

Non-syndromic multiple supernumerary teeth associated with Dens Invaginatus: a case report

Dientes supernumerarios múltiples no sindrómicos asociados a Dens Invaginatus: reporte de un caso

Mario Alejandro Ortiz¹

- ¹ Departamento de Morfología, Universidad del Valle. Cali, Colombia.
- * Corresponding author: mario.ortiz@correounivalle.edu.co

ABSTRACT

Background: Supernumerary teeth are an infrequent developmental alteration causing alteration in normal arch form and occlusion. Multiple supernumerary teeth can be associated with some syndromes. However, it can be present in patients without any systemic pathology. Dens invaginatus is a dental developmental variation resulting from an alteration in the normal growth pattern of the dental papilla of a tooth. Cone Beam Computered Tomography (CBCT) could be used to diagnosis supernumerary tooth and dens invaginatus. **Case report:** A case of a 10-year-old girl with multiple supernumerary teeth without syndromic association is presented. A rare association with dens invaginatus was present. Supernumerary teeth were located in maxilla and mandible and cause severe tooth displacement and lip incompetence. **Discussion:** Hyperodontia is a number dental alteration of unknown etiology with a prevalence that varies between 0.1 and 3.8%. Dens in dente is a dental shape alteration of unknown etiology with variable prevalence depending on the population. This alteration puts the integrity and health of the affected tooth at risk. Early diagnosis is important in the long-term prognosis. Radiological tools are the appropriate option for diagnosis and are shown in this article.

Keywords: Dens Invaginatus; Supernumerary teeth, Maloclussion.

RESUMEN

Introducción: Los dientes supernumerarios son una alteración del desarrollo poco frecuente que causa alteración en la forma normal del arco y en la oclusión. Varios dientes supernumerarios pueden asociarse con algunos síndromes. Sin embargo, puede estar presente en pacientes sin patología sistémica. Dens invaginatus es una variación del desarrollo dental que resulta de una alteración en el patrón de crecimiento normal de la papila dental de un diente. La tomografía computarizada de haz cónico (CBCT) podría usarse para diagnosticar dientes supernumerarios y dens invaginatus. Caso clínico: Se presenta el caso de una niña de 10 años con múltiples dientes supernumerarios sin asociación sindrómica. Estuvo presente una rara asociación con dens invaginatus. Los dientes supernumerarios se ubicaron en el maxilar y la mandíbula y causaron un desplazamiento dental severo e incompetencia labial. Discusión: La hiperodoncia es una alteración dental de etiología desconocida con una prevalencia que varía entre el 0,1 y el 3,8%. Dens in dente es una alteración de la forma dentaria de etiología desconocida con prevalencia variable según la población. Esta alteración pone en riesgo la integridad y la salud del diente afectado. El diagnóstico temprano es importante en el pronóstico a largo plazo. Las herramientas radiológicas son la opción adecuada para el diagnóstico y se muestran en este artículo.

Palabras clave: Dens Invaginatus; Dientes supernumerarios; Maloclusión.

Article History

Received: 09 01 20 Aceptado: 10 09 20 Publicado: 12 11 20

DOI 10.17081/innosa.102 ©Copyright 2020.



I. BACKGROUND

A supernumerary tooth or hyperodontia is a clinical condition in which are additional teeth to the normal series and can be found in almost any region of the dental arch(1, 2). The prevalence of supernumerary teeth varies between 0.1 and 3.8% and is more common in the permanent dentition(3). The etiology of supernumerary teeth is not completely understood. Both genetic and environmental factors have been considered(4). Supernumerary tooth could be found in both deciduous and permanent dentition. They can be classified according to location and morphology. Topographically (location) they can be mesiodens, paramolar, distomolar and parapremolar (5). Four different morphological types of supernumerary teeth have been described: conical, tuberculate, supplemental and odontoma (2).

Dens invaginatus, also known as dens in dente or dilated composite odontoma, is a rare malformation of teeth, showing a broad spectrum of morphological variations. The affected teeth radiographically show an in folding of enamel and dentine which may extend deep into the pulp cavity and into the root and sometimes even reach the root apex (6). The etiology of dens invaginatus malformation is controversial and remains unclear and prevalence varies from 0.3 to 10% depending of the population. Although a clinical examination reveals a deep pit or fissures on lingual surfaces of anterior teeth, the radiographic examination is the sine quo non for diagnosis of dens invaginatus (7).

This article reports one case of concomitant occurrence of non-syndromic supernumerary teeth and dens invaginatus.

