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Clinical report

Traumatic bone cyst and congenital muscular torticollis: Association or a chance?

Quiste óseo traumático y tortícolis muscular congénito: ¿Una asociación o una posibilidad?

Introduction

The traumatic bone cyst, also called simple bone cyst, hemorrhagic bone cyst, solitary bone cyst and idiopathic bone cavity[1–11] has been described for first time by Lucas in 1929 and since then this lesion has attracted great interest in dental literature for its unclear pathogeneses.[1] Although many theories have been exposed, none of them explains all the clinical and pathological characteristics of the lesion.[10] The traumatic–hemorrhagic theory seems to be the most accepted. Other theories include the inability of interstitial fluid to exit the bone due to blockage of drainage system, bone growth and development disorders, ischemic necrosis of medullary bone and local changes in metabolism resulting in bone osteolysis.

The World Health Organization rates traumatic cysts as non-neoplastic lesions, because of not having epithelium as true cysts. It usually occurs in the metaphyseal region of long bones and it is unusual in the maxillofacial region, with a prevalence of 0.5–1.2% of all jaw cysts.[1–5] Usually a little or no tissue is obtained for the histopathological diagnosis. The definitive diagnosis is mainly based on clinical and radiographic features,[4,9] along with surgical findings.[4]

The traumatic bone cyst is a benign intraosseous cavity characterized by an empty bone cavity or containing liquid, devoid of epithelial lining, sometimes clinically presented with a painless swelling in the affected area. When it affects gnathic bones, it mainly attacks the mandibular region, between the canine and third molar teeth.[6,7] The traumatic bone cyst usually appears in individuals in the second decade of life[8] and approximately 60% of cases occur in male patients.[4]

Several treatment modalities have been reported, including resection, curettage, bone grafting, corticosteroid injection and, more recently, injection of autologous medullary bone. However, surgical exploration of the cystic cavity has been recommended.[4,5,9] It is believed that in some cases there may be a spontaneous resolution.[9] On the other hand, other authors have suggested that treatment with a single punch and/or aspiration of the cyst content is sufficient for regression and treatment of the cyst.

Congenital muscular torticollis (CMT) is a condition characterized by contralateral deviation or vicious head position and progressive appearance of facial and cranial asymmetry in most cases.[14,15] It occurs due to the rupture of the sternocleidomastoid muscle and fibrosis in uterus or during birth. Treatment varies for each case, being physiotherapeutic or surgical.[13]

Possibly the case of traumatic bone cyst to be reported may be related to CTM associated with trauma during birth, or be a consequence of sternocleidomastoid muscle tension, which probably caused a local disturbance in mandibular growth and development.

Case report

A male patient, aged 13, was referred to the São José dos Campos Dentistry School – UNESP, for the review of a radiographic finding during a survey of third molar in August 2011. During anamnesis it was reported that the child had a history of congenital muscular torticollis, which had been treated surgically after birth, with myotomy of the sternocleidomastoid muscle. In the clinical analysis, it was found that the mandibular lesion was asymptomatic and there were no signs of mucosal alterations or swelling of cortical bone. The teeth vitality test was positive. A panoramic radiography revealed a well-circumscribed unilocular radiolucent lesion extending up the distal roots of the teeth 44, 45 and mesial of 46 to the base of the mandible (Fig. 1).

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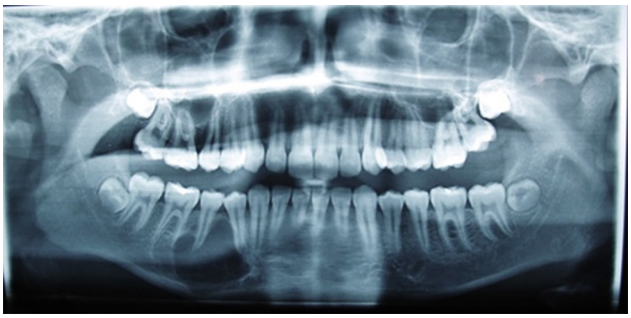

Fig. 1 – Initial panoramic radiograph.

