CASE REPORT

Management of non-syndromic dens evaginatus affecting permanent maxillary central incisors: a systematic review

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SUMMARY

To assess management of non-syndromic dens evaginatus affecting permanent maxillary central incisor, we performed a systematic review and also present a case report. We searched PubMed via MEDLINE and the reference lists of included reports. Eligible studies were any type of clinical studies describing the management of non-syndromic dens evaginatus affecting the crown of a permanent maxillary central incisor. We included 31 studies corresponding to 34 relevant case reports. Therapeutic options were complete reduction of the talon cusp in a single appointment (56%), periodic and gradual reduction of the cusp (26%), abstention (13%) or extraction (5%). We report an 8-year-old girl with unusual two-talon cusp, labial and lingual, on a right maxillary double central incisor. A multidisciplinary approach is key to management of permanent maxillary central incisors affected by coronary anomalies.

CASE PRESENTATION

An 8-year-old girl presented an abnormally shaped tooth. A labial view showed the presence of malformation on the right maxillary central incisor 11 (figure 1). The medical and familial history was unremarkable, and the patient exhibited no syndrome or history of dental trauma. A second dental anomaly was a prominent cusp-like structure on the palatal surface of the same tooth. The tooth appeared to be x-shaped when viewed occlusally (figure 2). The right lateral incisor (12) was rotated because of the tooth size-arch length deficiency in the maxillary segment. No periapical or periodontal alterations were detected radiographically (figure 2). Panoramic radiography also showed the double tooth 11 and rotation of tooth 12 (figure 3). However, the radiograph did not clearly define the described anomaly. To clarify the anatomy and to establish a definitive diagnosis, we referred the patient for a three-dimensional (3D)

Figure 2 Periapical radiograph of tooth 11.

Figure 1 Labial view of double tooth 11 with labial talon cusp in an 8-year-old girl.

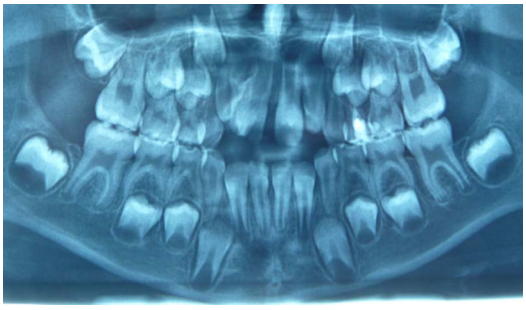

Figure 3 Panoramic radiography view.

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cone-beam CT (CBCT) of tooth 11. CBCT demonstrated the complex anatomy of tooth 11 and showed the radicular anatomy (figure 4A,B). The diagnosis was a double talon cusp (type 1) associated with a double tooth. The occlusion was Angle's Class I molar relationship on both sides.

For orthodontic purposes and complex anatomy, the treatment chosen was extraction of the tooth. Since the patient was relatively young, we started periodic selective grinding of the accessory cusp and application of fluoride varnish. When the patient was 9 years old, the double tooth was extracted (figure 5A). After 5 days into the wound healing process, the tissue re-growth stage began (figure 5B). Seven days after extraction, the sutures were removed and we took alginate impression. A transitional arc prosthesis sealed onto orthodontic bands was placed 15 days after extraction (figure 6A,B). Orthodontic treatment will begin when all primary teeth will have fallen out.

