Synovial Chondromatosis of the Temporomandibular Joint Successfully Treated by Surgery

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Received: 28 June 2014 / Accepted: 1 April 2015 / Published online: 26 April 2015
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Abstract Synovial chondromatosis (SC) is a chronic process, defined as a reactive cartilaginous proliferation, characterized by formation of cartilaginous nodules, usually loose in the joint space of the synovial membrane. It mainly affects large joints such as knee, hip, shoulder, and elbow, commonly in male patients. However, its manifestation in the Temporomandibular joint (TMJ), is a rare finding, occurring predominantly in females. This paper reports a case of a woman who presented to the service of Stomatology complaining of mouth opening limitations and pain in her left pre-auricular region. After clinical and radiographic analyses, the condition was diagnosed as SC of the TMJ. The loose bodies within the TMJ were removed under general anesthesia. Histological and follow-up features of this lesion are also discussed. To our knowledge, this is the second report of SC of the TMJ in Brazil.

Keywords Synovial chondromatosis · Temporomandibular joint · Metaplasia

Introduction

Synovial chondromatosis (SC) is a rare condition characterized by the presence of loose cartilaginous bodies, due to the abnormal proliferation of synovial membrane. These bodies can be attached or loose in the articular space. There are several reports of this pathology affecting joints of long bones. In these regions, male patients are most affected. Primary occurrence in the temporomandibular joint (TMJ) is rare in a proportion of female/male 1.5:1 [1–4]. Most of the cases are unilateral, at the right side with just a few cases of bilateral occurrence [1, 4–10].

Regarding the age, the lesion ranges from 39 to 55.4 years old and the symptoms are not pathognomonic. Symptoms could be similar to that of other TMJ disorders, such as swelling on pre-auricular region, pain and limitation or deviation of mandibular movements [3, 8, 11–13]. Patients can also present clicking and crepitus of the joint. Moreover, luxation of the mandible, displacement of the mandible to the opposite side, inability to close the jaw properly, vertigo, and tinnitus can also present in patients [3].

According to Guarda-Nardini et al. [5] the first report of SC in TMJ dates from 1933 with a case of a cartilaginous metaplastic process originating in synovial membrane.

SC etiology remains unclear and some authors mention trauma, joint disorder, infection, and inflammatory processes of the TMJ as possible causes of the condition [2, 9, 10, 14]. The more prevalent clinical signs presented by the patients are pain, swelling at the affected TMJ, crepitation, and limitation of jaw movements [1, 10]. Tomography, magnetic resonance imaging (MRI), and orthopantomography are used as diagnostic tools [5].

The treatment modality should be based on symptoms reported by the patient, structures involved, radiographic findings, and the evolution of the lesion. Removal of the

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cartilaginous bodies by open surgery with synovectomy [1] is the most recommended approach. Arthroscopy may also be used [10]. We report a case of a SC treated successfully through a surgical approach.

Case Report

A 54-year-old female patient presented at the Odilon Behrens Hospital (OBH), complaining of pain and swelling in her left TMJ, and difficulty of mouth opening. She was attended in our service carrying a Magnetic Resonance Imaging (MRI) exam, with a nonspecific diagnosis of “TMJ pathology.” The patient reported previous medical treatment, complaining of an intermittent and spontaneous pain at her left TMJ, with 1 year of evolution, which increased in the last month without any history of trauma. She also reported the presence of bruxism. In that occasion, she was prescribed analgesic and muscle relaxant drugs. However, the symptoms did not present any improvement.

After an extra oral examination, a pre-auricular swelling on left side was observed, followed by spontaneous pain, limited mouth opening (maximum of 20 mm), and mandibular deviation to the left side. Crepitation and exacerbation of the pain on palpation were additional findings. For additional diagnostic information, computed tomography (CT) was requested and revealed the presence of dense and multiple nodules, with some areas of calcification (Fig. 1a), anterior to left condyle, measuring approximately 35 mm (Fig. 1b) with no signs of temporomandibular dislocation. The TMJ bony structure was preserved.

