

Original article

Diagnosis of tooth ankylosis using panoramic views, cone beam computed tomography, and histological data: a retrospective observational case series study

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Summary

Objectives: The aim of this study was to determine whether cone beam computed tomography is a reliable radiological method to diagnose tooth ankylosis.

Materials and methods: A series of teeth clinically diagnosed as ankylosed were collected after extraction in a private practice from 2009 to 2015 and analyzed retrospectively. Inclusion criteria comprised permanent molars extracted due to failed tooth eruption in the absence of any visible mechanical obstruction, existing panoramic view (PV), and cone beam computed tomography (CBCT) and histological sections of sufficient quality. The CBCT scans and PVs were evaluated twice for signs of ankylosis by two independent observers using the following score: clear signs, possible signs, and no signs. The histological sections were evaluated and graded similarly to the radiographs by a specialist blinded to the radiographs and treatment.

Results: Out of an initial group of 22 patients, 9 subjects with 10 affected teeth were included for final evaluation. The age ranged from 8.3 to 17 years. No agreement was seen in comparing the PV scores to the histological sections. Fair to moderate agreement was seen in comparing the CBCT scores to the histological sections. All histologically confirmed ankylosis were detected in CBCT by both observers but some false positive results were found.

Limitation: Only a small sample size was available as the disorder is rare. It is difficult to distinguish ankylosis from primary failure of eruption.

Conclusion: CBCT images can be a useful adjunctive diagnostic tool to diagnose ankylosed teeth, but cannot be recommended as a single diagnostic modality as false positive results were found.

Introduction

Disturbance of normal tooth eruption can occur due to various factors such as mechanical obstruction, malpositioning of the tooth

bud, pathology of the dental follicle, primary failure of eruption (genetic), medical syndromes (e.g., craniofacial dysostosis, osteopetrosis, or hypothyroidism), or tooth ankylosis (1–6). Proper

diagnosis of ankylosis or primary failure of eruption, differentiation from idiopathic delayed dental development, and treatment of these conditions remain challenging (7). Furthermore, even the detailed mechanisms that result in physiological tooth eruption remain elusive (1, 3).

Ankylosis is histologically defined as fusion of cementum/dentin to bone in at least one area resulting in loss of the periodontal ligament space in that area. Fusion of bone and the tooth root leads to a vertical stagnation of tooth eruption resulting in an infraocclusion or even impaction of the respective tooth (7–10). In growing patients, it is often required to extract the ankylosed tooth to prevent deterioration of malocclusion and development of a lateral open bite due to a vertical growth inhibition of the alveolar process (11, 12). Other treatment options are luxation of the ankylosed tooth and attempt to align the tooth with orthodontic force, or, to perform a block distraction and align the tooth with its surrounding bone. However, those procedures are only described in case reports and are associated with risk of root fracture, re-ankylosis before reaching the desired tooth position or, in the case of a distraction, damaging of neighbouring structures (10,13). All those procedures are invasive and require surgical intervention. Therefore, a reliable diagnosis of ankylosis is mandatory prior to therapy.

Detection of dental ankylosis is generally established through clinical findings such as the presence of infraocclusion of the respective tooth, percussion testing, and missing tooth mobility (11). But accepted clinical tests such as the percussion testing and assessment of tooth mobility are not feasible in cases of impaction (14). As a very small ankylosed area located on the buccal or oral surfaces of the affected tooth can already inhibit tooth eruption, two-dimensional imaging such as intraoral radiographs or panoramic views (PV) are considered insufficient for proper diagnosis of ankylosis (15–17).

An accepted method of diagnosing ankylosis in impacted teeth is the lack of orthodontic movement over a defined period of time (11, 18). Applying an orthodontic force to the affected tooth requires a surgical intervention to bond a bracket or attachment to the respective tooth. In the absence of tooth mobility, and with the confirmation of true ankylosis, a second surgical intervention to extract the impacted tooth is required. If the correct diagnosis of ankylosis could be established early using three-dimensional radiographic methods such as a cone beam computed tomography (CBCT), this exploratory approach could be skipped and a second surgical intervention could be avoided. To our knowledge, only Paris *et al.* (2) examined whether an ankylosis could be correctly diagnosed using medical computed tomography (CT) and reported that three-dimensional reconstruction and analysis of CT scans enables the clinician to precisely diagnose ankylosis.

