b). Protostylid on tooth 44 showed eminence of cusp without groove outline (Fig. 2a). Protostylid on tooth 17 was touching the mesiobuccal cusp of tooth 47 whereby protostylid on tooth 27 did not occluding the lower teeth.

The periapical radiograph of tooth 12 demonstrated radiopaque invaginations from the palatal pits restricted to the crowns (Fig. 3a). These invaginations were of the enamel-lined limits of the crown and not extending outside the cemento-enamel junction. The pulp sensibility tests (electric pulp test and cold test) showed the tooth was vital. The bitewing radiographs revealed there

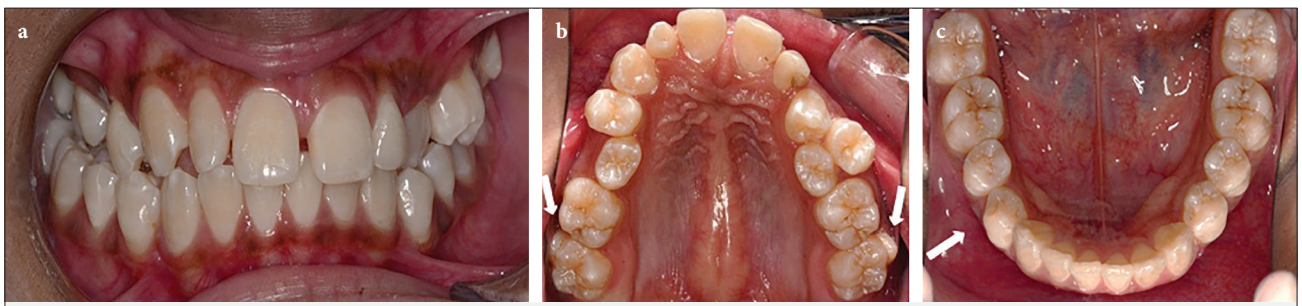

Figure 1. (a) The frontal view of the dentition of the patient. (b) The photo of the upper occlusal view of protostylid on the right and left maxillary second permanent molars. (c) The photo of the lower occlusal view of protostylid on the right mandibular first premolar

Figure 2. (a) The buccal view of protostylid on the right maxillary second permanent molars and right mandibular first premolar. (b) The buccal view of protostylid on left maxillary second permanent molars

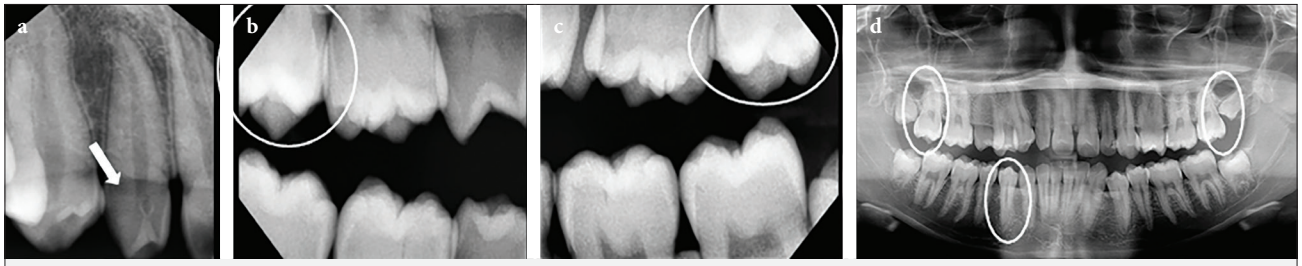

Figure 3. (a) The periapical radiograph of tooth 12 shows Type I DI. The invagination of the surface enamel above the cement-enamel junction is indicated using the white arrow. (b, c) The right and left bitewing radiographs showing there is no pulpal extension into the accessory central cusp of both right and left maxillary second permanent molars (white circle). (d) The orthopantomogram showing a permanent dentition stage

is no pulpal extension into the accessory cusp of both right and left maxillary permanent second molars (Figs. 3b, c). The orthopantomogram showing the unerupted tooth 18 and 28 is in the mesio-angular inclination and no obvious pulpal extension of into the accessory cusp of right permanent first lower premolars (Fig. 3d). The lateral cephalometry analysis showed Class I skeletal pattern and bimaxillary protrusion (ANB=1.5).