Based on clinical and imaging findings, a presumptive diagnosis of SC of the left TMJ was proposed.

Under general anesthesia, an L-inverted incision was performed to access the TMJ capsule exposing the articular

compartments. After performing the capsule incision, an extravasation of a light-colored and viscous liquid could be observed in a higher amount than the normal physiological of the TMJ content. Cartilaginous bodies were identified at the upper compartment (before the mandibular condyle) and they were completely removed (Fig. 2a). In a gross view, 11 irregular bodies of various shapes and sizes (the bigger measuring approximately 10 mm), could be identified. They were whitish and with consistency similar to cartilage (Fig. 2b). The surgical incision was closed in layers. There were no complications in the postoperative period. Tissue specimens were fixed in 10 % buffered formalin and sent for histopathological examination.

Hematoxylin and eosin staining disclosed a lesion characterized by slightly basophilic and irregular tissue interspersed with eosinophilic areas, showing scattered and hyperchromatic cells. The vascularity is scarce and no inflammatory cells could be visualized (Fig. 3a). Areas resembling hyaline cartilaginous metaplasia with multiple lacunae of different sizes, some of them filled by chondrocytes of variable shape, were visualized. Basophilic areas resembling calcified tissue were also observed (Fig. 3b). Therefore, histopathological examination was in accordance with the clinical diagnosis of SC.

The patient still remains in clinical and radiographic follow-up. Three years after the surgery, the patient showed reestablishment of TMJ function, mouth opening without pain or limitation (>35 mm), and no evidence of recurrence.

Discussion

Synovial Chondromatosis is a reactive process characterized by the formation of loose cartilaginous bodies, produced by abnormal proliferation of synovial membrane [5].

Fig. 1 Computed tomography (CT) imaging. **a** Axial view showing a hyperdense image located between the zygomatic arch and the condylar process of the mandible on the *left side*. **b** Coronal view

showing a hyperdense image between the condyle and skull base on the *left side*. Arrows showing the image corresponding to the cartilaginous lesion

Fig. 2 Surgical view. **a** Open joint surgery removing the mass lesion confined to a single joint compartment. **b** Macroscopic cartilaginous aspect of multiple irregular bodies

Fig. 3 Histologic features. **a** Lesion characterized by slightly basophilic and irregular tissue interspersed with eosinophilic areas, showing scattered and hyperchromatic cells. The vascularity is scarce and no inflammatory cells could be visualized. **b** Areas resembling hyaline cartilaginous metaplasia with multiple lacunae of different sizes, some of them filled by chondrocytes of variable shape, were visualized (black arrows). Basophilic tissue consistent with slight areas of calcification (white arrows) (hematoxylin-eosin $\times 400$ magnification)

A literature review of 242 cases showed that SC of TMJ affects mostly women, in a ratio of 1.5:1, mainly between the 4th and 5th decade of life. Generally, SC is unilateral

with higher prevalence at the right side [4, 8–10], but a few bilateral cases are reported. [1, 5–7, 15]. The upper compartment of the TMJ is more affected [5]. Our patient presented with the bodies restricted to the upper compartment of her left TMJ.

Although some publications present hypotheses about the etiology of SC, its pathogenesis is still a matter of debate [8, 18]. One work argues that SC is more a reactive metaplastic process than a neoplastic process [16]. Occlusal disorders have been appointed to trigger joint changes, which induce reactive processes [17]. This hypothesis can be considered for the present case, due to her bruxism, maybe producing functional overload to the TMJ. However, it is necessary to take into account the rarity of SC in this site, while bruxism is common in the population. Thus, some other aspects should be involved in the etiology of the SC.

The expression of growth factors is mentioned in the literature, such as the expression of FGFR-1 receptors by chondrocytes of SC, and the association of FGF-2 that seems to promote the lesion growth [18]. The presence of bone morphogenetic proteins (BMPs) in cartilaginous nodules suggests that BMPs synthesized by the synovia and by the loose bodies are able to promote bone and cartilaginous metaplasia, contributing to the pathogenesis of SC [19]. This information clearly warrants more research.