The aim of this study was to determine whether cone beam computed tomography (CBCT) is a reliable radiological method to diagnose tooth ankylosis. For this purpose, the findings of CBCT scans and two-dimensional radiographic images (panoramic views; PV) were compared to histological sections of a series of extracted teeth clinically diagnosed as ankylosed.

Material and methods

Material

The data analysed in the present study (extracted teeth, due to the diagnosis of ankylosis, as well as the corresponding two- and three-dimensional (2D and 3D) radiographs) were collected from 2009

to 2015 in a private practice specialized in oral surgery. Inclusion criteria were: permanent maxillary and/or mandibular molars extracted due to failed tooth eruption or stagnation of the continuous elongation after reaching the occlusion in the absence of any visible mechanical obstruction, thus clinically diagnosed as ankylosed; existing PV and cone beam computed tomography (CBCT) prior to treatment and existing histological sections of sufficient quality after tooth removal. Exclusion criteria were: teeth with possible previous history of dento-alveolar trauma, patients older than 30 years, invasive cervical resorption, suspicion of primary failure of eruption Typ I, genetic syndromes, or metabolic disorders (e.g., craniofacial dysostosis, hypothyroidism, hypopituitarism), and missing or insufficient quality of respective radiographs or insufficient quality of the histological sections.

The panoramic radiographs and CBCTs were all taken for treatment planning purposes prior to surgery using a Veraviewepocs R100 Pano 3D (Morita Corp., Kyoto, Japan) with a voxel size of 0.125 mm, and a slice thickness/interval of 0.125 mm.

All extractions were performed by the same oral surgeon under local anaesthesia (KD). The study protocol was approved by the institutional review board (Ethics Commission of Canton Bern, Switzerland; approval number KEK-BE 395/15). All patients included gave their informed consent to use their radiographic and biological material for further analysis.

Histological analysis

The extracted teeth were fixed in formaldehyde (4% neutrally buffered), washed in H₂O, dehydrated three times in ascending concentrations of ethanol, inserted in xylol twice, embedded in methylmethacrylate (MMA) and cut into 500-µm-thick ground sections in longitudinal and transverse axis, depending on the region of interest, using a slow speed diamond saw with a coolant (Varicut® VC-50, Leco, Munich, Germany). After mounting onto acrylic glass slides, the sections were ground to a final thickness of 80 µm and superficially stained with toluidine blue/Mc Neal combined with basic fuchsin. The histological sections were customized and stored in the Robert K. Schenk Laboratory of Oral Histology at the University of Bern.

The histological sections were evaluated by a specialist not involved in the treatment of the patients and blinded to the radiographs (DB) using the following criteria: clear signs of ankylosis (visible connection of bone to the cementum or dentin); possible signs of ankylosis (proximity of bone to the cementum or dentin and presence of resorption lacunae, but no connection/direct contact visible on the section); no signs of ankylosis (continuous periodontal gap with or without resorption lacunae). Further evaluations of the affected teeth included the presence of above average resorption lacunae or cementum hyperplasia.

Radiographic analysis

The PVs and CBCTs were assessed twice by two independent raters not involved in the treatment of the patients (MMB and FD) in an interval of at least 2 weeks on the same Dell Precision T3500 workstation (Dell, Round Rock, Texas, USA) with a 19-inch Eizo Flexscan monitor (resolution of 1280 × 1024 pixels; Eizo Nanao AG, Wädenswil, Switzerland). The radiographs were evaluated and graded similarly to the histological specimens: clear signs of ankylosis (no visible periodontal gap and visible resorption and presence of tissue replacement), possible signs of ankylosis (*only* signs of resorption with tissue replacement *or* no visible periodontal gap), and no signs of ankylosis (periodontal gap intact, no signs of resorption nor

replacement of tissue). Furthermore, it was evaluated if the affected teeth showed thickening of the cementum layer in comparison to the adjacent teeth in order to also assess the hypothesis of hypercementosis as a potential cause of non-eruption through mechanical obstruction.

Age, gender and localization of the affected tooth in the upper or lower jaw were recorded as secondary outcome variables. Orthodontic movement of affected teeth was also taken into consideration.

Statistical analysis

Cohen's kappa values were calculated in order to assess intra-rater agreement for the scoring of the PVs and CBCTs separately for the first series and the second series of image analysis. The first series of image interpretation of the PVs and the CBCTs of each observer was used for comparison with the respective histological sections calculating Cohen's kappa values (19, 20). Because of the three-level scoring, a sensitivity-respective specificity test was not feasible and descriptive statistics were used instead. All statistical analyses were performed using the Stata 14.1 statistical package (StataCorp, College Station, Texas, USA).