The final diagnosis was protostylid on tooth 27 (grade 4), 17 (grade 4) and 44 (grade 2), shovel tooth with DI type I on tooth 12, ICDAS 34 on tooth 22, and mesio-angular inclination of the unerupted teeth 18 & 28. The orthodontics diagnosis was Class III incisor relationship on a Class I skeletal base with an increased lower third vertical proportion. The patient was in permanent dentition with Class II molar relationship on right and Class I molar relationship on the left site, severe crowding of the maxilla with a buccally ectopic position of tooth 24 and mild crowding of mandible, 90-degree rotation of tooth 15, 24, and 25, bilateral posterior crossbite, upper midline diastema and upper midline shifted to the right 2mm. The fissure sealant and topical fluoride were placed on protostylid on the upper permanent second molars (tooth 17 and 27) (Figs. 4a-d). Topical fluoride applied on protostylid tooth 44. Restorative crown modification with composite (3M™ Filtek™ Universal Restorative) was performed on shovel-shaped tooth 12 and a composite filling on 22 (3M™ Filtek™ Universal Restorative) was carried out (Figs. 5a-c). Treatment objectives for orthodontic included correction of displacement and rotated teeth, correction of bilateral posterior crossbite, correction of molar relationship, correction of upper midline shift, and establishment of a stable occlusion. The proposed treatment plan was non extraction, upper arch expansion using arch wire coordination to correct a bilateral posterior crossbite, and alignment and levelling of the arches plan were formulated with fixed appliances using 0.022-inch slot MBT bracket systems. To reinforce the anchorage, temporary anchorage devices on the buccal aspect mesial to 16 and 26 are planned to prevent the mesial movement of the molars and retention with a Hawley appliance. A six-month review showed that the tooth was clinically and radiographically asymptomatic. The patient is under regular follow-up and waiting for orthodontics intervention.

Discussion

The concurrent existence of multiple dental anomalies has been reported previously, particularly in patients with chromosomal abnormalities who may present with multisystem abnormalities.[24] Multiple dental anomalies have also been reported in patients without generalized abnormality or disease as presented in this case. Furthermore, many studies reported the association between dental anomalies and skeletal malocclusion, and their genetic background is widely known.[25] To our best knowledge, no scientific literature has previously reported on a similar presentation among these types of dental abnormalities in a single non-syndromic patient.[6] Hence, the combination of multiple dental anomalies found in this case is considered unique and differs from previously published cases.

The management of a teenage patient with malocclusion and dental anomalies demands a thorough diagnosis and a treatment plan which involves aesthetic, occlusal, and functional judgment.[26,27] Therefore, upon diagnosis of such dental anomalies, the prognosis of the tooth in terms of long-term stability, restorability, periodontal status, and the patient's overall orthodontic condition must be taken into account.[28,29] A periodic observation schedule with orthodontic treatment to align the dentition, fissure sealant on the protostylid teeth with deep fissure, combined with the cosmetic restoration of the shovel-shaped lateral incisor would be an effective treatment plan as shown in the presented case report. The key to treatment is an early diagnosis combined with a treatment plan of continuity and long-term objectives.[26,30]

