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Mandibular buccal bifurcation cyst treatment: report of two cases and literature review

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Key words:
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Abstract – The mandibular bifurcation cyst is an uncommon lesion that was first described by Stoneman and Worth in 1983. It is a site and age-specific lesion affecting the first or second permanent mandibular molar in children between 6 and 13 years old. Even if the clinical and radiological signs are well defined, this lesion can be misdiagnosed in particular with forms which would have a spontaneous resolution. The patient care is less well defined, the range of treatments being rather wide, from the simple follow-up to the extraction of the involved tooth. Both two clinical cases presented will illustrate the diagnostic criterias associated to the buccal bifurcation cyst. The etiopathogenesis and the therapeutic decision criterias will be discussed through a literature review.

Mots clés :
kyste de furcation vestibulaire / kyste paradentaire / mandibule / molaire mandibulaire permanente / radioclarté

Résumé – Traitement du kyste de furcation vestibulaire : à propos de 2 cas et revue de la littérature. Le kyste de furcation vestibulaire est une lésion peu commune qui a été décrit pour la première fois par Stoneman et Worth en 1983. Elle a une topographie et un âge d'apparition spécifiques en touchant uniquement les premières et secondes molaires mandibulaires permanentes chez des enfants ayant entre 6 et 13 ans. Même si les signes cliniques et radiologiques sont bien définis, ces lésions peuvent passer inaperçues notamment à certaines formes qui auraient une résolution spontanée. En revanche, la prise en charge est moins bien codifiée il existe un large éventail de traitements allant de la simple surveillance à l'élimination de la dent impliquée. Les deux cas cliniques présentés illustreront les critères diagnostiques associés au kyste de furcation vestibulaire. L'étiopathogénie et les critères de décisions thérapeutiques seront discutés à travers la revue de littérature.

The radioluciency of the jaws can have polymorphic aspects. Their discovery is made most of the time in a fortuitous way on a panoramic X-ray which must be completed by the anamnesis and the clinical exam. They cover all populations and any range of age. However, some of these entities, such as cherubism, draw our attention because they appear only during childhood.

The buccal bifurcation cyst (BBC) is an uncommon lesion that was first described by Stoneman and Worth in 1983 [1]. But it only appeared in the WHO classification in 1992. This pathological entity has been included in the inflammatory odontogenic cysts family. It is a site and age-specific lesion affecting the first or second permanent mandibular molar in children between 6 and 13 years old. It is associated with the tilting of the mandibular molars which remain vital. This lesion does not occur in the maxilla, nor in adults and can be bilateral [2-4].

This article aims at describing two new clinical cases of the same pathology but different in presentation, and for which a conservative surgical approach was adopted, with almost 2 years of follow-up. A literature review will permit to better understand the etiopathogeny of this particular cystic lesion and the various types of treatments available.