II. CASE REPORT

A 10-year-old girl attended the author's clinic for a regular checkup. Her mother reported with complaint of abnormally erupted tooth in maxillary anterior region, severe tooth malposition and lip incompetence. Her medical history was unremarkable and there was no family history of supernumerary teeth or dens invaginatus. However, she had a history of two supernumerary deciduous upper central incisors extracted when she was 7 years old. Psychological affection was present due to facial appearance associated to lip incompetence and severe crowding. There was bullying at school.

At the time of initial examination, the patient had full permanent dentition (except third molars). Examination revealed Class II malocclusion, severe upper crowding, large overjet, lip incompetence, palatal eruption of one supernumerary teeth and buccal displacement of upper central incisors (**Figure 1 and 2**).

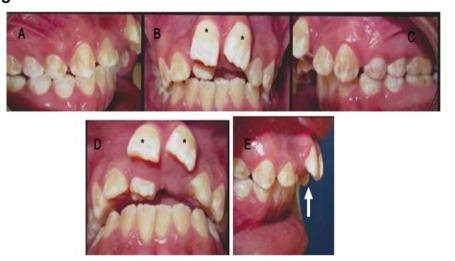
Figure 1. Clinical Evaluation before orthodontic treatment.





Upper occlusal view(A) and Lower occlusal view (B). Note buccal displacement of upper central incisors and supernumerary tooth eruption on the right side). **Source:** Author

Figure 2. Clinical Intraoral evaluation before orthodontic treatment.



Right lateral intraoral view (A), Frontal intraoral view (B), Left lateral intraoral view (C), Overbite view (D) and Overjet view (E). Observe severe upper central incisors (*) buccal displacement causing increased overjet (white arrow) and lip incompetence. A generalized amelogenesis imperfecta is present. Source: Author

Radiological evaluation was performed using panoramic radiograph and cone-beam computed tomography (CBCT). CBCT in the upper arch showed two supplemental mesiodens in palatal position to upper central incisors and severe buccal displacement of upper central incisor out of the alveolar process. Left mesiodens had a Oehlers's type 2 dens invaginatus. There was an additional supernumerary parapremolar tooth between upper left canine and first bicuspid, another one between lower first molar and second bicuspid and a last one between first and second lower bicuspid (**Figure 3 - 4**). There was a total of 5 supplemental supernumerary teeth.

Treatment involved the extraction of upper and lower supernumerary teeth in order to relieve the crowding. Orthodontic phase 1 treatment was performed in the upper arch in order obtain dental alignment and eliminate lip incompetence. A phase 2 comprehensive orthodontic treatment was planned in order to manage class II malocclusion.

IV. DISCUSSION

Multiple supernumerary teeth are more common when a syndrome is involved(8) such as: cleidocranial dysostosis, Gardener syndrome, Enlers-Danios syndrome, Fabry Anderson's syndrome, chondroectodermal dysplasia, incontinentia pigmenti and tricho rhino-phalangeal syndrome. Ysuof suggested that it may be rare to find

multiple supernumerary teeth without an associated syndrome; however, non-syndromic multiple supernumerary teeth can occur($\underline{9}$). Indeed, multiple authors have reported several cases of non-syndromic supernumerary teeth. Supernumerary teeth can be classified based on morphology and location($\underline{10}$ – $\underline{12}$). Those teeth can also present development alterations as Amelogenesis imperfecta, crown morphology, root morphology and dens invaginatus($\underline{1}$).

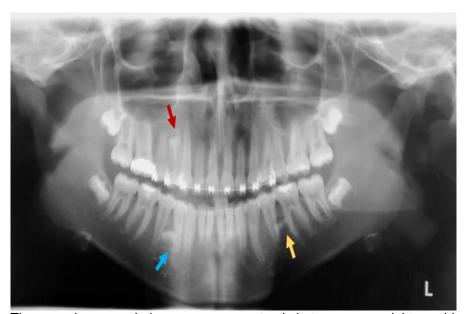


Figure 3. Panoramic x ray taken after two mesiodens extraction

There are three remainder supernumerary teeth; between upper right cuspid and first bicuspid (red arrow), between left lower second bicuspid and first molar (yellow arrow) and between first and second right lower bicuspids (blue arrow). **Source:** Author

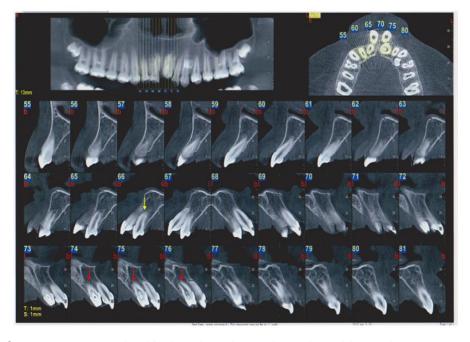


Figure 4. CBCT imaging.

