Australian Endodontic Journal
The Journal of the Australian Society of Endodontology Inc., the Australian and New Zealand Academy of Endodontists and the Asian Pacific Endodontic Confederation

Aust Endod J 2014; 40: 32–38

CASE REPORT

Diagnosis and management of a maxillary lateral incisor exhibiting dens invaginatus and dens evaginatus

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Keywords
dens evaginatus, dens invaginatus, fusion.

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doi:10.1111/aej.12056

Declaration: In accordance with the principles enunciated in the Declaration of Helsinki (revised 2004), parental and patient approval for publishing this report was sought and achieved. The authors adhered to QH's protocol for consent to treatment and release of records. The authors, employees of QH, applied to The Royal Brisbane and Women's Hospital Human Ethics Committee for permission to publish the manuscript. Approval was granted. The report attracted neither grant nor funding. The authors declare no financial interest.

Abstract

This case report, involving an indigenous Australian, presents the diagnosis and non-surgical endodontic management of a 22 with developmental abnormalities. They include a dens evaginatus and a dens invaginatus that extends to an apical burst in a second truncated root. Cone-beam computed tomography and endodontic microscopy were not available to the clinician. This case report focuses on ambiguities appearing in the literature relating to classification and terminology associated with the abovementioned developmental anomalies. It also demonstrates the need to methodically collect and cautiously interpret available information before initiating endodontic intervention. Axial inclination, distance perception, internal demarcation and spatial awareness, together with an understanding of dental anatomy, embryology and histology and associated physiology and pathology, allowed the clinician to accurately predict the point, the angle and the depth of coronal access. Sensibility of the dentino-pulpal complex was maintained. Critical thinking, experience, innovation, problem-solving and established principles can compensate for inaccessible technologies.

Introduction

This case report presents the diagnosis and non-surgical endodontic management of a 22 with multiple developmental abnormalities. They include a dens evaginatus (DE) and a dens invaginatus (DI) that extends to an apical burst in a second truncated root. To the authors' knowledge, this is the first documented case of DI and DE in an indigenous Australian. The innovative aspect of treatment is that the clinician, a specialist endodontist, maintained sensibility in the seemingly autonomous dentino-pulpal system. This phenomenon has been reported only once in the Australian endodontic literature and rarely in the international equivalent (1). Successful treatment was achieved without the aid of cone-beam computed tomography (CBCT) and endodontic microscopy. From the academic perspective, this case report exposes ambiguities appearing in the literature relating to classification systems and associated terminologies for the abovementioned developmental anomalies. From the general practitioner's view, it demonstrates the role of a specialist's experience in the preamble to endodontic diagnosis and intervention.

Critical thinking requires careful observation, effective analysis and objective judgment. With regard to the diagnosis, the clinician methodically collected and assessed evidence from the clinical examination, radiographs and special tests. This information, augmented by the clinician's knowledge of dental anatomy, embryology and histology, together with an understanding of associated physiology and pathology, allowed a provisional diagnosis. Paradoxically, a definitive diagnosis was elusive and largely irrelevant to treatment. With regard to the intervention, the clinician engaged difficult-to-learn concepts, namely, axial inclination, distance perception, internal demarcation and spatial awareness, to initiate the biomechanical phases of endodontic treatment. In unusual cases, clinical experience and the ability to think critically and analytically can often compensate for inaccessible technologies.

The literature relating to anomalous development of tooth number, size, shape and structure has notable features. Having a genesis in the 1950s, an era when endodontics was a young discipline, debates regarding definitions and systems for classification have generated a consensus regarding terminologies. Some early terms and definitions, for example, DI 'an infolding of the crown'; DE 'an anomalous outgrowth of tooth structure' and; fusion 'a "double" tooth resulting from the union of two adjacent tooth germs', appear in the American Association of Endodontists' *Glossary of Endodontic Terms* (2). However, anomalous dental development is an umbrella term that embraces many entities with an extensive range of morphologies. Classification systems and associated terminologies describe the developmental origins and ignore the endodontic landscape. Yet endodontists are heavily involved in treatment. Hence, confusing use of terminologies for anomalous dental developments appears in the endodontic literature.