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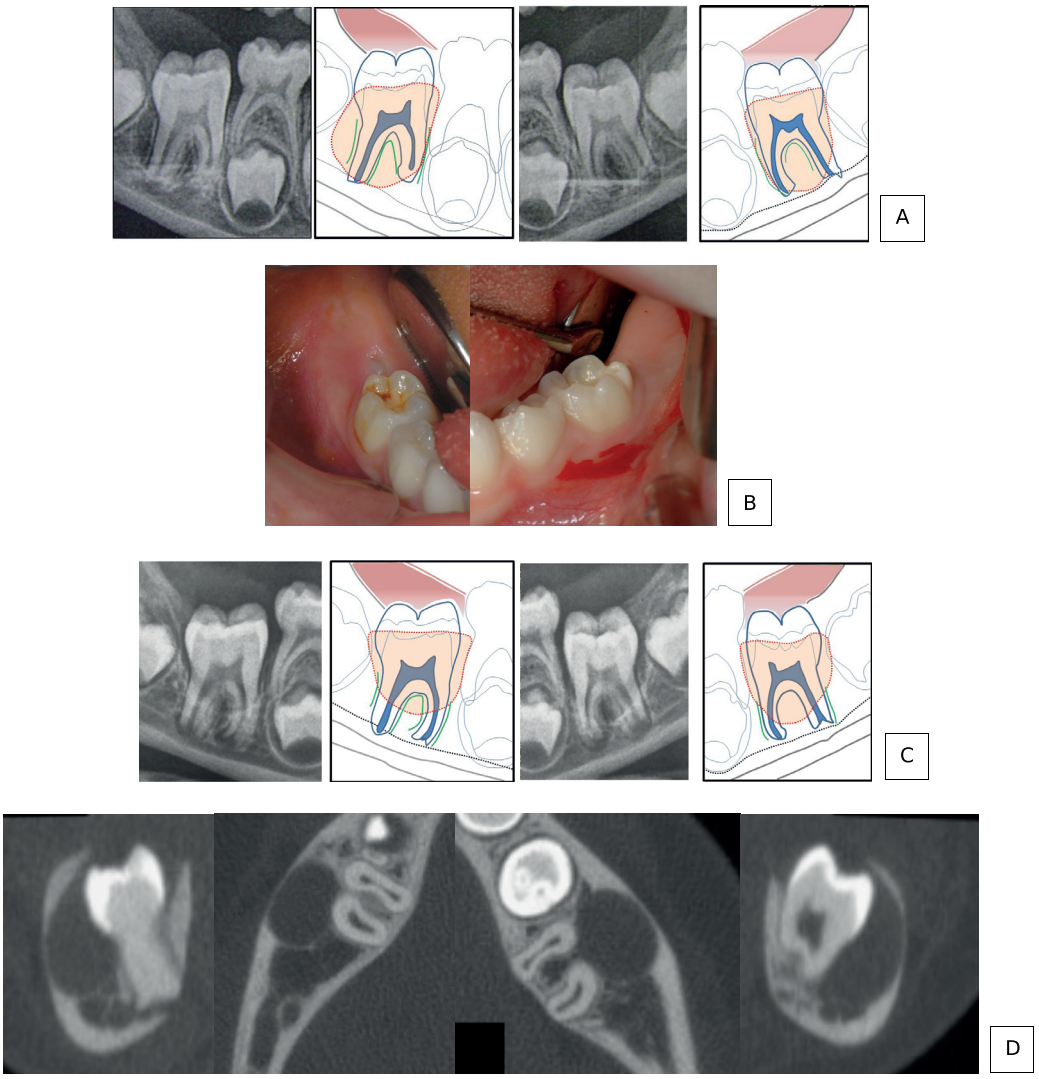

Fig. 1. Case 1: preliminary clinical and radiographic features. A: panoramic X-ray enlargements and their corresponding drawing centered on teeth 36 and 46 (september 2009); radiolucencies are very discreet. B: intraoral views showing the absence of the 36; only the mesiolingual cusp of the 46 is visible in the oral cavity. C: panoramic X-ray enlargements and their corresponding drawing centered on teeth 36 and 46 (april 2010); U-shape radiolucencies. D: axial and frontal scan-CT views (avril 2010), buccal radiolucencies lesions are associated to the roots of the 36 and 46 with a buccal tilting of the crown in the frontal plan.
Fig. 1. Cas 1 : examens cliniques et radiographiques initiaux. A : agrandissements du panoramique dentaire et schémas correspondant centrés sur les zones de 36 et 46 (septembre 2009) ; présence d'images radioclaires très discrètes. B : vue endobuccale montrant l'absence de la 36, seule la cuspide mésio-linguale de la 46 est visible. C : agrandissements du panoramique dentaire et schémas correspondant centrés sur les dents 36 et 46 (avril 2010) ; présence d'images radioclaires plus marquées en forme de « U ». D : coupes scanner axiales et frontales (avril 2010), lésions vestibulaires radioclaires en rapport avec les racines de 36 et 46, associées à une inclinaison vesibulaire des couronnes dentaires dans le plan frontal.

Case 1

An 8-year-old girl was referred by her orthodontist for delayed eruption of the first mandibular molars. Upon extraoral examination, no asymmetry and no swelling were noted. The girl did not complain on the palpation of the area or on chewing. The patient had a normal dentition for her age, except for the absence of the two first mandibular molars. All of the other teeth were asymptomatic. Only the mesiolingual cusp of the lower right mandibular molar was visible in the oral cavity. The overlying mucosa, including the buccal gingiva, appeared within normal appearance and no suppuration was noted. But we noticed an enlargment of the alveolar processes by bidigital palpation in the two posterior areas (Fig. 1).