Results

Population

Out of the initial 22 patients, 13 did not meet the inclusion criteria and were therefore excluded from further analysis. Thus, 9 subjects with 10 affected teeth were included for the final evaluation (Figure 1).

The age of the participants ranged from 8.4 to 17 years with a male to female ratio of 7:2. The ankylosed teeth were more frequently

localized in the upper jaw (upper:lower = 6:4). In total, three out of nine patients included had two teeth affected, but only one of them had available histological records for analysis of both teeth. In three out of nine cases, an orthodontic force had been previously applied to the affected tooth (Table 1).

Radiographic and histological analysis

For the analysis of the PVs, intra-observer agreement ranged from moderate to almost perfect (Kappa for observer 1 = 0.6 and for observer 2 = 1). There was a low interobserver agreement concerning the scoring of the PVs (Kappa for 1st scoring¹ = 0.2 and for 2nd scoring² = 0.09). Because of a slightly better interobserver agreement, the first scoring was used to compare the PV to the histological sections. No agreement was seen comparing the PV scores to the histological sections, which were acting as a gold standard (Kappa for both observers was 0; Table 2).

Comparing the PV scores to the histological sections in more detail, four teeth were scored falsely as ankylosed for observer 1 and one tooth for observer 2 in the PV. In contrast, none of the two histologically confirmed teeth with ankylosis had been detected in the PV, neither by observer 1 nor by observer 2 (Table 3).

For the cone beam computed tomographies (CBCT), the intra-observer agreement ranged from moderate to almost perfect (Kappa for observer 1 = 0.54 and for observer 2 = 0.84). There was a moderate interobserver agreement concerning the scoring of the CBCT scans (Kappa for 1st scoring¹ = 0.54 and for 2nd scoring² = 0.42). Because of a slightly better interobserver agreement, the first scoring was used to compare the CBCT scans to the histology. Fair to moderate agreement were found for the comparison of the CBCT scores to the histological sections (Kappa for observer 1³ = 0.37 and for observer 2⁴ = 0.53; Table 2).

A detailed comparison of the CBCT evaluation with the histological scores showed the following: two teeth (rated the same by both observers) were scored as teeth with possible signs of ankylosis, whereas histologically no ankylosis was detected; two teeth were scored by the first and one tooth by the second observer as radiographically showing clear signs of ankylosis, but histologically, these teeth only exhibited possible signs of ankylosis. Both of the histologically confirmed teeth with ankylosis were detected in CBCT by both observers as having clear signs of ankylosis (Table 4).

Regarding the thickening of the cementum layer, two teeth presented histologically with cementum hyperplasia at the root apex. Radiographically, hypercementosis could not be clearly diagnosed, neither in PVs nor CBCTs.

Discussion

The aim of this retrospective study was to determine whether high resolution CBCT is a reliable radiological method to diagnose tooth ankylosis in direct comparison to 2D imaging (PV) and histology. In the present study, low interobserver agreement was seen in the scores of the PVs, and no agreement was evident comparing the PV scores to the histological sections, indicating that diagnosis of ankylosis in PV is not reliable and very dependent on the individual clinician's perception.

Comparing the CBCT scores to the histological sections, fair to moderate agreement was seen. All the histologically confirmed ankyloses were detected in the CBCT scans by both observers (Figure 2), but also some false positive results were found (Figure 3). This indicates that ankylosis is detectable in CBCT when present, but additional clinical findings are still beneficial for the definite diagnosis