Supernumerary tooth with dens invaginatus is evaluated in sections 74, 75 and 76 (red arrow). Right supernumerary tooth generating upper central incisor displacement (yellow arrow). **Source**: Author

The aetiological basis of supernumerary teeth is poorly understood; however, there is clearly an important genetic component. Supernumeraries run in families, demonstrate racial variation and display sexual dimorphism (13). However, some of the most compelling evidence for a genetic basis is the fact that supernumerary teeth can also be a prominent feature of many developmental disorders(14). Familiar incidence and some genetic factors have been reported in the literature, including *sonic hedgehog* (shh), *fibroblast growth factor* (fgf), Tumor Necrosis factor pathway, Bone Morphogenic Protein signalling and WNT signalling(14,15).

Dens invaginatus is a developmental anomaly resulting in a deepening or invagination of the enamel organ into the dental papilla prior to calcification of the dental tissues(6). Oehlers classified dens invaginatus in three forms depending on the extent of the invagination. Type dens invagination presents an enamel lined form which invades the root but remains confined as blind sac. It may or may not communicate with dental pulp (16). The invagination presented in this case penetrated to the level of the root and did not communicate with the pulp. The permanent maxillary lateral incisor appears to be the most frequently affected tooth with posterior teeth less likely to be affected (17). Non-syndromic supernumerary teeth associated with dens invaginatus are rare(7,18).

The presence of an invagination is considered to increase the risk of caries, pulpal pathosis and periodontal inflammation (6). Therefore, management always depends on the type of supernumerary teeth and its position, relation to other tooth and its effects on adjacent teeth (1). Treatment plan depends of a correct diagnosis. Now days, CBCT is a very useful tool to

investigate multiple supernumerary teeth and its use has been increasing in order to decrease the exposure dose (19,20). Current case was evaluated using initial orthodontic recordings, including panoramic x-ray. Furthermore, CBCT was used to determine supernumerary tooth position, allowing dens invaginatus diagnosis. However, CBCT is not a standard diagnosis method in Orthodontics and must be used following the *American Academy of Oral and Maxillofacial Radiology Clinical recommendations regarding use of cone beam computed tomography in Orthodontics* (21). In conclusion, indiscriminate, routine use of CBCT for all orthodontic patients is considered an unacceptable practice. In the presented case, CBCT was justified because it was useful to evaluate supernumerary tooth position and allow clinical decision of tooth extraction and comprehensive orthodontics. CBCT is a usefull tool for early diagnosis of supernumerary tooth and dens invaginatus.

Authors Contributions: "Conceptualization, M.A.O.; methodology M.A.O.; investigation, M.A.O.; resources, M.A.O.; data healing, M.A.O.; writing: preparing the original draft, M.A.O.; writing: writing: review and editing, M.A.O. All authors have read and accepted the published version of the manuscript.

Conflicts of interest: The author declare no conflict of interest.