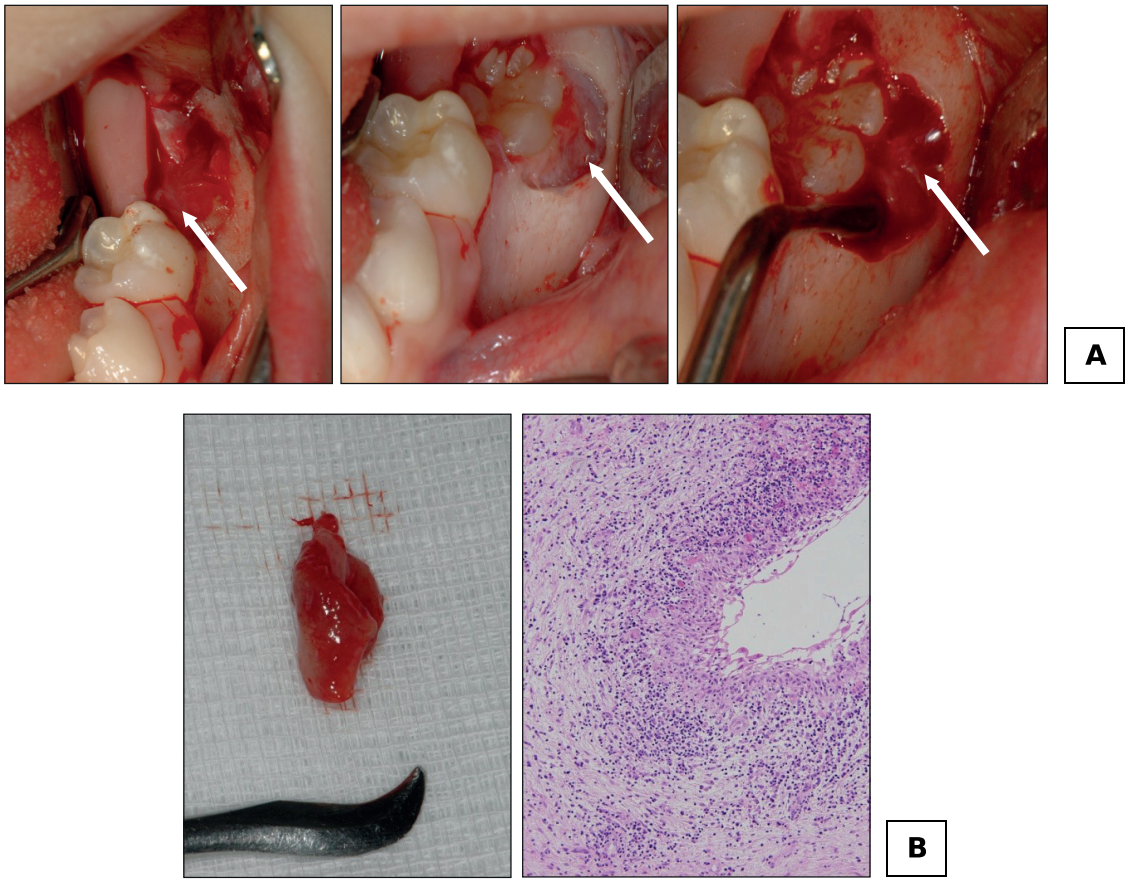

Fig. 2. Case 1: surgical management. A: left to right, full-thickness mucoperiosteal flap with peridental follicle, external cyst aspect after osteoectomy and cyst enucleation; B: macroscopic and microscopic (× 200) views of the BBC. The walls were made of fibrous connective tissue and lined by a non-keratinized stratified squamous epithelium of various thickness and morphology, according to the extent of inflammation; the cyst showed dense, chronic inflammatory cell infiltration and vascular proliferation.
Fig. 2. Cas 1 : phase chirurgicale. A : de gauche à droite, visualisation du sac péricoronaire après élévation d'un lambeau muco-périosté d'épaisseur totale, aspect externe du kyste après ostéoectomie, énucléation kystique. B : aspects macroscopique et microscopique du kyste de furcation vestibulaire. La paroi était composée d'un tissu conjonctif d'aspect fibreux et délimitée par un épithélium malpighien non kératinisé d'épaisseur et de morphologie variables en fonction de l'importance de l'inflammation ; il était noté la présence d'une inflammation chronique avec une infiltration cellulaire et vasculaire dense.

In panoramic radiograph, the aetiology was unknown. Only discreet bilateral radiolucent lesions with a sclerotic ring in the bifurcation area were seen in the mesial and distal buccal parts of the crowns, encompassing the roots. However, the teeth associated with the lesion seemed to be caries-free and had a normal morphology, but incomplete apices. The inferior border of mandible was intact. An increased prominence of lingual cusps showed a buccal tilting of the crowns.