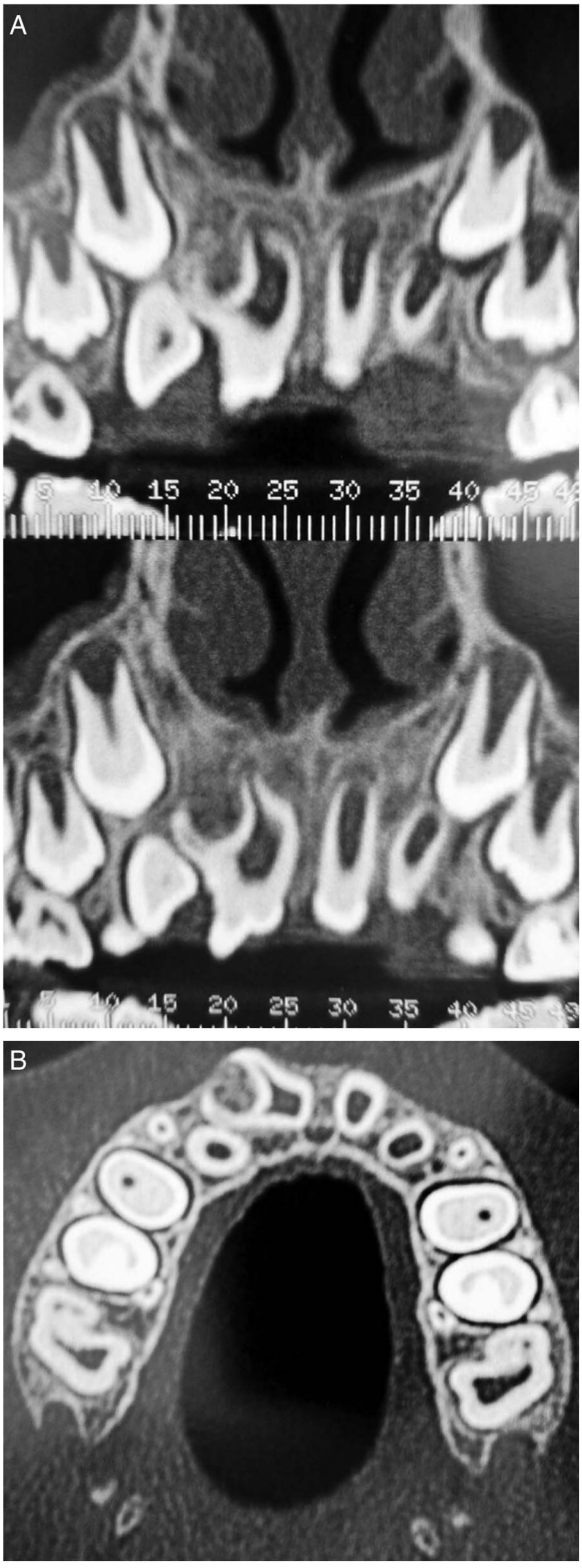

Figure 4 CBCT of tooth 11: (A) axial view and (B) frontal view. CBCT, cone-beam CT.

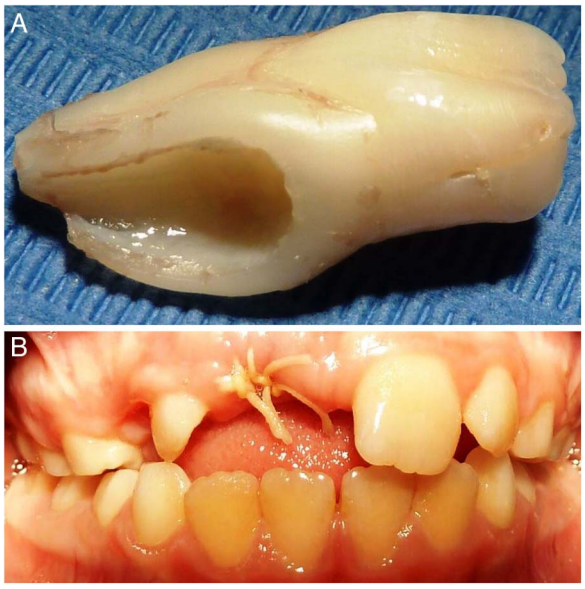

Figure 5 (A) Extracted tooth 11. (B) Frontal view 5 days after extraction.