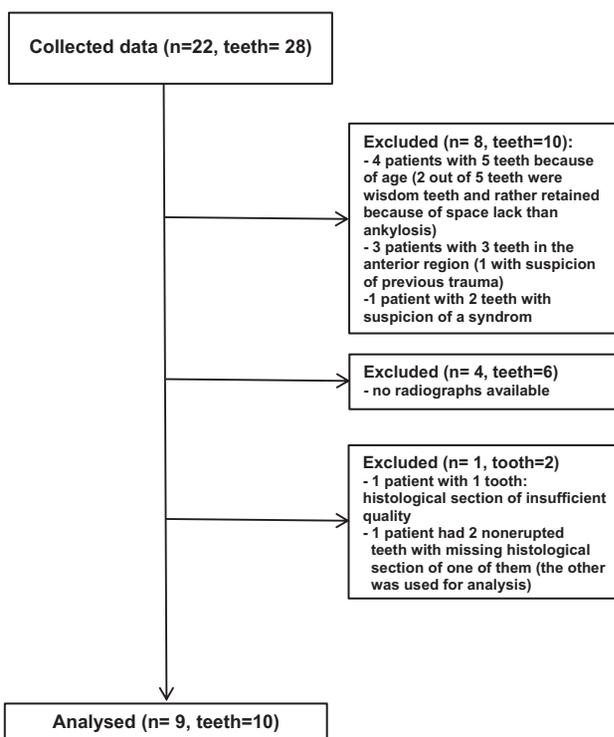


Figure 1. Enrollment of patients into the study for further analysis (according to <http://www.consortstatement.org>). *n* = numbers of patients.

Table 1. Descriptive data of the patients included in the study.

Age (years)	Gender	Upper/lower jaw	Affected tooth	Orthodontic force application
15.5	Male	Lower	46	Yes
16.6	Female	Upper + lower	(17) + 37	No
8.4	Female	Lower	46	No
16	Male	Lower	37	No
14.8	Male	Upper	16	Yes
17	Male	Upper	17	No
15	Male	Upper	17 + 27	No
11.3	Male	Upper	16	No
13.6	Male	Upper	16 + (26)	Yes

Teeth in clasps were not analysed due to missing histological records.

Table 2. Intra- and inter-observer reliability for evaluation of the PVs and cone beam computed tomographies (CBCT), as well as their agreement to the histological findings.

Measurement	Agreement (%)	Kappa
PVs (observer 1)	80	0.60
PVs (observer 2)	100	1.00
CBCT (observer 1)	70	0.54
CBCT (observer 2)	90	0.84
PVs (observer 1 versus observer 2)	60 ^a	0.20 ^a
	40 ^b	0.09 ^b
CBCT (observer 1 versus observer 2)	70 ^a	0.54 ^a
	60 ^b	0.42 ^b
PVs versus histology	30 ^c	-0.16
	50 ^d	-0.13
CBCT versus histology	60 ^c	0.37
	70 ^d	0.53

Kappa values: no agreement, <0; slight, 0–0.2; fair, 0.21–0.40; moderate, 0.41–0.60; substantial, 0.61–0.80; almost perfect, 0.81–1 [Landis and Koch (19, 20)]. For comparison of PV respective CBCT versus histology, the first scoring was chosen because of better interrater agreement.

^aFirst scoring, ^bsecond scoring, ^cobserver 1 (FD); ^dobserver 2 (MB).

because of low specificity, which could possibly lead to a false treatment or overtreatment. Furthermore, observer 2 was more experienced in diagnostic imaging (a specialist in oral and maxillofacial radiology; observer 1 is an orthodontist), which is reflected in the higher intra-observer agreement and in the higher kappa-value comparing CBCT to histology.

There are several reasons for a disturbance in tooth eruption, but two of them are clinically very difficult to distinguish without knowledge of a prior trauma, the previous treatment history or additional genetic information: primary failure of eruption (PFE) and ankylosis. In contrast to ankylosis, there is no fusion of bone and cementum in teeth affected by PFE, but a disturbance in the eruption mechanism itself causing a non-ankylosed tooth to fail to fully erupt. It primarily affects posterior teeth, especially the first molars, and all teeth located posterior to the most anterior tooth affected (7). Teeth affected with PFE do not respond to orthodontic force, and even tend to ankylose after applying orthodontic force (7, 12, 21).

Due to the retrospective study design with no information about detailed clinical findings, treatment history or genetics, teeth affected with PFE, in particular with PFE type 2, instead of ankylosis cannot be ruled out with certainty in this study. In the absence of an applied orthodontic force, those teeth would not show any signs of ankylosis—neither histologically nor in the CBCT scans. Hallmark features

Table 3. Tabulation of the scores for the PVs to the scores of the histological sections per observer.

Histo	PV				Total
	N.s.	P.s.	C.s.		
Observer 1	N.s.	2	4	0	6
	P.s.	1	1	0	2
	C.s.	2	0	0	2
	Total	5	5	0	10
Observer 2	N.s.	5	1	0	6
	P.s.	2	0	0	2
	C.s.	2	0	0	2
	Total	9	1	0	10

C.s., clear signs; N.s., no signs; P.s., possible signs.

Table 4. Tabulation of the CBCT scores to the scores of the histological sections per observer.