Axial cone-beam CT view demonstrated well-defined radiolucent lesions involving the buccal area of 36 and 46 with bony expansion and thinning of the buccal cortex. An anterior view of a 3D cone-beam CT reconstruction showed the close association of the well-circumscribed lesions with the buccal bifurcation area of the first mandibular molars and the characteristic tilting of apices toward the lingual cortex.

Surgical management was performed under general anesthesia due to several considerations like the young age of the girl, the bilateral location of the lesions, the limited access and the proximity with the dental mandibular nerve. The operation consisted in a bilateral cyst enucleation with tooth conservation (Fig. 2). That was a conservative approach to allow a physiological dental and bone development. After reflexion of a full-thickness mucoperiosteal flap and eviction of peridental follicle, the blue color of the cystic membran was noticed, confirming the buccal location of the cyst. It was removed and involved molar were left in situ. The flap was repositioned with resorbable sutures.

Histological features were similar to those of the other inflammatory paradental cysts. The walls were made of fibrous connective tissue and lined by a non-keratinized stratified

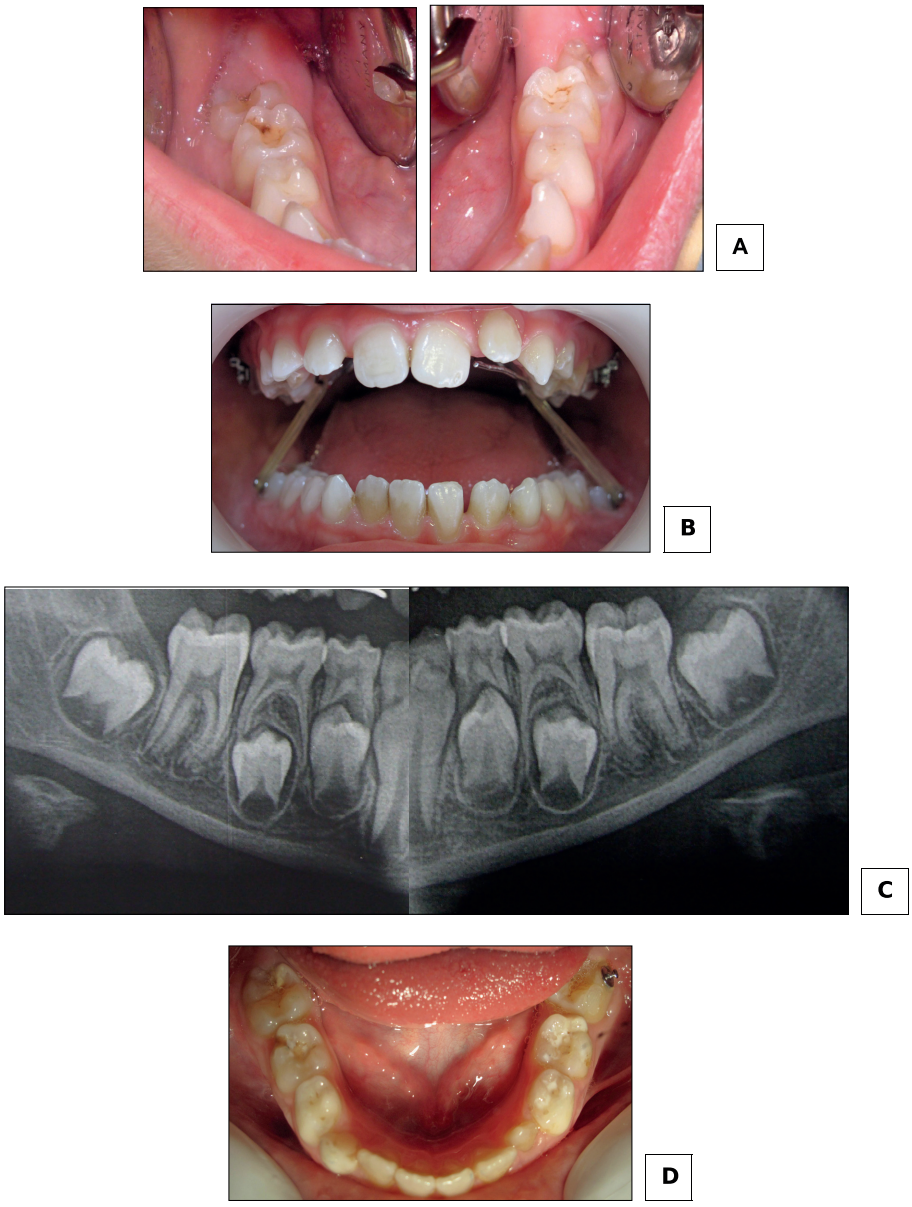