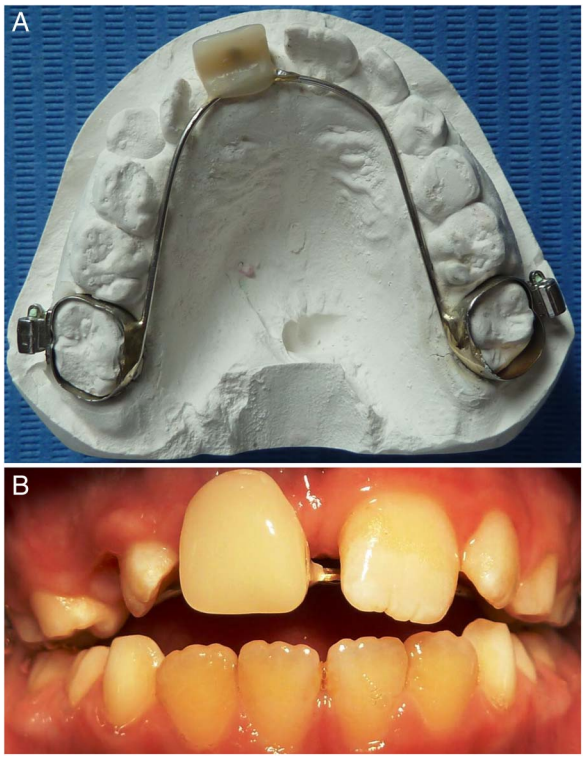

Figure 6 (A) External view of the temporary prosthesis. (B) Frontal view 2 weeks after extraction, with temporary prosthesis.

GLOBAL HEALTH PROBLEM LIST

Dens evaginatus is a coronal anomaly referring to an accessory cusp-like structure projecting from the cingulum area or cementoenamel junction of an anterior tooth. The prevalence varies from 0.04% to 10% in permanent teeth.[1] The permanent maxillary central incisor is the most affected tooth (50%).[2] Hattab *et al*[3] classified this anomaly into three types based on degree of formation and extension. Type 1 (true talon) is an additional cusp that projects towards the palatal surface to at least half the length between the cementoenamel junction and incisal edge (figure 7). Type 2 (semi talon) refers to an additional cusp of ≥1 mm in length that extends less than half of the length between the cementoenamel junction and incisal edge (figure 8). Type 3 (trace talon) is a protruding cingulum that has a tubercle-like appearance (figure 9). Type 1 is the most frequently described (52%).[4]

The aetiology of dens evaginatus is unknown and could be multifactorial, with genetic and environmental causes.[5–8] These anomalies can induce carious lesion or pulpal necrosis, occlusal interference, periodontal disease and poor aesthetics.[9 10] Diagnosis and treatment planning are difficult for the dentist, and we lack consensus on management regardless of type.

Here, we systematically reviewed the current literature describing management of permanent maxillary central incisors affected by non-syndromic dens evaginatus to analyse the different therapeutic options. As well, we report one case of this coronary anomaly.

GLOBAL HEALTH PROBLEM ANALYSIS

MATERIALS AND METHODS

Systematic review

A systematic review of the literature was performed.

Figure 7 True talon (type 1 according to Hattab's classification) on the labial aspect of a right maxillary central incisor.

Figure 8 Semi talon (type 2 according to Hattab's classification) on the lingual aspect of a left maxillary lateral incisor.

Figure 9 Trace talon (type 3 according to Hattab's classification) on the lingual aspect of a left maxillary lateral incisor.

Criteria for considering studies for this review

Eligible studies were published randomised controlled trial, comparative non-randomised study, cohort study, case series or case report describing the management of non-syndromic dens evaginatus affecting the crown of a permanent maxillary central incisor.

Search methods for identification of studies

To identify studies, we searched the database MEDLINE via PubMed for articles published in English and French, with no restriction on date of publication. Search equation combined free text words and controlled vocabulary pertaining to the condition and interventions (see online supplementary file 1). The last search for articles was in April 2015. We checked the references of all eligible articles for relevant studies and scanned reference lists from identified review articles for further studies. We searched ClinicalTrials.gov for the protocols of included studies and to identify ongoing trials.[11]

Data collection

Two authors independently and in duplicate screened the title and abstract of records retrieved by the search, then screened the selected full-text reports. Any disagreements were resolved by discussion. Finally, we included studies after eliminating duplicate publications.