One week after analysis of radiographic findings and physical examination, an incisional biopsy was scheduled. The pattern procedure of intra- and extra-oral antisepsis and installation of drapes was performed, followed by local infiltrative anesthesia, mucoperiosteal incision, exposure and osteotomy to perform puncture aspiration. The contents of the cyst were collected, which was a bright bloody fluid (Figs. 2 and 3).

After the puncture aspiration, bone cavity was inspected in tentative to obtain material for pathological examination. However, it was not possible to identify any fragment or granulation tissue that could be consistent with a possible cystic capsule. Bone walls were free and undamaged. The study proceeded with the irrigation of the cavity with physiological solution and flap closure with sutures. The patient had no postoperative complications and was instructed to visit weekly during the first month and every month from the second month.

In 2012 during of return evaluation, it was reported that a second surgery was performed, for muscle relief of sternocleidomastoid with the objective to improve the movement of the neck. In 2013, after 2 years follow-up, the remission of signs and bone repair of the mandibular lesion was observed (Fig. 4).

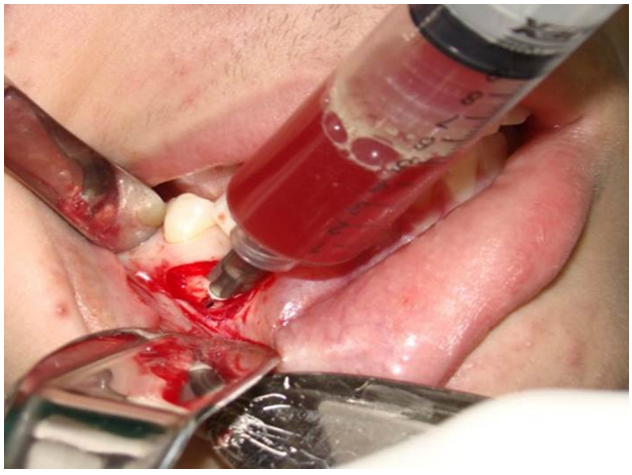

Fig. 3 – Aspiration of the contents of the cavity showing bloody and brilliant appearance.

Discussion

The traumatic bone cyst is a non-neoplastic lesion, characterized by an empty bone cavity or containing liquid, presenting the radiographic characteristics a unilocular radiolucent area with clipping effect, the teeth involved are vital and do not show root resorption.[4,9] In the present case, it was observed that the characteristics cited in the literature,[3,4,9,11] confirmed the diagnosis of traumatic bone cyst. It was not a histopathological analysis which could confuse us with aneurysmal bone cyst; the final diagnosis was confirmed after regression of the lesion with follow-up without recurrence of two years, differing of the aneurismal bone cyst, that will have a recurrence rate of 6–60% of cases in eight months and this is due to incomplete resection of lesion.[10]

The pathogenesis of traumatic bone cyst remains a matter of controversy and several theories have been suggested. Trauma is the most common etiological factor discussed. It leads to intraosseous hematoma formation, in which the blood clot liquefies, leading to osteoclastic bone resorption caused by enzyme activity.[15] In this case, the absence

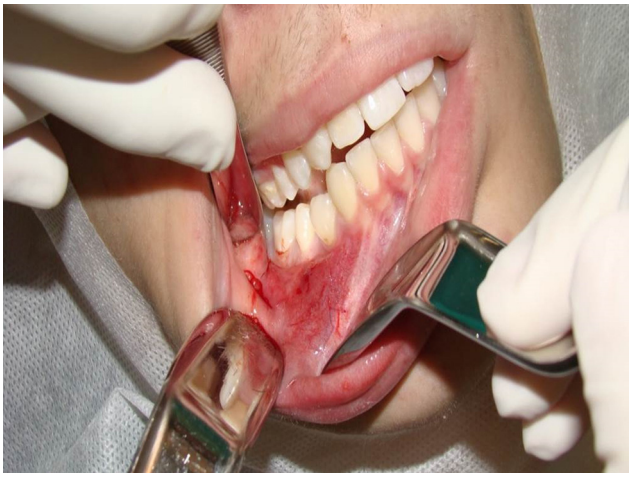

Fig. 2 – Clinical intraoral aspect showing a normal mucosa.

