

## Winged Maxillary Premolar in a Patient of Neurofibroma: A Rare Case Report

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### Abstract:

Dens evaginatus is a rare developmental anomaly characterized by the presence of extra cusp like projection from the occlusal surface of an affected tooth commonly seen on premolars. This study reports an unusual case of a 26-year-old female with projections from the mesial and distal surface of maxillary second premolar, a finding that has previously not been reported.

**Key Words:** Dens evaginatus, fusion, neurofibroma, projections

### Introduction

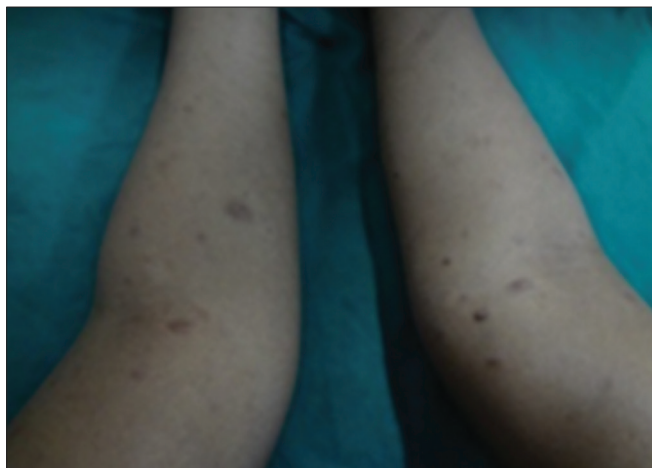
Accessory cusp projections are uncommon in primary or permanent teeth. Talon cusp is the preferred term when describing accessory cusp like projections in anterior teeth and Dens Evaginatus (DE) for posterior teeth.<sup>1</sup> DE is a rare developmental aberration of a tooth resulting in the formation of an accessory cusp whose morphology has been variously described as an abnormal tubercle, elevation, protuberance, excrescence, extrusion, or bulge. This uncommon anomaly projects above the adjacent tooth surface, exhibiting enamel covering a dentinal core that usually contains pulp tissue that on occasion may have a slender pulp horn.

It may be present unilaterally or bilaterally in males or females. In the majority of cases, it is present on the occlusal surface between cusps and a very few cases have been reported documenting DE on proximal surfaces of premolars.

Neurofibroma Type I (NF-I) is present in seven different forms out of which two distinct forms are generally accepted namely a peripheral form known as NF-I and a central form known as NF-II.<sup>2</sup> NF-I representing the classic form, is a genetic disorder inherited as an autosomal dominant trait. The major features of NF-I include NFs, café-o-lait macules and lisch nodules.<sup>3</sup> Oral lesions are usually discrete, non-ulcerated nodules varying from normal mucosal color to red even yellow, occurring in 2-7% of cases. They have been located in the soft tissues such as the cheek, palate, tongue, the floor of mouth, and lips. The tongue is the most common affected site.

### Case Report

A 26-year-old female reported to the Department of Conservative Dentistry & Endodontics, with a chief complaint of pain and pus discharge with the upper left back region since 1-month medical history revealed NF-I. Family history revealed that her mother and grandmother were known patients of NF. General examination revealed Café-o-lait Spots on flexor surface of the left and right forearm (Figure 1). Intraoral examination disclosed pigmentation on the tongue, missing left maxillary first premolar, with mesial migration of the second premolar. An intraoral sinus was seen in relation to the left second premolar. Exophytic projections of the crown having the color as that of enamel were seen arising from the mesial and distal aspect of the tooth just below the marginal ridges were seen on the second premolar (Figure 2). The distal projection was larger than the mesial. The junction between the mesial projection and the crown was found carious. Response to percussion was normal, and pulp tests yielded no response. The clinical differential diagnosis considered were fusion of supernumerary or odontome to the second premolar and gemination or DE of the second premolar. Radiographic examination confirmed missing the first premolar. Periapical radiograph in relation to the second premolar revealed Class I composite restoration on the distal pit and proximal caries at the junction of mesial projection and mesial surface. The projections had almost the same density as that of enamel and dentin. The single large root canal was found which ruled out the possibility of fusion or gemination of the second premolar. Irregular radiolucency >1 cm was seen periodically (Figure 3). Treatment plan advocated was root canal treatment followed by fixed prosthesis. After Rubber dam application, (Figure 4) access cavity preparation was done using the high speed air-rotor hand piece with diamond points. A single large canal was found, working length was determined by placing 15 no. K file (Figure 5). 1 mm short of this length was decided as working



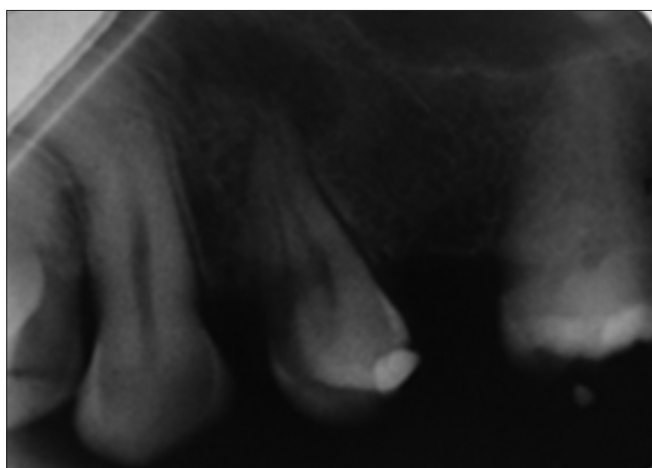
**Figure 1:** Café-o-lait spots on flexor surface of forearms.