Another attribute of the published research relating to these anomalies is multidisciplinary authorship. Surgeons, paediatricians, orthodontists, endodontists, radiologists and periodontists often focus on the management of a specific clinical circumstance and its environs. The high prevalence of some anomalies in ethnic groups has generated national streams of investigation. For instance, in Turkey, the reported prevalence of anatomical variations in endodontic morphology of maxillary lateral incisors ranges can be as high as 22% (3). Case reports frequently feature individuals of Turkish and Chinese descent. Moreover, access to dental services, which also affects research direction, varies internationally and regionally. These influences, together with associated language barriers to communication, have historically created isolated silos of ethnic-based inquiry. Accordingly, systematic reviews of anomalous dental development are often limited in scope and contain diverse opinions regarding optimal management and, to a lesser extent, terminology.

Hallett provided the first classification of 'palatal invagination of maxillary incisor teeth' (4). Based on 'degree of affection' from 'normal', it embraces fourteen subdivisions of aberrancy. Hallett acknowledged an obvious problem: 'deciding exactly what is a normal incisor, so wide were the variations discovered'. Three years later, Oehlers categorised DI according to apparent radiographic extension of the invagination (5). In simple terms, Group I are limited to the crown. Group II penetrate the crown and root. Group III penetrate the crown and root and communicate with the periodontal ligament. The pseudo-foramen, which Oehlers termed a burst, can be located either laterally or apically. Munir *et al*. have summarised alternative proposals and modifications to Oehlers' classification (6). However, its simplicity and clinical relevance have ensured its widespread acceptance in contemporary dental literature.

A brief mention of closely aligned Australian literature is worthwhile. Variations in dental anatomy, including tooth fusions, have captured Australian dentists' interest from the early 1950s (7). Barker reported dual roots in a central incisor in the *Australian Dental Journal* (8). Barker *et al*. published two of many manuscripts that related to dental anthropology, anatomy and aberrant tooth form, including DI (9,10). Mupparapu and Singer, Mupparapu *et al*., Lee *et al*., O'Reilly, Ozden *et al*. and Turker and Karaca published reports relating to anterior teeth in the same journal (11–16). Likewise, Collins, John, Low and Chan, McClelland, Pahl and George *et al*. are notable contributors to either the *Australian Endodontic Journal* or its precursor the *Australian Endodontic Newsletter* (1,17–21). Several features are worthy of comment. First, many of the reports in the Australian literature were either based overseas or involved ethnic populations resident in Australia. Second, John, who enjoyed the diagnostic benefits of CBCT, is the only author who provided endodontic management that parallels this case. However, the affected tooth in John's investigation lacked both a palatal evagination and a second root. Moreover, John did not specify the ethnic background of the patient. To the authors' knowledge, this case report is the first to document DI and DE in an indigenous Australian. Hence, this report adds to a scant stream of literature.

Case report

Located in the Brisbane central business precinct, the Brisbane Dental Hospital provides both general and specialist dental services to eligible patients. The patient, hereafter P, was a 15-year-old female with no relevant medical history. Prior dental records are sparse. A district dental officer ordered an orthopantomogram to assess periodontal bone height (Fig. 1). A radiologist coincidentally discovered and reported 'The morphology of the 22 is unusual and this is consistent with a developmental anomaly. The appearance of the 22 is most suggestive of a dens invaginatus. There is a reasonably well defined lucency lying between the 22 and 23 roots and this has resulted in some separation of the roots of these teeth. Lucency in the 22/23 region is most suggestive of longstanding inflammatory disease associated with a nonvital 22.' The dental officer's referral advised that the 22 was asymptomatic and requested 'Treatment for non-vital (distal canal) 22 – dens invaginatus evident on PA.' It also cautioned '22 responded positively to cold.'