Clinical findings often overlap with the symptoms presented by patients with joint disorders and/or parotid tumors [20]. Pain, pre-auricular swelling, limitation of mandibular movement, and noise are the most commonly reported symptoms [5, 9, 14, 15]. Other symptoms such as limitation of mouth opening, deviation to the affected side, occlusal changes, sounds, and closed lock are also found [21], and were observed in our patient. Although less prevalent, complaints of headaches, vertigo, otalgia, and obstruction of the external auditory canal [22]; sclerosis of

the articular fossa and eminence; resorption of the articular surface of the temporal bone, and thinning of the glenoid fossa and erosion of the temporal bone have also been reported [11, 23]. In the present case these signs and symptoms were not observed.

Nevertheless the condition arises in the intra-articular spaces, and in some cases the SC may extend into the condyle [24], out of the TMJ capsule [11, 25] or even invade the middle cranial fossa [11, 26], being locally destructive or present a risk of malignancy [24].

The clinical findings, mainly in association with intermittent symptomatology and slow growth rate of our patient condition suggests a benign process. The use of auxiliary image exams (radiographs, CT, MRI, and arthroscopy) must be highlighted for differentiating from other conditions like osteoarthritis, osteochondroma, villonodular synovitis, and focal osteochondritis [6].

Radiographic findings are limited and are present only in 45 % of cases [27]. These data include increased joint space, presence of radiopaque structures, and irregular or sclerotic surfaces of the joint and bony changes. Panoramic X-ray (orthopantomograph) is a good option for this assessment, providing good visualization of the surrounding structures with a lower radiation dose, compared with CT. However, this exam presents a limitation in the tridimensional location of the lesion [10, 23]. CT enables the better definition of the size, shape, and location of the cartilaginous bodies and defines and displays the lesion [23]. As the MRI carried by the patient at her first appointment pointed out only a generic “TMJ pathology”, we opted to request CT images. They were essential to establish the tridimensional position of the lesion, guiding the surgical approach and the removal of the loose bodies. However, we must take into account that the MRI is absent of ionizing radiation [1, 10], and can be enough for the assessment of cartilage degeneration [2], determination of the borders of the lesion and of surrounding structures, and the distension of the joint capsule [14]. Nevertheless, bony structures cannot be visualized clearly. The evolution of diagnostic image tools, particularly CT and MRI, will probably be helpful for the increase in the number of diagnoses of SC in the near future.

Another method rarely used for diagnosis, but also included among the forms of treatment, is arthroscopy [15]. It allows the visual inspection of intra-articular compartments and of the cartilaginous bodies [3, 12, 14, 28].

SC treatment depends on the size of the nodules, structures involved, and the extent of the lesion. Although there are reports of clinical and imaging follow-up (when there is no interference in jaw functions) [29], the first choice of treatment, by most authors, is the removal of the cartilaginous bodies, through arthroscopic or open surgery, combined with synovectomy [2, 5, 9, 11, 14, 15, 26].

Surgical treatment was the method of choice in the presence of symptomatology and calcified lesions with considerable dimensions. When the lesion involves other joint structures, it should be treated more aggressively.

Besides clinical and imaginological features, the histopathological examination is a very important tool for the final diagnosis. Histological sections shows nodular hyaline fibrocartilaginous fragments with variable cellularity of clustered chondrocytes. Focal atypical chondrocytes exhibiting nuclear pleomorphism, bizarre and binucleated cells, and myxoid change of the matrix are commonly seen only as a focal phenomenon [30, 31]. Areas of calcification and ossification may also be present. Malignant transformation of SC to a chondrosarcoma is rare and its progression has been recently reported by some authors [30, 32, 33]. The follow-up period of SC is variable. However, periods of follow-up ranging from 1 month to 18 years are reported [5].

Although rare, recurrences can occur [15, 34, 35]. Thus, regardless of treatment modality, it is imperative to proceed to complete removal of the cartilaginous nodules and synovia [4, 36]. The assessment of synovia as well as its removal seems to be associated with the prevention of SC recurrence [34, 35].