Fig. 3. Case 1: follow-up. A: intraoral view 2 months later, partial eruption of 36 and 46. B: orthodontic appliance, quad-helix and traction elastics. C: panoramic X-ray, 10 months later, demonstrating a full bony healing and a quite normal eruption pattern. D: intraoral view, 18 months later, physiological position of 36 and 46.
Fig. 3. Cas 1 : suivi. A : vues intra-buccales deux mois après l'intervention, éruption partielle de 36 et 46. B : appareillage orthodontique avec quad-hélix et élastiques de traction. C : panoramique dentaire montrant, dix mois après, une bonne cicatrisation osseuse et une éruption dentaire quasi-normale. D : vue intra-buccale dix-huit mois après, 36 et 46 sont en position physiologique.

squamous epithelium of various thickness and morphology, according to the extent of inflammation. The cyst showed dense, chronic inflammatory cell infiltration and vascular proliferation (Drs Marcellin and Renard, Strasbourg).

Intraoral examination after 2 months showed only partial eruption of the mandibular first molar (Fig. 3). However, the buccal tilting was still present. Involved teeth remained vital. Orthodontic traction of the first molar allowed achievement of eruption into a rather physiological position in about 6 months. Panoramic radiograph 10 months after enucleation demonstrated bony fill, healing of the lesions and normal eruption pattern. We still noticed the persistence of buccal tilting of the crowns.

The control 18 months after surgery revealed a complete regression and no recurrence of the cyst.