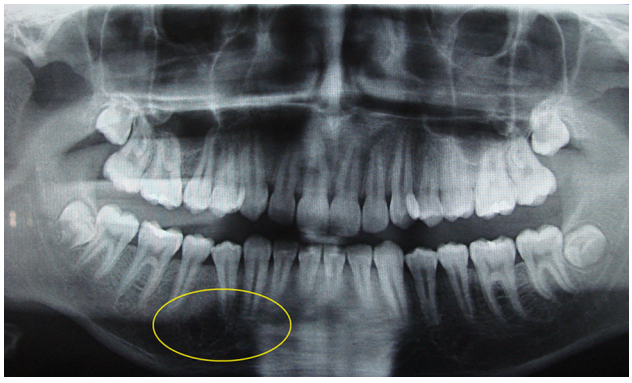

Fig. 4 – Panoramic radiograph showing bone repair at the lesion site follow-up 24 months.

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of traumatic events in the maxillofacial area and the history of CMT showed a probable etiology associated to local disorder in cranio-maxillofacial growth and development. Unilateral muscle shortening caused by CMT can lead to sleep-wake postural changes, contributing to musculoskeletal growth and development changes. Such changes may cause unbalanced pressure in the skull and facial bones in development, and can consequently lead to remodeling in the facial bones and facial hemi hypoplasia or plagiocephaly.[12–15]

The diagnosis of the traumatic bone cyst is made accidentally in a routine radiographic examination as a unilocular radiolucent area with "clipping effect", the teeth involved are vital and do not shows root resorption.[4] Definitive diagnosis is invariably made in exploratory surgery, when an empty bone cavity without epithelial lining is observed. During curettage of the cavity walls, normal bone tissue and occasionally fibrous tissue can be observed, as in the case presented. In some cases, a straw-colored liquid or bright blood is observed.[9,10] The traumatic bone cyst can make differential diagnosis with aneurysmal bone cyst because both lesions are preferentially affecting the long bones and, when that affects the maxillofacial region, which may preferably be present in the posterior mandible. Both the aneurismal bone cyst and traumatic bone cyst prevail in the second decade of life, but with regards to gender, traumatic bone cyst has a predilection for males[4–7] and aneurysmal bone cysts to female.[10] A simple classification of aneurysmal bone cysts was introduced in three stages according to radiological and clinical aspects by Capanna et al.[10] The inactive stage presents with complete periosteal and sclerotic borders. The active stage shows incomplete periosteal boarders with defined margins. The third, aggressive, stage is described as a uniform osteolysis with diffuse boarders of the lesion. Active and aggressive cysts tend to recur, whereas inactive cysts do not show any proliferation.[10]

Congenital torticollis is characterized by shortening and fibrosis of the sternocleidomastoid muscle detected at birth or shortly after birth. It is the third most common congenital muscle skeletal anomaly.[12–16] Muscular torticollis can be subdivided into three groups: Group 1 is the sternocleidomastoid tumor group, which consists of torticollis with a palpable tumor, that is, fibromatosis colli. This is a hard, movable mass within the substance of the sternocleidomastoid muscle detected at birth. This mass may be tender to palpation and usually regresses within the first year of life. This is the most common presentation. Group 2, known as muscular torticollis, consists of torticollis with tightness of the sternocleidomastoid muscle, but no palpable tumor. The last group, Group 3 (also known as POST), is a postural torticollis without a mass or tightness of the sternocleidomastoid muscle.[15,16]

The ideal treatment of congenital torticollis is controversial. Treatment modalities include observation, manual stretching, braces, physiotherapy, botulinum toxin, and different surgical procedures.[14]

As traumatic bone cyst, muscular torticollis also has its etiology associated with trauma.[3,15] The association of both pathologies could be justified not by a direct traumatic event, but by growth and developmental changes that can stimulate or locally press the mandible,[12] resulting in the formation of a traumatic bone cyst.