At interview, a parent expressed two concerns. The first related to 'the future of the tooth'. The second related to

Figure 1 The orthopantomogram (OPG) accompanying the referral. Note the presence of the radiolucency near the 22, the major root and the vertical radio-opacity ascending to an apparent apical burst in a truncated root.

inconvenience. Travel to Brisbane was onerous for family members. P is one of many siblings. The parent requested a minimum number of visits. There is no familial history regarding dental anomalies and no personal history of either dental or facial trauma. The deciduous dentition had allegedly developed, erupted and exfoliated without concern. Indigenous ancestry was acknowledged. P confirmed that the 22 was asymptomatic.

The clinician used enhanced lighting (headlamp), magnification (loupes), transillumination and dental (straight and sickle) and periodontal probes to evaluate the coronal colours, contours and accessible root anatomy. The clinically visible dentition showed neither crowding nor displacement. From the labial view, the appearance of the 22 crown was normal: the vertical-to-horizontal crown ratio was harmonious. There was no aesthetic concern with balance, proportion, surface texture, shape and size of the clinical crown.

The palatal enamel generally appeared of normal texture and colour. However, a 6 mm-long clinically non-cavitated (to sharp sickle probe), inverted, U-shaped, stained fissure was present on the palatal surface. The fissure was limited to the coronal enamel. A significant evagination without occlusal interference was present (Fig. 2a). The fissure captured a brownish discoloured block of enamel. Moreover, in comparison to the 12, the 22 had subtle (maximum 2 mm) broader labio-palatal contours. A cingulum was present but the normal 22 palatal concavity was largely obliterated by the base of a 3 mm-high evagination with an apparent invagination at its peak. No swelling of either the gingiva or mucosa was detected. Pigmentation in the attached gingiva was noted.

The results of the special tests, given the radiolucency on the 22, were confusing. The 21, 22 and 23 responded positively to cold sensibility testing. The following tests were either negative or of little diagnostic value: controlled multidirectional percussion; firm palpation from labial and palatal; and mobility tests. Clinical analysis and study models identified no occlusal interferences. No parafunctional habits were acknowledged. Multidirectional and sequential periodontal probing suggested no aberrancies in the accessible root contour. No enamel-cemental fissuring either on or through the gingival margin, characteristic of a palatal groove defect, was detected.

A preoperative periapical radiograph was taken (Fig. 2b). The clinician used a commercial film holder and a modified paralleling technique in an attempt to standardise film angulation and to maximise accurate reproduction and visualisation of the apex. The radiograph confirms aberrancies in root anatomy: one major but delicate and curved root, ostensibly with an intact lamina dura and endodontic patency; and a second truncated root with a delineated outline and a diffuse and open apex. A circumscribed, periapical radiolucency appears to be associated with the 22. The curved and spindly dominant root suggests atypical crown-root angulation and dimensions. A fragile root form is present.

The radiograph also suggests abnormal coronal histology and morphology. At the palatal opening of the invagination, an ascending radio-opacity resembling that of enamel, appears to extend apically well above the level of the cemento-enamel margin. The radio-opacity, possibly surrounding either a canal or a fissure, seems fine but demarcated. The ill-defined radio-opacity across the crown suggests diffuse calcification of the dentine and minimal, if any, pulpal horns. This radiographic appearance is inconsistent with that of most 15-year-olds.

The radiographic peculiarity of the 22, in conjunction with the positive sensibility response, together with the absence of subgingival palatal grooving and the presence of localised coronal discolouration, a palatal aperture and two roots, suggested to the clinician that the dentino-pulpal complex in the main root was both autonomous and healthy. If these assessments were correct, non-surgical management of the invagination had to be considered as a serious option. Given the paucity of reports regarding these phenomena and their non-surgical management, these judgments confirm the clinician's experience and ability to think both critically and laterally.