Histo	CBCT				Total
	N.s.	P.s.	C.s.		
Observer 1	N.s.	4	2	0	6
	P.s.	0	0	2	2
	C.s.	0	0	2	2
	Total	4	2	4	10
Observer 2	N.s.	4	2	0	6
	P.s.	0	1	1	2
	C.s.	0	0	2	2
	Total	4	3	3	10

C.s., clear signs; N.s., no signs; P.s., possible signs.

of a PFE are 1. at least one infraoccluded first molar, 2. supracrestal presentation of the infraoccluded teeth, and 3. an eruption pathway completely lacking any obstruction and clear of alveolar bone occlusal to the tooth (7, 22). In the present study, 3 out of the 10 included teeth were partially erupted and presented orthodontic attachments. All of those teeth were either upper or lower first molars. Radiographically, they presented with an occlusal step towards the neighbouring tooth with impediment of vertical growth of the alveolar bone. These signs are indicating a high probability of existing PFE, but also of ankylosis due to previous orthodontic traction.

For treatment reasons, it is important to distinguish between PFE and ankylosis (7). In case of PFE, all teeth located posterior to the first affected tooth will be also affected, whereas in a case of ankylosis the affected tooth can be removed and the space closed by moving the posterior teeth anteriorly (1, 23). For future studies, specifically genetic testing for a PTH1R-mutation could help to distinguish between PFE and ankylosis (7). Nevertheless, a negative PTH1R-mutation-test does not exclude the possibility of PFE with a different yet unknown mutation (7, 24).

In general, the following criteria can be clinically used to decide whether a tooth is likely to show an eruption disorder, such as ankylosis or PFE, and to justify the indication of an intervention: A discrepancy in tooth eruption of more than 6 months between the contralateral sides requires further radiological examination, e.g. a PV.