Although there is no correlation in literature between diseases, the present case study, based on history, could allow the hypothesis of etiological association. Moreover, a new front is opened for discussion and monitoring of patients suffering from the same diseases and it may contribute to the improvement of methods of monitoring, diagnosis and treatment.

REFERENCES

1. Lucas C, Blum T. Do all cysts of the jaws originate from the dental system. J Am Dent Assoc. 1929;16:659–61.
2. Saia G1, Fusetti S, Emanuelli E, Ferronato G, Procopio O. Intraoral endoscopic enucleation of a solitary bone cyst of the mandibular condyle. Int J Oral Maxillofac Surg. 2012 Mar;41:317–20.
3. Harnet JC, Lombardi T, Klewansky P, Rieger J, Tempe MH, Clavert JM. Solitary bone cyst of the jaws: a review of the etiopathogenic hypotheses. J Oral Maxillofac Surg. 2008 Nov;66:2345–8.
4. Lago CA, Cauás M, Pereira MA, Portela L. Cisto ósseo traumático em mandíbula: relato de caso. Rev Cir Traumatol Buco-Maxilo-Fac. 2006;6:23–8.
5. Saito Y, Hoshina Y, Nagamine T, Nakajima T, Suzuki M, Hayashi T. Simple bone cyst: a clinical and histopathologic study of fifteen cases. Oral Surg Oral Med Oral Pathol. 1992;74:487–91.
6. Kuttenberger J, Farmand M, Stoss H. Recurrence of a solitary bone cyst of the mandibular condyle in a bone graft. Oral Surg Oral Med Oral Pathol. 1992;74:550–6.
7. Motta AFJ, Torres SR, Coutinho ACA. Traumatic bone cyst: report of a case diagnosed after orthodontic treatment. Rev Odonto Cienc. 2007;22:377–81.
8. Xanthinaki AA, Choupis KI, Konstantinos T, Pagkalos VA, Papanikolaou SI. Traumatic bone cyst of the mandible of possible iatrogenic origin: a case report and brief review of the literature. Head Face Med. 2006;2:40.
9. Neuschl M, Reinert S, Gülicher D. 3rd, Neuschl J, Hoffmann J. Aneurysmal bone cyst of the ascending ramus mandible. A case report. J Craniomaxillofac Surg. 2014;42:e36–8.
10. Campidelli C, Di Tommaso L, Zanetti G. Aneurysmal bone cysts of the nasal cavity. Description of a case and review of the literature. Pathologica. 2003;95:103–7.
11. Martins-Filho PR, Santos TdeS, Araujo VL, et al. Traumático osso cisto da mandíbula: uma revisão de 26 casos. Braz J Otorhinolaryngol (Brasil). 2012;78:16–21.
12. Keller EE, Jackson IT, Marsh WR, Triplett WW. Mandibular asymmetry associated with congenital muscular torticollis. Oral Surg Oral Med Oral Pathol. 1986;61:216–20.
13. Lopes I, Alves A, Cunha A, Grande CC, Barroso J. Torcicolo Muscular Congénito: A Propósito de Um Caso Clínico. Arq Med [periódico na Internet]. 2009;23:7–9.
14. Pombo Castro M, Luaces Rey R, Vázquez Mahía I, López-Cedrún Cembranos JL. Congenital muscular torticollis in adult patients: literature review and a case report using a harmonic scalpel. J Oral Maxillofac Surg. 2014;72:396–401.
15. Cheng JC, Tang SP, Chen TM, et al. The clinical presentation and outcome of treatment of congenital muscular torticollis in infants—a study of 1086 cases. J Pediatr Surg. 2000;35:1091–6.

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16. Do TT. Congenital muscular torticollis: current concepts and review of treatment. Curr Opin Pediatr. 2006;18:26–9.

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