For each randomised controlled trial, two authors independently and in duplicate recorded the year of publication, inclusion/exclusion criteria specified, number of arms in the trial, treatments compared, detailed description of interventions, number of patients enrolled, number of treated teeth, mean age of participants, duration of follow-up and outcome data. We assessed the risk of bias for each trial by the Cochrane Collaboration Risk of Bias tool, which includes the following items: selection of participants, blinding of participants and personnel, blinding of outcome assessors, incomplete outcome data and selective outcome reporting.[12] For non-randomised studies, we considered the risk of selection bias as high and also assessed

the risk of bias related to confounding factors. For each randomised or non-randomised trial, each domain was rated as low, high or unclear risk of bias. Then, each study was assigned an overall risk of bias score: low risk (low for all key domains), high risk (high for one key or more domains) or unclear risk (unclear for one key or more domains). The two review authors compared evaluations and resolved any disagreements by discussion. For each cohort study, case series or case report, two authors independently and in duplicate recorded the year of publication, gender and age of patients, teeth involved, type of dens evaginatus according to the Hattab *et al* classification, pulpal anatomy for the double tooth (number of the pulpal chambers and number of the roots), presence or absence of aesthetic, periodontal, occlusal or caries problems, presence or absence of associated anomaly, detailed description of treatment and requirement of endodontic treatment and orthodontic management.

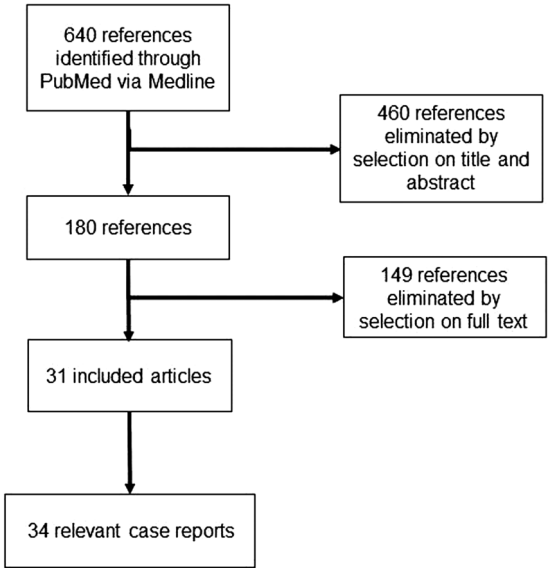

Figure 10 Systematic review flow chart of case reports describing the management of non-syndromic dens evaginatus affecting the crown of a permanent maxillary central incisor.

Analysis

We did not perform any meta-analysis, but we described the study characteristics and results qualitatively with number, percentage or mean (min–max).

RESULTS

Systematic review

Literature search

The search yielded 640 potentially eligible articles. We included 31 articles corresponding to 34 case reports.[1 13–42] The flow of article selection is in figure 10. We found no published randomised or non-randomised trial, no published cohort study and no protocol of ongoing trials at ClinicalTrials.gov.

The median year of publication was 2005 (range 1971–2014); 9 articles were published before 2000, 15 between 2000 and 2010 and 7 between 2010 and 2014.

Reported cases

A total of 21 cases involved men (60%). Across the 34 case reports, the mean age of the patients was 13.6 years. The age range varied across studies from 7 years to 47 years. Dens evaginatus was bilateral in 5 cases (15%), so 39 incisors were affected by dens evaginatus. Among the 34 cases, 12 featured dens evaginatus associated with double tooth.[16 20–23 26 29 31 34 39 40] According to the Hattab *et al* classification, the anomaly was type 1 for 32 incisors (82%), type 2 for 2 (5%) and type 3 for 2 (5%); we had insufficient information to characterise the type for 3 incisors. Localisation of dens evaginatus was lingual for 35 incisors (90%), and the condition caused occlusal interference for 25 (74%). Caries or pulpal necrosis and periodontal disease were detected in nine and four incisors, respectively (33%). For five teeth, the patient considered the abnormality unattractive (13%). Details and references of corresponding case reports are given in online supplementary file 2.

Therapeutic options for treating dens evaginatus are presented in figure 11. Details and references of corresponding case reports are given in online supplementary file 3.

For 13% of patients, the tooth with dens evaginatus without occlusal interference, irritation on the soft tissue, poor aesthetics, caries or pulpal necrosis did not need treatment. For 87% of cases, the following two options were described.

