

# Nasopalatine Duct Cyst Associated with Dens in Dente and Semi-Talon Cusp: Case Report

## Dens in Dente ve Semi-Talon Tüberkülü ile İlişkili Nasopalatin Kanal Kisti

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**ABSTRACT** The purpose of this report is to present an unusual clinical case presenting with simultaneous combination of dental anomalies such as nasopalatine cyst, dens in dente and semi-talon cusp. Radiographic examination of a 37-year-old male patient revealed a radiolucent area of approximately 3 × 5 cm in size underneath the 12<sup>th</sup> tooth. The vitality of the tooth was negative. The cyst was thought to be a radicular cyst and was enucleated surgically. Pathologic evaluation of the enucleated fragment showed that the cyst was lined with non-keratinized stratified columnar epithelium, squamous epithelia and chronic inflammatory cells; the findings were concordant with a nasopalatine duct cyst. An unusual feature of this cyst is that it was formed under the non-vital 12<sup>th</sup> tooth, instead of in the midline of the anterior part of the maxilla and was associated with dens in dente and talon cusp. Clinical and radiographic examination is suggested when a tooth is associated with other dental anomalies. The present case illustrates that dental invagination can be associated with other dental anomalies. He was resolved by endodontic treatment and enucleation of the cyst after evaluation of the maxillary and mandibular dental discrepancy and the clinical status of the patient.

**Key Words:** Dens in dente; cystic duct

**ÖZET** Bu çalışmanın amacı, nasopalatin kist, dens in dente ve semi-talon tüberkülünün kombine olarak birarada görüldükleri, sık rastlanılmayan bir klinik olgunun takdim edilmesidir. 37 yaşındaki bir erkek hastada, radyografik inceleme sonucunda 12 no'lu dişin altında yaklaşık 3 x 5 cm boyutunda radyolüsent bölge saptanmıştır. Dişin vitalometrik incelemeye verdiği cevap negatiftir. Bu oluşumun bir radiküler kist olduğu düşünülerek, kist cerrahi yolla enükle edilmiştir. Enükle edilen fragmanın patolojik değerlendirmesinde, non-keratinize epitel ile çevrili ve kronik inflamatuvar hücreler içeren bir nasopalatin kanal kisti olduğu belirlenmiştir. Bu kistin non-vital bir dişle bağlantılı olarak görülmesi, anterior orta hat içinde olmaması ve dişte semi-talon tüberkülü ile dens in dente'nin aynı anda görülmesi ender karşılaşılan bir durumdur. Bir diş, diğer dental anomalilerle ilişkili bulunduğu klinik ve radyolojik inceleme önerilir. Mevcut olgu dental invajinasyonun diğer dental anomaliler ile birlikte görülebildiğini ortaya koymaktadır. Bu vaka, hastadaki bu olağan dışı durumun değerlendirilmesinden sonra endodontik terapi ve kistin enükleasyonu ile tedavi edilmiştir.

**Anahtar Kelimeler:** Dens in dente; kistik kanal

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**R**adiolucid areas are occasionally observed between the roots of the maxillary lateral incisor and canine teeth in patients with diseases such as radicular granuloma, radicular cyst, nasopalatine cyst and dens invaginatus.<sup>1</sup>

Dental invagination or dens in dente is a rare malformation with a widely varied morphology.<sup>2-4</sup> The synonyms for this condition are as follows: dens in dente, dens invaginatus, gestant odontome, invaginated odontoma, tooth inclusion, dilated odontoma and dentoid in dente. The different terms reflect the varied theories on its etiology. Numerous mechanisms have been proposed as being responsible for this phenomenon, including local delay in enamel formation, infolding of the enamel organ within the dental pulp, and local external effects on the tooth germ.<sup>5</sup> This condition is a developmental abnormality of a normal tooth and occurs before crown calcification and coronal invaginations originate from an anomalous infolding of the enamel organ into the dental papilla. This dental abnormality is most frequently seen in the permanent maxillary lateral incisor and commonly occurs bilaterally; its prevalence is reported to be 1.7 to 10%.<sup>6,7</sup> Men are more affected by this condition than women in a ratio of 3 to 1.<sup>8,9</sup> A recent study on patients undergoing orthodontic treatment showed a prevalence of 26.1% and a higher incidence among women.<sup>10</sup> Different classifications have been proposed for this anomaly. Hallet proposed four types of invagination: type 1, a fistula in the palatal enamel at the cervical level that runs vertically with no expansion or dilatation; type 2, the invagination extends toward the pulp of the canal and a small perforation is formed in the cingulum; type 3, the invagination extends deep within the pulp and is dilated; and type 4, the invagination fills the coronal pulp and may extend through the amelocemental junction.<sup>11</sup>