Discussion

Diagnosis without CBCT, which option P's parent refused, was vexatious. Many characteristics of an Oehlers type III DI are present: a normal labial appearance, a palatal enamel-lined invagination with root involvement that ends at an apical burst and an exaggerated lingual

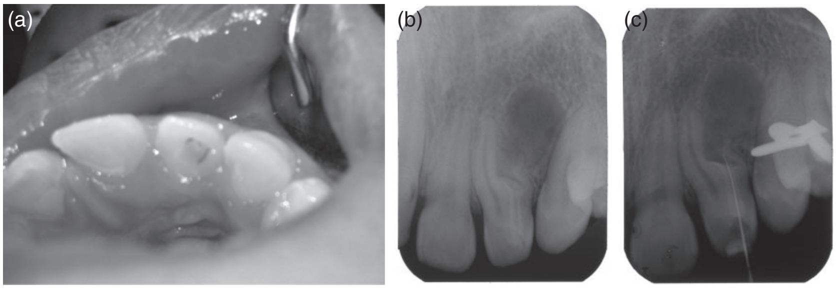

Figure 2 (a) Note the abovementioned features on the palatal of the 22. (b) The preoperative periapical radiograph confirms the presence of the radiolucency near the 22, the major root with an apparently intact lamina dura and a vertical radio-opacity with a seemingly patent and enamel-lined invagination ascending to an apical burst in a truncated root. (c) A post-access periapical radiograph showing an exploratory gutta-percha point.

cingulum. Additional supportive evidence for DI is a periapical radiolucency apparently associated with the truncated root, a positive sensibility test to cold and no overt pathological radiolucency on the main root. Moreover, given the depth of the invagination into the root, it is reasonable to suggest that inner enamel epithelial proliferation within the enamel organ and into the dental papilla during the morpho-differentiation phase of tooth development was pronounced. An aggressive in-folding of inner enamel epithelial cells could not only extend the invagination through the dental papilla to lay the foundations for the accessory root but also create a simultaneous reverse-pressure out-folding to produce the evagination carrying an aperture. Furthermore, the interface between the internal enamel epithelium and the dental papilla would be the only boundary to carry a basement membrane, which is critical to both amelogenesis and dentinogenesis. If correct, these propositions carried consequences for treatment planning. The enamel-lined aspect of the crown and truncated root did not contain a dentino-pulpal complex. The provisional diagnosis had to include Oehlers type III DI. Most importantly, the dentino-pulpal complex in the main root did not require endodontic intervention.

A diagnosis of Oehlers type III DI assumes that the anomalous dental development is derived from a single tooth bud. Some evidence suggests that a supernumerary tooth bud may have been fused onto the 22 at an early stage of tooth development. The subtle (maximum 2 mm) broader labio-palatal contours and the presence of correct tooth count are two potential indicators of fusion (22). Location is another: the maxillary lateral incisor region is a common site for a supernumerary tooth. Moreover, tooth fusion is ‘an ill-defined term’ and has been implicated in the aetiology of DI (6,14). Furthermore, the presence of the second root is inconsistent with Oehlers’ portrayal of a type III DI, which unlike Hallett’s classification, does not provide for fused combinations with a supernumerary element. These considerations were considered in a provisional diagnosis, which focused on DI and DE but did not rule out fusion.

A myriad of confounding factors affected prospects for a definitive diagnosis. The morphology of the maxillary lateral incisor is the most variable in the human dentition. For instance, Kottoor *et al*. reported ‘a maxillary lateral incisor with four root canals’ (23). Croll and Killian, Killian and Croll and Schuurs and van Loveren report shortfalls with assigning a single diagnostic term to a anomalously developed tooth (24–26). Bhargava *et al*. and Killian and Croll suggest that a diagnosis of fusion, in the potential presence of supernumerary tooth, is difficult (25,27). Bockow *et al*. concurred but resolved the issues with CBCT (28). With regard to this 22, there are major objections to a fused supernumerary hypothesis: the presence of the tubular enamel-lined invagination and the apparent absence of adjoining enamel interfaces and associated dentino-pulpal complexes. Nonetheless, without CBCT, the potential presence of a fused supernumerary lingered in the clinician’s mind.