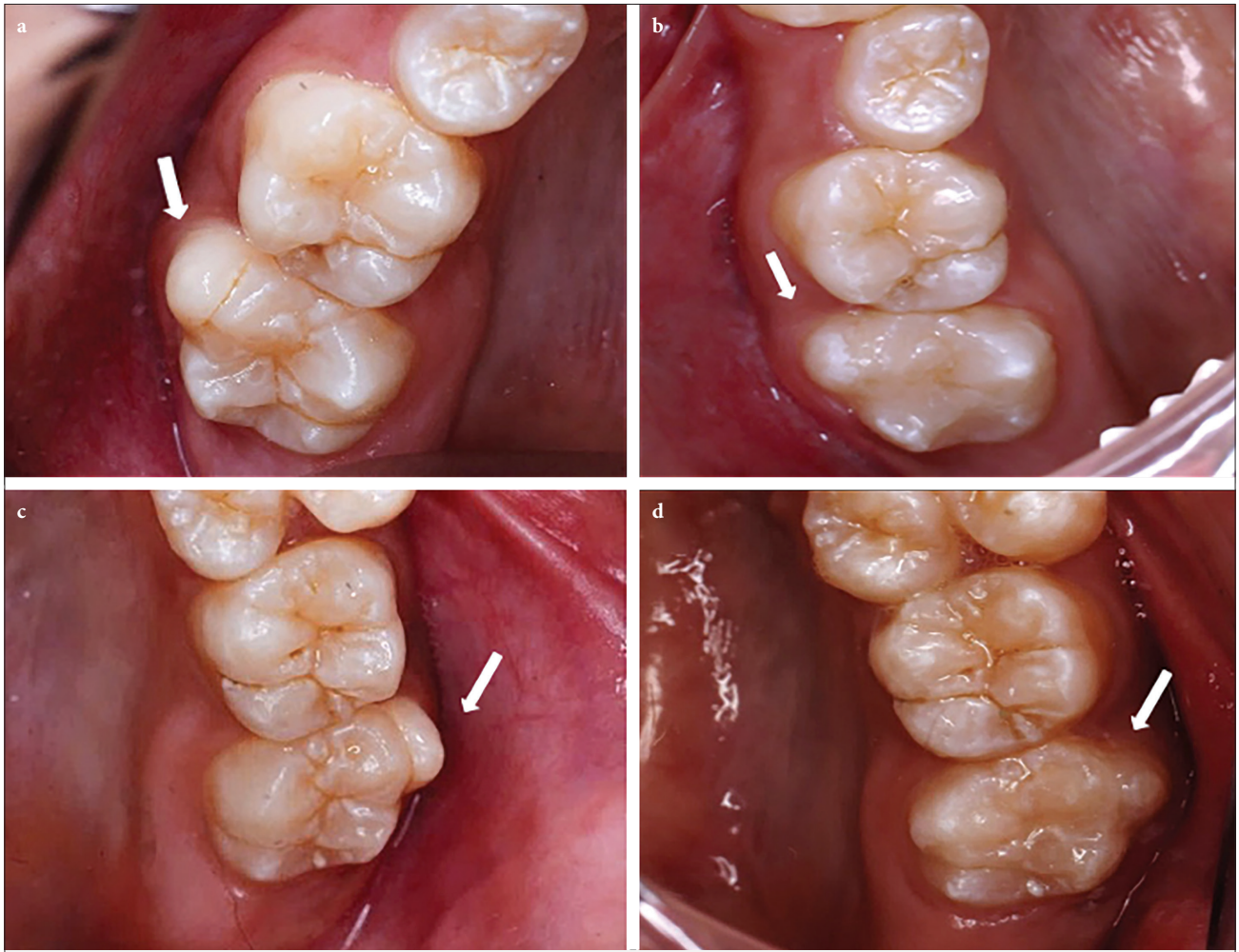

Figure 4. (a) Preoperative photo and (b) Postoperative photo of the occlusal view of protostylid on the right maxillary second permanent molars after placement of fissure sealant (c) Preoperative photo and (d) Postoperative photo of the occlusal view of protostylid on the left maxillary second permanent molars after placement of fissure sealant (close image)

SSIs, so called because the lingual marginal ridges enclose a fossa and giving the appearance of a "coal shovel", has been considered a Mongoloid trait of the dentition since Hrdlička[8] reported. Shovel trait frequently occurs in all Mongoloid groups, including Bunun aborigines, Chinese, Eskimos, and American Indians.[17] A usually large, unusual crown, peg or barrel-shaped crown, or a deep foramen caecum may clinically accompany dens invagination.[30] The altered or conical shape of the shovel incisors may lead to significant aesthetic discomfort.[30] Also, with such a small clinical crown, it becomes very difficult to bond the bracket in the center and achieve the desired tooth movement for orthodontic management.[7] In terms of management of SSIs, studies recommended either to close or open the space.[7,8] The SSI can be extracted and the resultant space closed. However, this will often give a narrow aesthetic smile. The other option is to open or maintain the space and create a proper space for a normal sized lateral incisor.[7] The aim of treatment should be to give the lateral a normal looking crown structure that appears unremarkable.[7] As sufficient space exists between the central and canine to allow restoration, the direct placement of tooth colored restoration to alter the shape, length and increase the surface area for placement of orthodontic bracket was carried out in the present case.

DI is more frequent in shovel-shaped incisors as seen in the present case.[31] The most common type of DI is the type I.[31] The teeth most affected are the maxillary lateral incisors.[32] Bilateral occurrence as high as 43% has been reported by Hülsmann.[32] DI can sometimes cause aesthetic issues, occlusal interference compromised overjet and occlusal stability.[13] Since the SSIs with DI did not irritate the tongue during speech and mastication and did not interfere with the occlusion, no occlusal adjustment was needed.[13] DI also considered