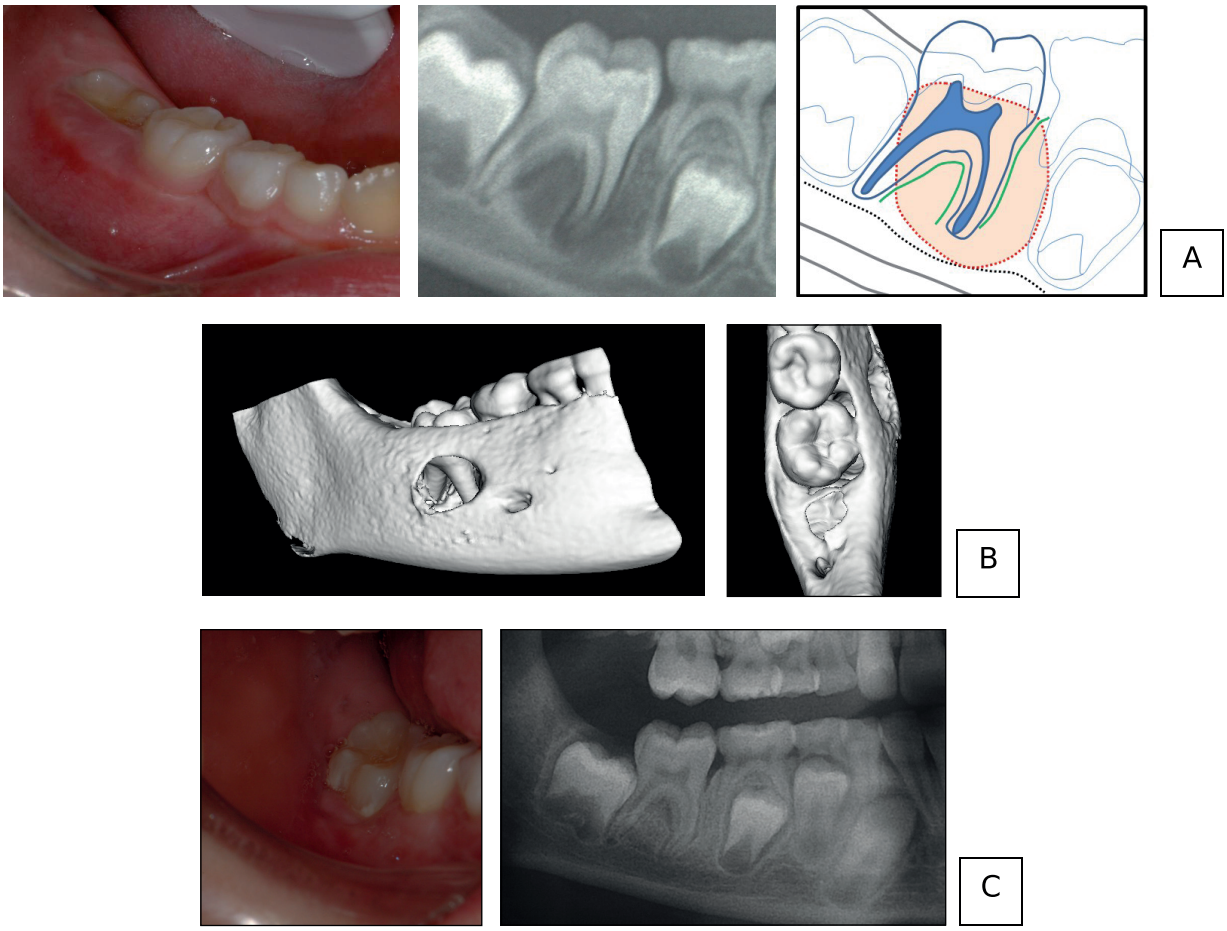

Fig. 4. Case 2. A: initial situation, partial eruption of 46, associated to a radiolucency lesion on panoramic x-ray. B: 3D reconstructions showing a fenestration of the external cortical, associated to the buccal tilting of the crown. C: intraoral view, 2 months after surgical management, showing eruption of the lower right first molar and panoramic radiograph, 3 months later, demonstrating a full healing of bone.
Fig. 4. Cas 2. A : situation initiale, éruption partielle de la 46 associée à une image radioclaire visible sur le panoramique dentaire. B : reconstruction tridimensionnelle montrant une fenestration de la corticale vestibulaire de la mandibule associée à une inclinaison vestibulaire de la couronne de la 46. C : vue intrabuccale, deux mois après l'intervention, montrant l'éruption de la 46 et panoramique dentaire, 3 mois plus tard, indiquant une cicatrisation osseuse complète.

Case 2

A 9-year-old boy consulted for delayed eruption of the lower right molar with history of pain and swelling. Intraoral view showed partial eruption of the lower right molar and initial panoramic radiograph revealed a well defined unilocular radiolucency covering the root area of 46 (centered on the mesial root) and responsible for the displacement of the second premolar. Axial and 3D-conebeam CT views demonstrated attachment of the cystic lesion to the area of tooth furcation and the perforation in the external cortical in front of the mesial root (Fig. 4). The buccal tilting of the crown was seen too.

Surgical management was also performed under general anesthesia and consisted of cyst removal with conservation of the involved tooth.

Two months after surgery, intraoral view showed eruption of the lower right molar with persistence of the buccal tilting of the crown. Involved tooth remained vital. Panoramic radiograph two months after enucleation showed bony fill and ongoing healing of the lesion.

Discussion

The aetiology of the BBC is still debated, and several theories have been proposed. Developmental theories support that the cyst could have originated from the crevicular epithelium, the cell rests of Malassez, the reduced enamel epithelium or the dental follicle. A local inflammatory stimulus beneath an epithelial junction seems to have an important role in epithelial