The presence of the evagination on the 22 is also intriguing. Oehlers, who used the term DI but not DE, recorded ‘exaggerated lingual cingula’ as an integral component of type III DI (5). A plethora of alternative terms, for instance DE, ‘accessory cusp’, ‘supernumerary cusp’ and ‘talon cusp’, emerge in later literature (29). The American Association of Endodontists’ glossary not only stipulates the presence of ‘enamel, dentin and pulpal tissue’ in DE but also ignores the aforementioned alternative terms (2). Dankner *et al*. reviewed the literature regarding DE from 1970 to 1995 and stipulated that enamel, dentine and pulpal tissue have to be present in a DE (30). Seow stated that DE ‘usually contains pulp tissue which communicates with the main pulp chamber’ (31). Gehlot *et al*. assert that a DE ‘may or may not contain pulpal tissue’ (32). Hence, the use of the term DE across the literature is inconsistent.

One group of researchers has claimed prominence from this ambiguity in both definition and terminology. In the *Australian Dental Journal*, Mupparapu *et al*. stated that 'the concurrence of DE and DI within the same tooth is a rarity and has never been reported' (12). However, when compared with photographs displayed in Oehlers type III DI nearly 50 years previously, one has to question if Mupparapu *et al.'s* assertion is as distinctive as they claim (5). Agrawal *et al*., Anthonappa *et al*., Gehlot *et al*., Kiswani and Marya *et al*. have subsequently recorded the presence of DE and DI in the same tooth (32–36). Hence, if one accepts this trend in recent literature, this case report adds to a scant pool of evidence. Moreover, to the authors' knowledge, the location of the invagination at the peak of the tubercle has rarely, if ever, been reported.

Regarding endodontic intervention, the clinician made further pivotal decisions, namely, the origins and diagnosis of the problems were irrelevant to the scheme of treatment. Attention focused on problems relating to management. Did the positive sensibility result imply an autonomous and healthy dentino-pulpal complex in the main root? Did the enamel-lined DI and dentino-pulpal complex connect? Assuming the sole source of infection was the invagination throughout the truncated root, what were the consequences for obturation regarding the immature and open root apex, enamel-lined and thin apical walls and the absence of an apical stop? Was the osseous lesion either cystic or granulomatous? Did the size of the lesion warrant surgery? Should the surgical amputation and sealing of the extraneous root be seriously considered? Like the diagnosis, treatment presented challenges.

A primary approach involving apical surgery was ruled out for the following reasons. The enamel-lined invagination possibly communicated with the dentino-pulpal complex. The potential for periodontal defects, for instance surgically induced fenestration and dehiscence, was another concern. The preservation of radicular integrity of the remaining spindly and curved root, especially in the event of either occlusal overload or prosthodontic intervention, was a further priority. A high lip line, intracoronal breakdown products from necrotic pulpal tissue, apparently hyper-calcified coronal dentine, adolescent fixation with aesthetics, eventual coronal discolouration and potential effects of internally placed bleaching agents and later challenges for either a veneer or a post-retained crown: all featured in treatment planning. Hence, the resolution of endodontic issues was only one priority. The preservation of coronal and radicular dentine and the potential for long-term aesthetic concerns were others.

Understanding the nature and extent of the anomaly is a prelude to both informed consent and competent endodontic intervention. Moreover, the doctrine of autonomy, namely, the patient voluntarily decides treatment direction, is fundamental to informed consent. At this juncture, the parent for logistical reasons based on inconvenience to other family members, rejected a recommendation for a CBCT image. Accordingly, P and the parent were advised that the clinical scenario was unusual; there was some doubt about the diagnosis; endodontic intervention was recommended; conservative options would be attempted first; preliminary assessments of success for conservative modalities would require at least 6 months of observation; surgical intervention was a last resort; the 22 may be aesthetically compromised throughout teenage years; either a crown or a veneer in later life could be problematic; the 22 may have to be extracted either during or after treatment; and failure to treat 22 would probably lead to its eventual loss. Based on this advice and associated discussions, treatment proceeded.