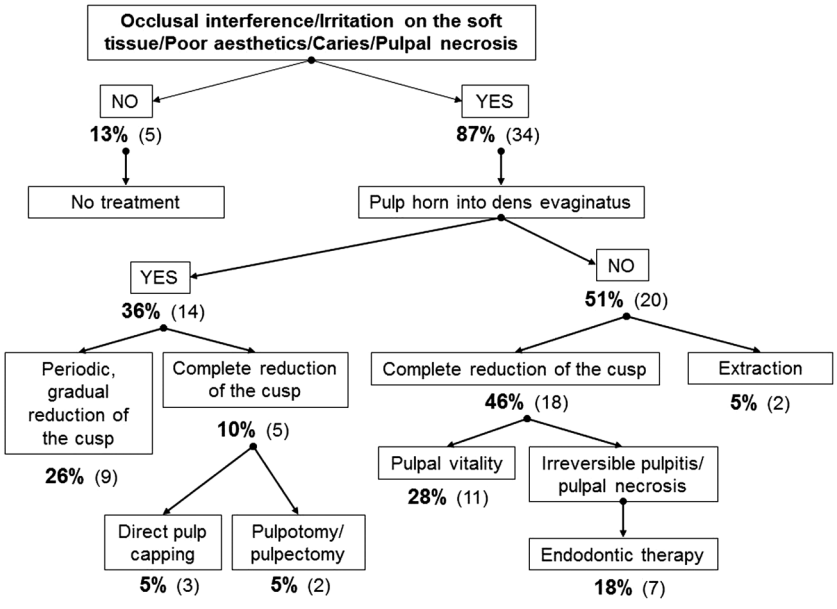

Figure 11 Therapeutic options for the 39 permanent maxillary central incisors reported in the 34 included case reports.

First, if radiographic examination showed the presence of enamel, dentin and pulp horn into the talon cusp, there were two therapeutic options. For nine incisors, the treatment plan was a gradual reduction of the talon cusp on consecutive visits and an application of a desensitising agent (fluoride) at each visit; for five of these incisors, the desensitising agent was applied without other treatment, and for one case, pulpal necrosis appeared after application of the desensitising agent. For the other four incisors, the desensitising agent was applied, followed by direct restoration at the final visit. With the second option, five patients were scheduled for complete reduction of the talon cusp in a single appointment; three incisors underwent direct pulp capping with mineral trioxide aggregate or calcium hydroxide followed by a final restoration with glass–ionomer cement then resin composite; one incisor underwent pulpotomy with formocresol and calcium hydroxide followed by resin composite application and the final incisor underwent pulpectomy.

In the second main treatment option, if radiographic examination did not show the presence of a pulp horn into the talon cusp, patients were scheduled for complete reduction of the talon cusp in a single appointment (18 incisors) or extraction of the incisor (2 double incisors exhibiting type 2 dens evaginatus). Among patients scheduled for complete reduction of the cusp, direct tooth restoration was performed in cases of pulpal vitality (11 incisors) and endodontic treatment followed by direct or indirect restoration in cases of irreversible pulpitis or pulpal necrosis (7 incisors).

Orthodontic management achieved aesthetics and occlusion in 29% of cases. In addition, 15 case reports reported the monitoring of the patient. The duration of follow-up was 12 months in median (mean 13 months, min–max 3–24 months).

DISCUSSION

To the best of our knowledge, no recent study has systematically reviewed the literature on the management of non-syndromic dens evaginatus affecting the permanent maxillary central incisor. Moreover, our case report illustrated the need for early and correct diagnosis of a talon cusp, which requires an individualised treatment plan and a multidisciplinary approach.

Dens evaginatus is a disturbance in tooth development that produces a tubercle of hard tissue on the surface of the tooth. Our results are similar to those of other authors, who reported type 1 as the most frequent.[4] The accessory cusp is most commonly located on the lingual aspect of an anterior tooth (90%) but can also be on the labial aspect or on the lingual and labial aspects of the same tooth.