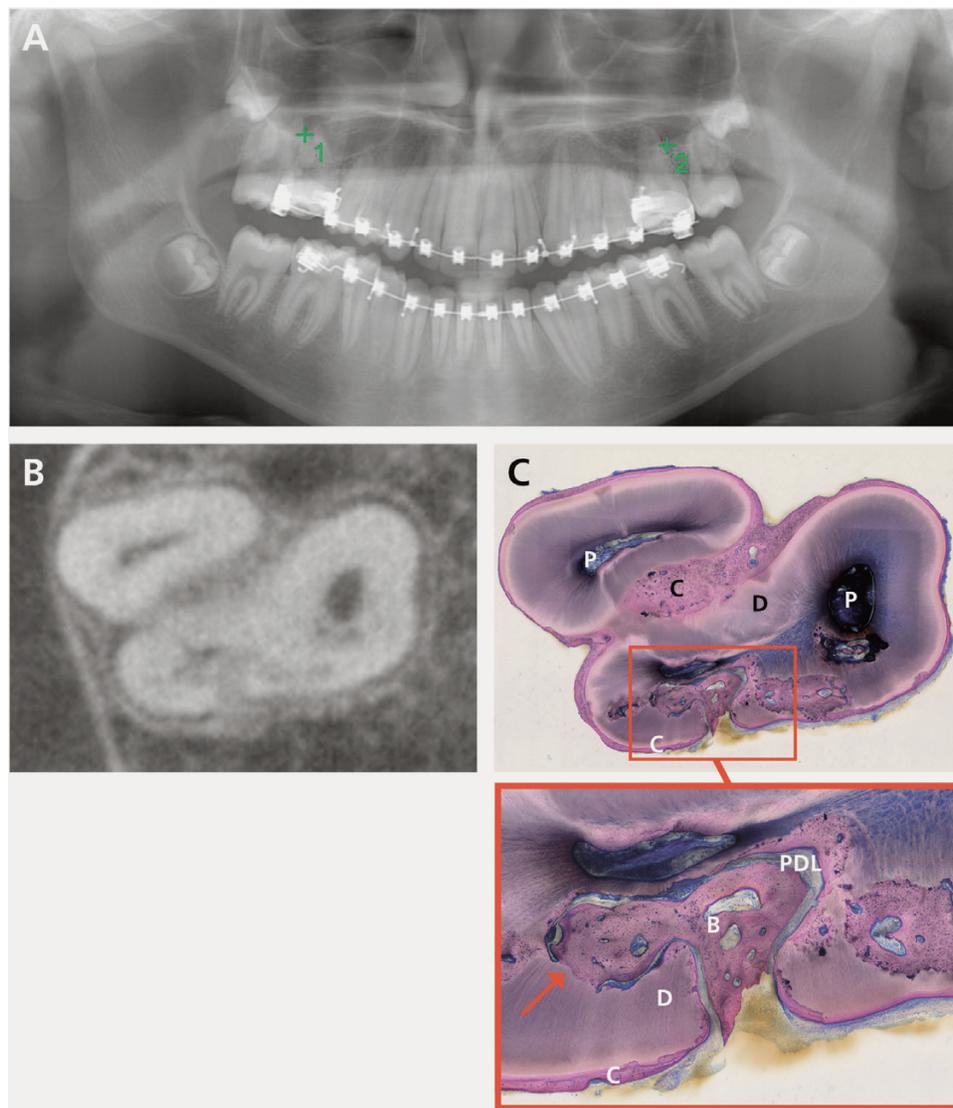


Figure 2. Suspicion of ankylosis of tooth 16 and 26 in the PV (A) (only 16 was histologically available). Clear signs of ankylosis in CBCT (B) and in the corresponding histological section (C). The ankylosis is clearly seen as a fusion of bone to the dentin (arrow). B, bone; C, cementum; D, dentin; P, pulp; PDL, periodontal ligament.

Usually teeth start to erupt when three quarters of the root has been formed and continue the root formation for two to three years after reaching the occlusion; Changes in the sequence of eruption are much more reliable for a local disturbance than a generalized delay; In growing patients, treatment is recommended as soon as severe infraocclusion of the respective tooth occurs with incipient tipping of the adjacent teeth (25).

To the best of our knowledge, only one comparable study has been published by Paris *et al.* (2), where they examined whether ankylosis can be correctly diagnosed using medical computed tomography (CT). Nowadays, CBCT is the method of choice for three-dimensional radiographic diagnostics in dentistry because of lower radiation dose and higher spatial resolution compared to the CT scans, and thereby resulting in beneficial representation of bone and teeth (26, 27). Therefore, in our study, we used CBCT and not medical CT for three-dimensional imaging. Paris *et al.* concluded that three-dimensional reconstruction and analysis of the CT scans enables the clinician to precisely diagnose ankylosis. They examined

15 impacted or retained teeth due to unknown aetiology or after unsuccessful orthodontic traction (7 anterior teeth, 3 premolars, and 5 molars). Out of the 15 included teeth, the authors assessed 2 histologically to confirm ankylosis, the other 13 were exclusively analysed radiographically by means of three-dimensional volume reconstructions. The authors rated whether there were any visible signs of resorption in the three-dimensional volume reconstructions of the CT data, which they considered as a confirmation of ankylosis. In contrast to these findings, the present study found that CBCT alone is not sufficient to diagnose ankylosis and further diagnostics are recommended (mainly a thorough dental history including possible trauma and clinical diagnostics). Because of a known superiority of analyses performed on multiplanar 2D CBCT images in comparison to measurements done on volume rendered images (28, 29), the present study exclusively used multiplanar 2D images for the assessments.

The presence of root resorption seen in radiographs or histological sections does not equal an ankylosis. In the present study, some