REFERENCES

- 1. Shah A, Gill DS, Tredwin C, Naini FB. Diagnosis and management of supernumerary teeth. Dent Update [Internet]. 2008;35(8):519–20. DOI 10.12968/denu.2008.35.8.510
- 2. Garvey MT, Barry HJ, Blake M. Supernumerary teeth--an overview of classification, diagnosis and management. J Can Dent Assoc [Internet]. 1999;65(11):612–6. https://pubmed.ncbi.nlm.nih.gov/10658390/
- 3. Rajab LD, Hamdan MAM. Supernumerary teeth: Review of the literature and a survey of 152 cases. Int J Paediatr Dent [Internet]. 2002;12(4):244–54. DOI: 10.1046/j.1365-263x.2002.00366.x
- 4. Hall A, Onn A. The development of supernumerary teeth in the mandible in cases with a history of supernumeraries in the pre-maxillary region. J Orthod [Internet]. 2006;33(4):250–5. DOI 10.1179/146531205225021735
- 5. Parolia A, Kundabala M, Dahal M, Mohan M, Thomas MS. Management of supernumerary teeth. J Conserv Dent [Internet]. 2011;14(3):221–4. https://www.jcd.org.in/article.asp?issn=0972-0707;year=2011;volume=14;issue=3;spage=221;epage=224;aulast=Parolia
- 6. Hülsmann M. Dens invaginatus: Aetiology, classification, prevalence, diagnosis, and treatment considerations. Int Endod J [Internet]. 1997;30(2):79–90. <u>DOI: 10.1046/j.1365-2591.1997.00065.x</u>
- 7. Mupparapu M, Singer SR. A rare presentation of dens invaginatus in a mandibular lateral incisoroccurring concurrently with bilateral maxillary dens invaginatus: Case report and review of literature. Aust Dent J [Internet]. 2004;49(2):90–3. DOI: 10.1111/j.1834-7819.2004.tb00056.x
- 8. Scheiner MA, Sampson WJ. Supernumerary teeth: A review of the literature and four case reports. Aust Dent J [Internet]. 1997;42(3):160–5. DOI 10.1111/j.1834-7819.1997.tb00114.x
- 9. Yusof WZ. Non-syndrome multiple supernumerary teeth: literature review. J Can Dent Assoc [Internet]. 1990;56(2):147–9. DOI 10.1016/0278-2391(90)90508-y
- 10. Gündüz K, Muğlali M. Non-syndrome multiple supernumerary teeth: A case report. J Contemp Dent Pract. 2007;8(4):81–7. https://pubmed.ncbi.nlm.nih.gov/17486191/

- 11. Ansari AA, Malhotra S, Pandey RK, Bharti K. Non-syndromic multiple supernumerary teeth: Report of a case with 13 supplemental teeth. BMJ Case Rep [Internet]. 2013. DOI 10.1136/bcr-2012-008316
- 12. Yagüe-García J, Berini-Aytés L, Gay-Escoda C. Multiple supernumerary teeth not associated with complex syndromes: A retrospective study. Med Oral Patol Oral Cir Bucal. 2009. https://pubmed.ncbi.nlm.nih.gov/19300360/
- 13. Brook AH. Dental anomalies of number, form and size: their prevalence in British schoolchildren. J Int Assoc Dent Child [Internet]. 1974;5(2):37–53. Available from: https://pubmed.ncbi.nlm.nih.gov/4535299/
- 14. Cobourne MT, Fleming PS, Xavier GM, Dibiase AT. Revisiting the supernumerary: The epidemiological and molecular basis of extra teeth. Br Dent J [Internet]. 2010;208(1):25–30. Available from: DOI 10.1038/si.bdi.2009.1177
- 15. Khambete N, Kumar R. Genetics and presence of non-syndromic supernumerary teeth: A mystery case report and review of literature. Contemp Clin Dent [Internet]. 2012;3(4):499–502. DOI: 10.4103/0976-237X.107455
- 16. Oehlers FAC. Dens invaginatus (dilated composite odontome). I. Variations of the invagination process and associated anterior crown forms. Oral Surgery, Oral Med Oral Pathol [Internet]. 1957;10(11):1204–18. Available from: DOI 10.1016/0030-4220(57)90077-4
- 17. Alani A, Bishop K. Dens invaginatus. Part 1: Classification, prevalence and aetiology. Int Endod J [Internet]. 2008;41(12):1123–36. DOI 10.1111/j.1365-2591.2008.01468.x
- 18. Jiménez-Rubio A, Segura JJ, Jiménez-Planas A, Llamas R. Multiple dens invaginatus affecting maxillary lateral incisors and a supernumerary tooth. Endod Dent Traumatol [Internet]. 1997;13(4):196–8. DOI: 10.1111/j.1600-9657.1997.tb00039.x
- 19. Akhila R DDS. Management Of Multiple Impacted Supernumerary Teeth In A Non-Syndromic Patient Using Cone Beam CT. Dentistry [Internet]. 2014;4(2). Available from: 10.4172/2161-1122.1000190
- 20. Wang WM, Wang X, Wang TM. Evaluation of nonsyndromic multiple supernumerary teeth using three-dimensional computerized tomography: A case report and literature review. J Contemp Dent Pract [Internet]. 2010;11(1):E081-7. https://pubmed.ncbi.nlm.nih.gov/20098970/
- 21. Radiology AA of O and M. Clinical recommendations regarding use of cone beam computed tomography in orthodontic treatment. Position statement by the American Academy of Oral and Maxillofacial Radiology. Oral Surg Oral Med Oral Pathol Oral Radiol [Internet]. 2013;116(2):238–57. Available from: DOI 10.1016/j.0000.2013.06.002