Oehlers described three categories of dental invagination according to the depth of penetration and the presence of communication with the periapical tissue: category 1, the invagination ends in a blind sac at the crown; category 2, the invagination extends apically, following the external amelocemental junction; and category 3, the invagination also extends through the amelocemental junction and forms a second apical foramen in the periapical tissues.<sup>12,13</sup>

Radiographically, the affected tooth shows an infolding of the enamel and dentine that can extend to within the pulp cavity and the root and sometimes into the root apex. A thin opaque line superimposed on the pulp is seen on radiographic examination of an affected tooth. Most cases of dens in dente are discovered radiographically if acute apical periodontitis does not progress. The invaginated teeth in certain cases can be successfully treated only with an endodontic approach.

Talon cusp has been reported as a rare and uncommon condition, and presents as a cusp-like structure that varies considerably in size, ranging from an enlarged cingulum to a well-delineated anomalous cusp extending to at least half of the crown height and usually occurring on the occlusal or lingual surfaces. It may be found on both primary and permanent incisors and this hyperplasia consists of enamel that may become susceptible to caries. Other names for Talon cusp include dens evaginatus, interstitial cusp, evaginated odontoma, occlusal enamel pearl, occlusal anomalous tubercle and supernumerary cusp. Type 2 (Semi-Talon) Talon cusp is an additional cusp with a length of 1 mm or more, but extending less than half the distance from the cemento-enamel junction to the incisal edge and is seen as a radiopaque image which is superimposed on that of the crown of the incisor. The treatment of talon cusp in cases where the cusp effects the occlusion or where caries occur is the slow removal of the cusp.<sup>3-4</sup>

The synonyms for nasopalatine duct cysts (NPDC) are incisive canal cyst, median palatine cyst or median anterior maxillary cyst. NPDC are classified as epithelial non-odontogenic cysts according to the 1992 WHO histological typing of odontogenic tumors.<sup>12</sup> NPDC develops from the remnants of the nasopalatine duct and accounts for about 10% of jaw cysts which form in the midline of the anterior part of the maxilla.<sup>14</sup> The incidence of these cysts is greatest among patients in their fourth through sixth decades. These cysts are asymptomatic. This type of cyst is located in the nasopalatine foramen and on radiographic examination the periphery is usually found to be well-

defined, oval in shape, and totally radiolucent whilst the area surrounding the cyst is radiopaque. This type of cyst is occasionally responsible for root resorption. On diagnosis the cyst can be differentiated as a radicular cyst or granuloma. A vitality test can facilitate the diagnosis of the cyst. The cyst can be treated by enucleation or if it is large, marsupialization of cyst can be considered. Recurrence after treatment is rare.

## CASE REPORT

A 37-year-old man whose major symptom was headache, presented at our dental faculty in Samsun, Turkey. His medical history did not reveal any systemic disease. Intraoral examination revealed moderate to poor oral hygiene and the maxillary incisors showed cingulum hypertrophies and a foramen coecum was observed on tooth 12. Large palatal and small nasolabial swellings were noticed at the apex of this tooth although mobility was normal (Figure 1). The palatal swelling was fluctuant, painless, non-bleeding and approximately with  $3 \times 5$  cm in diameter with a normal mucosal color. The patient reported a history of palatal swelling but had not sought dental treatment. He had used an antibiotic on an annual basis with the consequent reduction in the size of the swelling after each administration. Panoramic, periapical and occlusal radiographs were taken in order to determine the dimension of the