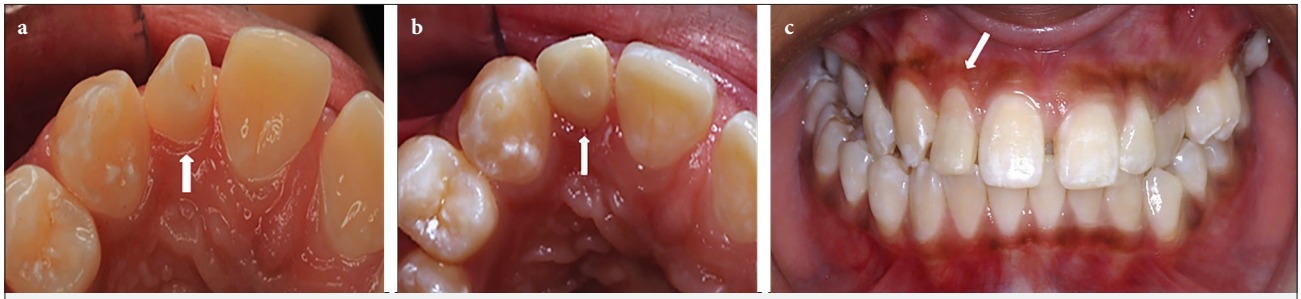

Figure 5. (a) Preoperative photo (b) Postoperative photo of composite build-up of shovel shaped maxillary right lateral incisor (close image). (c) The photo is showing the frontal view of composite build-up of shovel shaped maxillary right lateral incisor

increasing the risk of caries and pulpal pathology.[26] Thus, maintaining an optimum standard of oral hygiene is crucial especially for a patient undergoing orthodontic treatment as shown in this case report. Treatment of DI ranges from the conservative restoration of the opening to endodontic treatment or extraction.[12,26] Treatment decisions should be based on an in-depth pre-operative evaluation of the gravity and complexity of the invagination.[1,12] The invagination frequently communicates with the oral cavity, transmitting the access of irritants and microorganisms directly into the pulpal tissue.[27] Sometimes fine canals expand between the invagination and the pulp chamber causing in pulpal and periapical pathology even without dental caries.[28] In our case, the invagination was minimal and confined to the crown and thus, the flowable composite resin was a good material to fill the entrance of the invagination.[13] The possible consequence of undiagnosed and untreated invaginated teeth are pulp pathosis, late in eruption, cysts, and internal resorption.[29] Extraction is indicated in certain situations such as the presence of mesiodens, non-restorable tooth, very wide open apex, inaccessibility of conservative or surgical endodontic treatment.[29] Alternatively the resulting space can be orthodontically closed (to avoid a prosthesis) or unaesthetically wide spaces should be orthodontically optimized to allow prosthodontic rehabilitation.[28]

Very scarce case reports in the literature mention the existence of protostylid in human teeth.[21] The formation of the cuspal form was not observed among Negroes, Filipinos, and Hawaiians, whereas a greater incidence in both primary and permanent molars of southwestern Indians has been described.[17,33] Studies have shown that some Mongoloids, including southern Chinese, have a higher prevalence of protostylid traits than Caucasians.[5] These morphological variants can represent racial and regional variations.[21,34] The presence of protostylid in maxillary second molars and mandibular premolars is particularly unusual ranging from 0.4% to 2.8% in various populations.[35] The protostylid derives from the outfolding of inner enamel epithelium and focal hyperplasia of peripheral cells of mesenchymal dental papilla, during morphodifferentiation stage of tooth development.[36] Clinically, the occurrence of protostylid on teeth can range from a groove of varying depth and size to well-developed tubercle.[36] The groove type protostylid or surface irregularity is more familiar than the other form. Bilateral symmetry of this trait is common, and it is usually seen in the form of a buccal pit, while expression as a cusp is rare.[5]

Generally, protostylids have no functional significance because they are located on the buccal side of the related tooth and do not contact the opposing tooth during occlusion.[21] However, these morphological variations can lead to various clinical implications such as a tooth with pit and fissure caries, sensitivity, and devitalization due to fracture of the certain portion of cusp leading to pulpal exposure.[21,37] During orthodontic treatment, protostylid may interfere with the cementation of brackets and in the alignment of arch wires and thus often necessitates its removal by ameloplasty.[28] In view of our case, the protostylids does not causing any occlusal interference and happen to have occurred on the upper second permanent molar. Therefore, the anomalies will not be interfering with the orthodontic band placement and alignment of arch wire. Apart from that, these dental anomalies can be precious traces to cultural nature and enhance distinctive recognition of an individual's dentition. Consequently, the reporting and preservation of these structures can be crucial from the viewpoint of forensic odontology.[37]

Conclusion

The occurrence of shovel-shaped incisors, dens invaginatus, multiple protostylid, and malocclusion are such that dental practitioners should be prepared for appro-

priate interdisciplinary treatment planning to obtain excellent results. Thus, a comprehensive multidisciplinary treatment approach involving a general dentist, restorative dentist, radiologist, and orthodontist is requisite while managing dental anomalies and malocclusion. Also, maintaining the vitality of pulp or preserving the tooth by the least invasive method is needed. Preventive measures and close monitoring are crucial as soon as the dental anomalies are diagnosed.

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