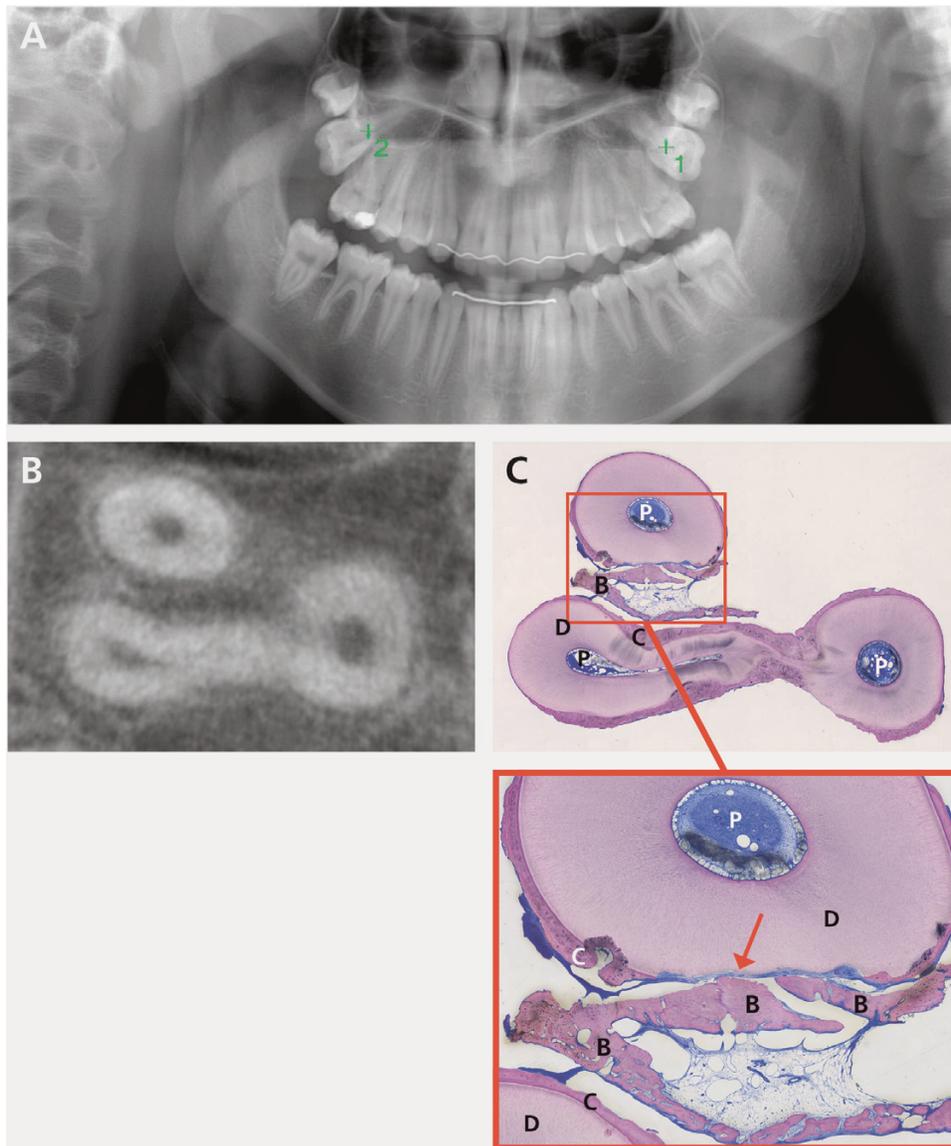


Figure 3. Suspicion of ankylosis of tooth 17 (and 27) in the PV (A) scored as possible signs of ankylosis in CBCT (B) and no signs of ankylosis in the histological section (C). The resorption of the distobuccal root extends in the dentin but is clearly demarcated from the alveolar bone with periodontal ligament (arrow). B, bone; C, cementum; D, dentin; P, pulp.

cases of root resorption were seen histologically, but the root still presented a continuous periodontal ligament (Figure 4). Spontaneous idiopathic resorption of the root surface has been described in 20% of 86 analysed premolars, extracted shortly after their eruption due to orthodontic reasons. Consequently, root resorptions are considered to a certain degree as physiologique (30–32). In a case of extensive root resorption with a pronounced resorption lacuna, without fusion of bone to the cementum and/or dentin, a mechanical retention could be a feasible reason for non-eruption, if the bone extends into the space created by the resorption process.

Even a very small ankylotic area on the root is sufficient to prevent tooth eruption, which can be easily missed in histological slices, and even more on 2D radiographs. Furthermore, through cutting teeth for histological evaluation with a sawblade followed by polishing, tooth material in between the sections gets lost, thus also eliminating possible small regions of ankylosis. This could

lead to an underestimation of the histological scoring because of non-detection of an ankylosed area. For further studies, the use of a micro-CT prior to cutting the teeth could be beneficial in regard to the detection of ankylosis in the histological sections (33).

This is a retrospective case series with some limitations. An evident limitation of the present study is the small sample size with 9 patients and 10 affected teeth after applying the strict inclusion and exclusion criteria. Failed tooth eruption of permanent first and second molars without any mechanical obstruction such as cysts or odontomas is a rare disorder with reported prevalence rates of 0.01–0.08% (34). Thus, achieving a sufficient case number in a prospective study could be difficult due to the rare phenomenon, which underlines the need for a multicentre approach. Regarding the limited statistical power of the present investigation, the results have to be interpreted with caution.