**FIGURE 1.** Incisal view of the subject's maxilla showing the presence of Semi-Talon cusp and cingulum hypertrophies.



**FIGURE 2.** Panoramic radiograph showing the cyst.



**FIGURE 3.** Occlusal radiography showing cyst and teeth structures.

lesion (Figure 2, 3). The radiographic appearance of tooth 12 was different from that of the incisors affected by cingulum hyperplasia. It was therefore suggested that tooth 12 was affected by a semi-talon cusp. The radiographs showed that the radiolucent area was between the right second



**FIGURE 4.** Endodontically treated maxillary lateral incisor.

premolar and the left lateral tooth, approximately 6 × 4 cm in diameter. The vitalities of the teeth were checked and only the right lateral incisor which was diagnosed as dens in dente and semi-talon cusp was negative. Our treatment plan consisted of endodontic treatment of tooth 12



**FIGURE 5.** The surgical operation of the cyst.

followed by enucleation of the cyst around this tooth followed by histopathological evaluation of fragment.

After endodontic treatment of the tooth (Figure 4), the cyst was totally enucleated. N. infraorbitalis, n. incisivus and n. palatinus were blocked with articain hydrochloride containing 0.006 mg/ml epinephrine. The incisions were made at the palatal sulcular area and between right I. molar and I. premolar teeth (Figure 5). After elevating the palatal mucoperiosteal flap, cyst perforation on the palatal bone was observed and the cyst cavity door was spread out. The totally enucleated cyst epithelium and cyst cavity were curetted. Apical resection was applied to the right lateral tooth. The cavity door was closed using a bovine collagen membrane (30 × 40 mm; Mem-lock). The flap was sutured with 3/0 silk. Amoxicillin 1 g (2 × 1), flurbiprofen 100 mg (2×1) and klorhexi dinediglukonate gargle (3×1) was administered postoperatively.

The enucleated fragment (Figure 6) was evaluated by a pathologist. The epithelial lining of this cyst was formed from non-keratinized stratified columnar epithelia, squamous epithelia and chronic inflammatory cells. The pathology result was concordant with nasopalatine duct cyst (NPDC). Informed consent was taken from the patient before the procedure.



**FIGURE 6.** The enucleated cyst fragment.

## DISCUSSION

The present case illustrates a problematic diagnostic challenge. The patient in the current case showed various dental anomalies in his teeth. The simultaneous presence of multiple dental anomalies has been previously reported, especially in patients with chromosomal alterations who may present with multisystemic alterations.<sup>15</sup>

Several authors have described the difficulty in diagnosing radiolucent areas related to invaginated teeth. A combination of dental anomalies also has been reported in patients without generalized abnormalities or disease.<sup>16</sup>

Several hypotheses have been postulated regarding the development of an invaginated tooth. The earliest hypotheses attributed the malformation to the incomplete fusion of two tooth germs or to the attempted division of one, an abnormal proliferation of enamel invading the interior of the dental pulp, or a local delay in the growth of the enamel epithelium during dental development. However, most authors considered dental invagination to be a deep infolding of the apical foramen during dental development, resulting in some cases, in a second apical foramen. On the other hand, the invagination can also start from the incisal edge of the tooth. If the hypothesis of genetic predetermination of tooth size is correct, invagination may also be predetermined, and if the tooth germ matures,

then the tooth will be affected by the same dental anomaly.<sup>17</sup>

This case is of particular clinical interest as it is rare for a NPDC to be seen together with dens invaginatus and semi-talon cusp. In the largest study of 334 NPDCs, the overall mean age of the subjects was 42.5 years.<sup>18</sup> The age of the patient in the present study was 37 years. A total of 40% of the cases was completely asymptomatic and the cysts were found only during routine clinical examination.<sup>19</sup>

Contrary to these findings, in this case the patient was suffering from continuous headache. The devitalization of the teeth associated with NPDC as reported in this study has also been documented previously.<sup>20</sup> We believe that the pressure of cysts and dens in dente lead to the devitalization of the tooth. Consequently our case has demonstrated the atypical clinical and radiographic features of NPDC.

The present case illustrates that dental invagination can be associated with other dental anomalies. He was resolved by endodontic treatment and enucleation of the cyst after evaluation of the maxillary and mandibular dental discrepancy and the clinical status of the patient.

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