On the basis of limited evidence (only case reports), our results show that dentists now have a broad range of management options for similar coronary anomalies involving a central incisor depending on a patient's pulpal features. The management of a talon cups includes no treatment (13%); sequential grinding allowing for tertiary dentin deposition, which may also be considered to remove occlusal interferences (26%); restorative treatment (28%); pulp therapy (28%) and extraction of the affected tooth (5%). We could add a preventive therapeutic option: pit and fissure sealants (not recorded in our selected case reports). Besides pulpal and radicular anatomy and treatment options, the choice of these treatments could relate to the degree of patient cooperation.[43] In addition, only 15 case reports reported the monitoring of the patient. The duration of follow-up was 12 months in median (mean 13 months, min–max 3–24 months). The therapeutic option failed in one case with a pulpal necrosis appearing after application of the desensitising agent. The other 19 included case reports did not report the monitoring of the patient. Consequently, we do not know if a therapeutic option failed or not.

Periapical radiography is commonly used for diagnosis and provides important information about the root anatomy. Nevertheless, periapical radiographs are 2D dental imaging and are not accurate or conclusive.[20 44] Moreover, CBCT provides submillimetre spatial resolution with short exposure times in the range of 20 s and radiation exposure similar to a full mouth series.[45–48] Therefore, CBCT should be systematically performed. CBCT provides 3D dental imaging and can help determine the best treatment.[49 50] CBCT allows for visualising any pulpal extension into the dens evaginatus. However, we found only two articles, published in 2014, describing the performance of CBCT.[39 42] About 30% of included articles were published before 2000, which could explain the low use of CBCT.

Our clinical case illustrates these results with an accurate diagnosis from a CBCT and follow-up. We performed extraction of the double tooth with dens evaginatus (then followed by an orthodontic management planned) as 5% of cases reported in our systematic review. Another therapeutic option would be difficult to plan, whether for aesthetic, endodontic or orthodontic problems. First, the type of dens evaginatus was three according to Hattab *et al* classification, and its localisation was lingual and labial. Moreover, complex anatomy of tooth 11 revealed by CBCT was contrary to perform an orthodontic treatment. Finally, the condition caused occlusal interference, and patient needed orthodontic management. Thus, we think that extraction of the tooth was necessary in any cases to avoid aesthetic, endodontic or occlusal problems. We did not perform a histological examination of the extracted double tooth, but it would have been interesting to send the extracted tooth to an histopathological laboratory for examination of hard and soft tissues.

Our study has some limitations. First, we identified only case reports with overall low quality of evidence and no randomised trial. Thus, the true clinical relevance of the findings is somewhat lacking. Moreover, we searched only one database and we did not search 'grey' literature, and we acknowledge that unidentified studies may exist. However, our search strategy was extensive and we consulted the largest trial registry ClinicalTrials.gov run by the US National Library of Medicine at the National Institutes of Health[51] to find unpublished randomised trials and hence the risk of publication bias in our study is low.[52 53] In addition, in spite of the fact that systematic reviews of randomised controlled trials provide the most reliable evidence about the effects of healthcare interventions, systematic reviews of case reports can be performed in some circumstances to highlight a mistreated specific medical area and stimulate further investigation.[54] For example, some authors have performed systematic reviews of case reports for cases that are out of the ordinary or for the conditions where randomised controlled trials were difficult or impossible to achieve.[55–58] Our remarks could have implications for future research. Decisions about which treatment is best are driven by the results of randomised trials and systematic reviews.[59] In this regard, high-quality randomised trials controlling for clinically relevant parameters and constraints are needed to determine what therapeutic option is a worthwhile clinical procedure for treating non-syndromic dens evaginatus affecting permanent maxillary central incisors.

CONCLUSIONS

A talon cusp is associated with problems such as compromised aesthetics, occlusal interference, tooth displacement, caries and periodontal problems. A correct and early diagnosis can prevent

these complications. The presence of a talon cusp is not always an indication for dental treatment, but the association of talon cusp with other dental abnormalities suggests a specific diagnosis for adequate care and a multidisciplinary approach.

Learning points

- A dens evaginatus is among the most challenging problems in dentistry.
- Owing to additional cusp(s) with or without pulp horn as well as misalignment, treatment usually requires a multidisciplinary approach to address endodontic and aesthetic considerations.
- Since two-dimensional periapical radiographs cannot be totally conclusive for the diagnosis, CBCT should be systematically performed in all cases because it provides three-dimensional dental imaging to help determine the best treatment.

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Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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