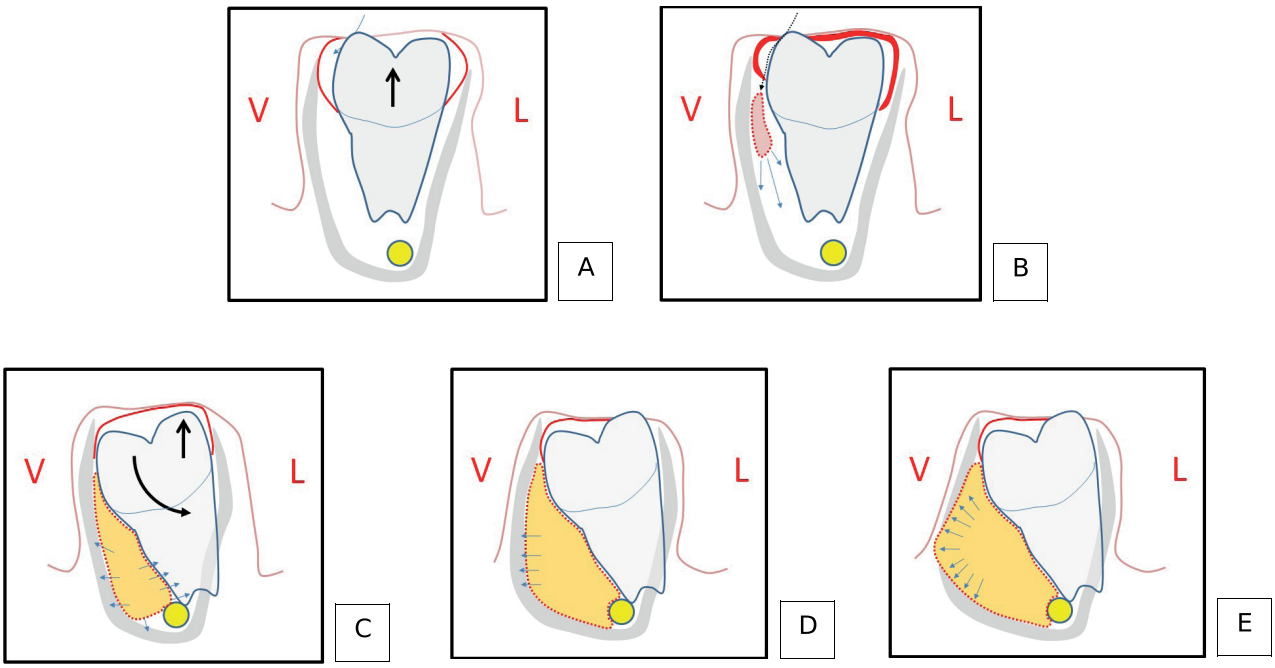

Fig. 5. Different stages of a BBC's development: theoretical hypothesis. A: physiological eruption of the mesiobuccal cusp through the epithelium. B: local inflammation of the epithelial attachment area. C: BBC's development, leading to a buccal tilting of the crown and a proeminence of the lingual cusps. D: extension of the BBC to the external cortical. E: fenestration of the external cortical.
Fig. 5. Hypothèse physiopathogénique pour les étapes du développement d'un kyste de furcation vestibulaire. A : éruption physiologique de la cuspide mésio-vestibulaire à travers l'épithélium buccal. B : inflammation locale située en regard de la zone de l'attache épithéliale. C : développement du kyste de furcation vestibulaire menant à une inclinaison vestibulaire de la couronne dentaire s'accompagnant d'une proéminence des cuspides linguales. D : extension du kyste vers la corticale vestibulaire. E : fenestration de la corticale vestibulaire.

proliferation. During molar eruption, the mesiobuccal cusp is the first to break through the epithelium. That induces a microscopic communication between the pericoronal space and the oral environment, which could lead to an inflammation localized in the epithelial attachment area, leading to the BBC's development [5, 6] (Fig. 5). This can easily explain the buccal tilting of the crown seen later on X-rays.

Although the BBC is included within the inflammatory paradental cysts family in the WHO classification of odontogenic cysts and tumors, its existence as a distinct entity is not accepted universally because of correlation of clinical, radiographic, surgical, and histologic findings with other lesions. The presence of radiolucency associated with the permanent mandibular molars in children should include the BBC in the differential diagnosis with the following lesions:

- a paradental cyst of the third molar tooth extending to the first molar;
- a keratocyst: it generally affects adults (30–50 years old) and can be found in the maxilla or in the mandible whereas BBC only exists in the mandible of children;
- a dentigerous cyst: the radiolucency is usually associated with the crown of the tooth, whereas the BBC's radiolucency surrounds the furcation and the roots of the tooth;
- a lateral periodontal cyst: it is very rare before the age of 20 and is usually located in the anterior area of the maxilla.
- an ameloblastoma: it usually involves the angle of the mandible and presents a typically radiological appearance like bubbles of soap.

In fact, histology of BBC is non specific because its features are similar to those of the other inflammatory paradental cysts. Today, the place of the BBC in the WHO classification seems to be unclear. The BBC is defined as a paradental cyst because localized on the lateral or other aspect of the root of a tooth but affects only mandibles of children, that's why the BBC could create a new specific entity in the WHO classification.