**Figure 4:** Rubber dam application.

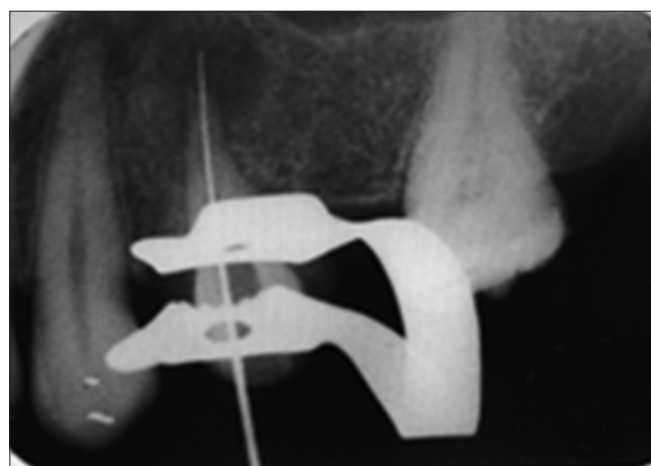


**Figure 2:** Mesial and distal exophytic projections on left maxillary second premolar.



**Figure 3:** Pre-operative intraoral radiograph.

length. This was followed by cleaning and shaping the canal using Protaper (Dentsply Maillefer). During the preparation Glyde file prep (Dentsply Maillefer) was used as a lubricant with 5.25% sodium hypochlorite (Primedent) as a disinfectant followed by intermittent copious irrigation with normal saline. The canal was closed with metapex (Meta Biomed)



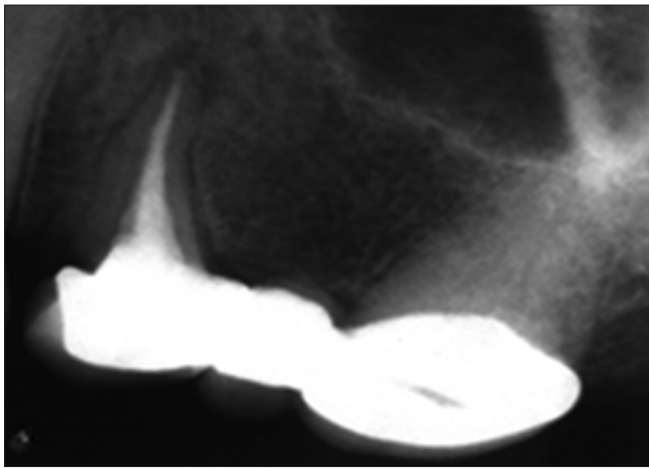
**Figure 5:** Working length determination.

for 2 weeks. Master IOPA was taken with Protaper GP point F<sub>2</sub>. The canal was dried with paper points (Dentsply India) and was obturated with Protaper GP point F<sub>2</sub> and AH-Plus sealer (Dentsply). The orifice was sealed with glass ionomer cement (GC Fuji II) followed by access cavity filling with Filtek Z350 (3M ESPE) composite.

The mucoperiosteal flap was raised, and the projections were sectioned to the level of alveolar crest and sent for histopathological examination. A ground section of the tooth revealed enamel rods and dentinal tubules, thereby suggesting that the projection was in continuation with the proximal surface of the tooth. The patient was referred for fixed prosthesis. After 6 months follow-up period, the tooth was asymptomatic with favorable periapical changes (Figure 6). The patient is still under regular clinical and radiographical reevaluation.

#### Discussion

Fusion is a rare developmental dental anomaly caused by the union of two tooth germs to form a single tooth. It has a complex morphology that can give rise to reduced esthetics, misalignment, dental caries, and periodontal problems. Fused



**Figure 6:** Post-operative radiograph follow-up intraoral periapical.

teeth are usually united by enamel and dentin, whereas the pulp chambers and pulp canals are either unified or separated. It is more frequent among Japanese population and American Indians. Fusion can occur between two teeth or a normal tooth with supernumerary teeth and odontoma.<sup>4</sup> A communication of the regular and fused root canal system exist in all fused posterior teeth. In this case, a single large root canal was found which made us rule out fusion.

DE is a rare dental anomaly with a prevalence of 1-4% among Asians. It is more commonly seen on the occlusal surface between the cusps of mandibular premolars and rare on molars. It may be present unilaterally or bilaterally in primary or permanent teeth, with more predilections for females.<sup>5</sup>

It has been suggested that it may have a multifactorial etiology including genetic, environmental factors, and hyperactivity of the dental lamina. The pathogenesis of the lesion is thought to be proliferation and evagination of internal enamel epithelium and the adjacent odontogenic mesenchyme into the stellate reticulum of the enamel organ.<sup>6</sup> The cells in the center of enamel organ are densely packed and form the enamel knot. The current view is that the enamel knot represents an organizational center, which orchestrates cuspal morphogenesis, patterning the cusps and hence the shape of the tooth crown.<sup>7-9</sup>

Only one case has been reported a detailing projection of tooth from the mesial surface of premolar,<sup>8</sup> and this is the first case of projections on both mesial and the distal surface of maxillary premolar in dental literature. The etiology of the present case might be similar to the other DE presentations, which include both genetic and environmental factors, and this case could have been just a rare oddity. Presently no title exists for such an appearance, we grouped it under DE and suggest terms like “dental horn” or “winged premolar.”

### Conclusion

The aim of this report is to emphasize and increase awareness of this unique finding among the dental fraternity. We report a never before described clinical finding of a proximal wing-like structure of the maxillary left first premolar as a result of an anomalous development of the enamel, and probably the underlying dentine.

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