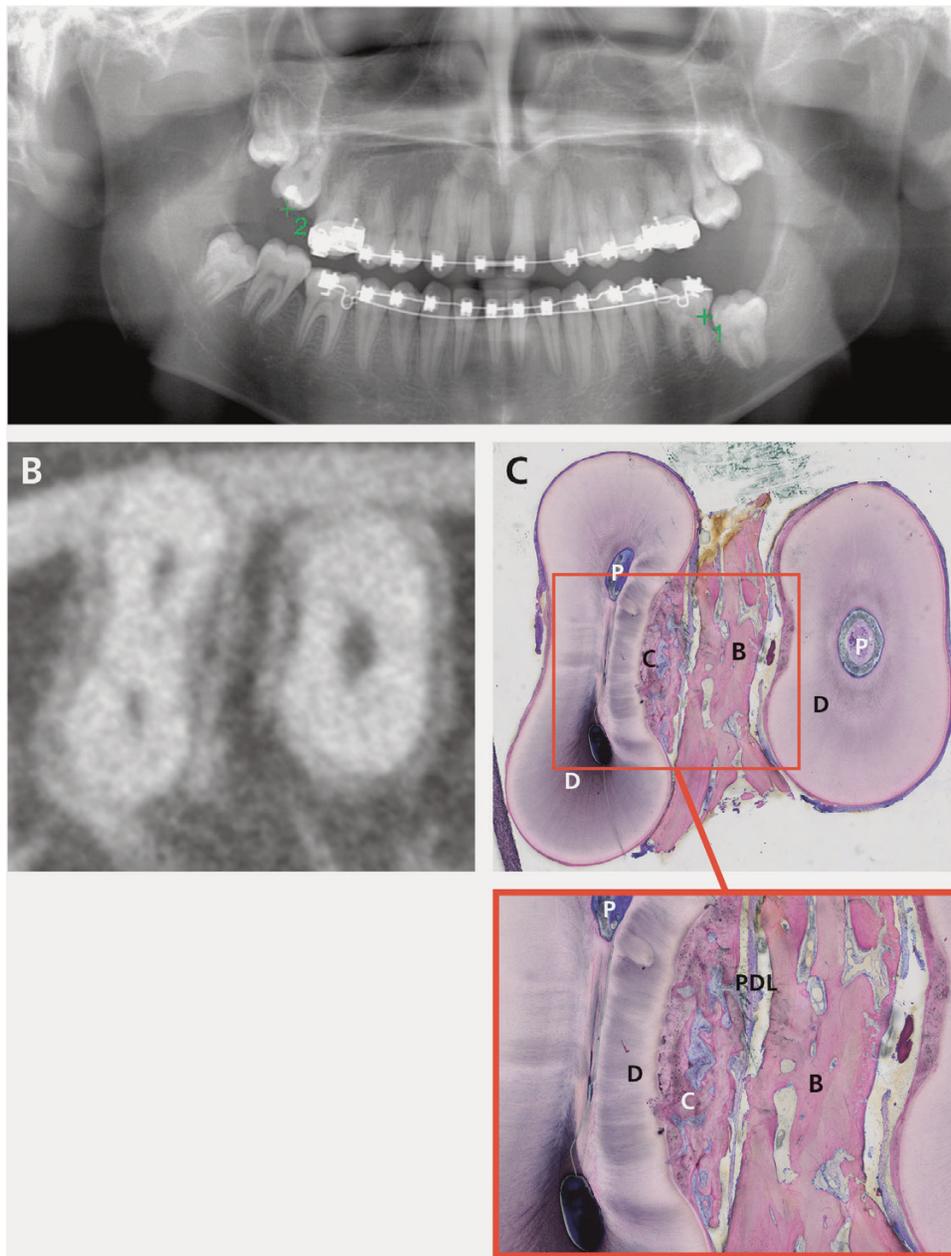


Figure 4. Suspicion of ankylosis of tooth 17 and 37 in the PV (A) (only the histological section of tooth 37 was available). No signs of ankylosis were scored in the CBCT (B) as well as in the histological section (C). Some resorption lacunae are detectable, repaired with cementum. The alveolar bone is clearly separated from the tooth by periodontal ligament. Primary failure of eruption cannot be ruled out. B, bone; C, cementum; D, dentin; P, pulp; PDL, periodontal ligament.

Conclusion

To diagnose an ankylosed tooth, CBCT images can be useful as an additional diagnostic tool supplementing clinical findings, dental and treatment history and possibly genetic information. CBCT as a single diagnostic tool is not recommended for the diagnosis of ankylosis, as some false positive results were found in the present study. PVs are considered as inappropriate for the diagnosis of ankylosis. Due to the small sample size and retrospective nature of the study, larger investigations ideally using also a multi-centre approach are encouraged.

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Conflict of interest

None to declare.

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