Several therapeutic approaches have been described including marsupialization, enucleation and curretage of the lesion associated or not with extraction of the involved molars. A more conservative non surgical approach (daily irrigation of the pocket with saline) has also been reported [5, 7]. Marsupialization can be useful to reduce the volume of a big cyst to facilitate its enucleation later on. But usually, surgical management by cyst enucleation without extraction of the involved tooth must be the treatment of choice. The extraction of the tooth associated to the cyst is appopriate when the lesion is very voluminous and when its resection compromises

Table I. Therapeutical approachs for BBC.
(CE: cyst enucleation; TP: tooth preservation; TE: tooth extraction; BBC: buccal bifurcation cyst; Mpz: marsupialization)
Tableau I. Approches thérapeutiques pour le kyste de furcation vestibulaire.
(CE : énucléation kystique ; TP : conservation de la dent ; TE : extraction de la dent ; BBC : kyste de furcation vestibulaire ; Mpz : marsupialisation)

	Vedtofte and Praetorius [13]	Packota et al. [14]	Wolf and Hietanen [15]	Pompura et al. [3]	Shohat et al. [6]	Thurnawald et al. [16]	Lacaita et al. [17]	Glock et al. [18]
Number of BBCs	13	5	6	44	5	13	14	2
First molar	6	5	3	44	3	13	12	1
Second molar	7	0	3	0	2	0	2	1
Age	7–15	6–8	6–14	5–11	8–13	5–9	6–9	Unreported
Surgical management	CE + TE	CE + TP	CE + TP for 4 BBCs CE + TE for 2 BBCs	CE + TP	CE + TP for 3 BBCs CE + TE for 2 BBCs	CE + TP for 9 BBCs Mpz for 1 BBC	CE + TP for 14 BBCs	TE + CE
Recurrence and follow-up	2 at 10–12 months	None	None at 6 years	None at 3 years	None at 3 years	None	None at 2 years	None

the sustainability of this tooth. The extraction of the causal tooth can be also prescribed in case of an agenesis or of a serious decay of the controlateral tooth to allow a symmetric development of the mandible [8].

The study of Yavuz et al. concerning 165 teenagers from 13 to 18 years old, to whom the unilateral extraction of a first permanent molar was necessary, demonstrates that, in 75% of the cases, the development and the implementation on the arch of the second and the third molar are accelerated compared to the controlateral side. In about 22% of the cases, this phenomenon remains identical on both sides. The extraction of a first mandibular permanent molar seems therefore to be an interesting therapeutic option, given the good implantation of the following teeth on the lower jaw [9]. When the tooth is present on the arch, its extraction is not prescribed when it is linked to a reduced size cyst.

On the other hand, De-la-Rosa-Gay et al. present two additional contraindications to the extraction of the tooth concerned by a BBC: a stage of Nolla superior to 8 (two-third of the radicular apexogenesis) or an angulation superior to 30 degrees. On the 74 second permanent mandibular molars, in 5.4% of the cases, the third molar did not achieve its eruption. Besides, in 19% of the cases, the third molar presented a defective proximal contact [10].

Let us also note that the germs of the wisdom teeth appear at about 8.5 years old on the panoramic X-ray, which is often the period when the therapeutic decision of the practitioner has to be known. Therefore, if wisdom teeth are absent, it seems hardly conceivable to extract the molar linked to a BBC because it would result in a short dental arch.

The therapeutic option of extraction must be considered only as a last resort because the extraction of a mandibular permanent molar in children can lead to an asymmetry, occlusion disorders, and masticatory dysfunction [6].

Taking advantage of the surgical operation, it is also possible to proceed to the cystic enucleation followed by the extraction of the tooth and then the immediate reimplantation in a physiological position. As the roots are not apexified, a phenomenon of pulpal revascularization can arise. An interesting alternative is the cystic enucleation associated with a simple luxation of the tooth concerned in a physiological position [11]. This one is then maintained by a flexible contention during about ten days, to compensate for the buccal pathological slope in the frontal plan, and can dispense the child from a later orthodontic treatment. However, this technique can present some risks: pulpal necrosis, ankylosis and root resorption [12]. The table I shows different therapeutical approaches and their follow-up, according to several authors [3, 6, 13-18].

Conclusion

BBC is a pediatric entity that involves the buccal region of the mandibular first molar and usually appears mainly in the first decade of life. It is localized near the furcation of the first or second vital permanent mandibular molar. The positive diagnostic can be made using epidemiological, clinical and radiographical criterias, because of a non-specific histology. Surgical management by cyst enucleation without extraction of the involved tooth seems to be the treatment of choice. A clinical and radiographical following after several months is essential to check the healing of the mandible. According to the position of the teeth obtained, an orthodontical treatment can be appropriate.

Competing interests: none

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