An experienced clinician can visualise both the canal morphology and optimal end-shape of an endodontic preparation. These skills are elusive and involve distance perception (length), spatial awareness (length, width and height), internal demarcation (insight as to outline of dentino-pulpal complex and DI) and, of course, angular discernment (differentiation between long axis of tooth and long axis of entry). The incisal edge of 22 provided a fixed, constant, reproducible reference point for endodontic measurements with minimal parallax error. With a half round bur, access was made via the tip of the evagination and then followed an overt invagination.

Experience and forethought are obvious in the clinician's determination of the axial inclination of the access cavity: down the long axis of the tooth, parallel and palatal to the expected location of the dentino-pulpal complex. As suspected from assessments of the PA radiograph and to the clinician's relief, the coronal access cavity and all visible (to a depth of approximately 5 mm) radicular preparation, appeared to be surrounded by enamel. Scouting of the invagination with a gutta-percha point and an exploratory endodontic file suggested that the enamel extended almost to the apex of the second root (Fig. 2c). There was exudate but no haemorrhage. A smooth, continuous, parallel preparation provided both centrality and a glide path that facilitated straight-line access into the apical aspect of the invagination. There was a paradox. Throughout all the visits for endodontic treatment, the 22 responded to cold.

Conventional biomechanical shaping and cleaning were tailored to the circumstance. An apex locator gave inconsistent readings. Sustained tactile exploration with larger hand files again implied that most, if not all, of the radicular aspect of the preparation was lined with enamel. The clinician noted that caution was warranted

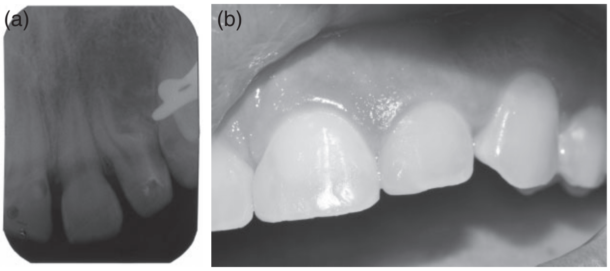

Figure 3 (a) Note the osseous healing. (b) The buccal view with minimal, if any, discolouration.

because the walls of the preparation were so hard that files could inadvertently bind. This observation was a dual-edged sword. The shortcomings were token use of nickel titanium rotary instrumentation and minimal mechanical preparation. The benefit was that, in all probability, the enamel-lined invagination formed a discrete entity. Connection to the dentino-pulpal complex was unlikely. Irrigation involved 4% sodium hypochlorite (NaOCl). A calcium hydroxide dressing was sealed in place using a temporary restorative material covered with self-cure glass ionomer cement.

After 2 months, no symptoms had developed. The 22 continued to respond to cold. Treatment of the endodontic preparation then involved irrigation with NaOCl, followed at the end of biomechanical preparation by ethylenediaminetetraacetic acid and a final rinse of NaOCl. White mineral trioxide aggregate was sealed into the radicular aspect and overlaid with a self-cured temporary restorative material and a chemical-set glass ionomer restoration. A 6 month review radiograph (Fig. 3a) and photograph (Fig. 3b) confirm osseous healing and minimal coronal discolouration. These observations lend further support to evidence suggesting the presence of an autonomous pulpal system and DI in the 22. P did not present for a 12 month review radiograph.

Conclusion

This case report demonstrates the importance of many themes in endodontic practice. The diagnosis confirms the roles of history taking, visual and tactile examinations, sensibility testing, transillumination and radiology as essential preambles to intervention. Of course, interpreting the findings, for instance, accurate assessment of dental anatomy, authoritative diagnosis of pulpal morphology and status and careful planning of intervention, require knowledge, skill and experience. The last was obvious not only in assessments of the locations and the morphologies of the pulpal system and the enamel-lined invagination but also in executing the site, outline, angulation and depth of access. The elusive skills of angular discernment, distance perception, internal demarcation and spatial awareness underpinned appropriate access to the coronal and radicular dimensions of the invagination. While there could be debate concerning the nature and classification of the anomalies, the clinician understood the clinical implications and fashioned conventional chemo-mechanical principles to the circumstance. The definitive diagnosis, probably DE and DI, is irrelevant to treatment outcome.

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