Conclusion

Indeed, SC is a rare pathological entity, especially in TMJ. To our knowledge this is the second case reported in Brazil. Thus, as clinical and imaging findings of SC may be inaccurate or very similar to other joint diseases, it is important to search for a diagnosis method that provides the best view of the structures involved in the lesion. In all cases, histological examination is very important to the establishment of the final diagnosis. Its association with the clinical and imaginological findings allows for a better and less empiric professional approach.

References

1. Guijarro-Martínez R, Torres MP, Mateo MM, García IS, Alba LM, Gimilio MEI, et al. Bilateral synovial chondromatosis of the temporomandibular joint. *J Cranio Maxillo Facial Surg.* 2011;39:261–5.
2. Abuabara A, Miles DA, Cruz GV, Passeri LA. Radiological study of three cases of loose bodies in the temporomandibular joint. *Braz J Oral Sci.* 2010;9(2):133–6.
3. Moses JJ, Hosaka H. Arthroscopic punch for definitive diagnosis of synovial chondromatosis of the temporomandibular joint: case report and pathology review. *Oral Surg Oral Med Oral Pathol.* 1993;75:12–7.
4. Bonatti BDS, Patrocínio LG, Costa SAA, Costa JMC, Patrocínio JA. Condromatose sinovial de articulação têmporo-mandibular. *Rev Bras Otorrinolaringol.* 2008;74(3):480.

5. Guarda-Nardini L, Piccotti F, Ferronato G, Manfredini D. Synovial chondromatosis of the temporomandibular joint: a case description with systematic literature review. *Int J Oral Maxillofac Surg*. 2010;39:745–55.
6. Balasundaram A, Geist JR, Gordon SC, Klasser GD. Radiographic diagnosis of synovial chondromatosis of the temporomandibular joint: a case report. *JCDA*. 2009;75(10):711–4.
7. Keogh CF, Torreggiani WC, Munk PL. Bilateral synovial chondromatosis of the temporomandibular joint. *Clin Radiol*. 2002;57:862.
8. Pau M, BiSCák Á, Reinbacher KE, Feichtinger M, Kärcher H. Surgical treatment of synovial chondromatosis of the temporomandibular joint with erosion of the skull base: a case report. *Int J Oral Maxillofac Surg*. 2014;43:600–5.
9. Von Lindern JJ, Theuerkauf I, Niederhagen B, Bergé S, et al. Synovial chondromatosis of the temporomandibular joint: clinical, diagnostic, and histomorphologic findings. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2002;94:31–8.
10. Meng J, Guo C, Yi B, Zhao Y, et al. Clinical and radiologic findings of synovial Chondromatosis affecting the temporomandibular joint. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2010;109:441–8.
11. Neville BW, Damm DD, Allen CM, Bouquot JE. *Patologia oral and maxillofacial*. 2nd ed. Rio de Janeiro: Guanabara Koogan; 2004.
12. Cai XY, Yang C, Chen MJ, Jiang B, et al. Arthroscopically guided removal of large solitary synovial chondromatosis from the temporomandibular joint. *Int J Oral Maxillofac Surg*. 2010;39:1236–9.
13. Adachi PLG, Kaba SP, Martins MT, Hueb CH, Shinohara EH. Arthrocentesis in the treatment of loose bodies of the temporomandibular joint associated with synovial Chondromatosis. *Br J Oral Maxillofac Surg*. 2008;46:320–1.
14. Holmlund AB, Eriksson L, Reinholdt FP. Synovial chondromatosis of the temporomandibular joint. Clinical, surgical and histological aspects. *Int J Oral Maxillofac Surg*. 2003;32:143–7.
15. Shah SB, Ramanojam S, Gadre PK, Gadre KS. Synovial chondromatosis of temporomandibular joint: journey through 25 decades and a case report. *J Oral Maxillofac Surg*. 2011;69(11):2795–814.
16. Milgram JW. The classification of loose bodies in human joints. *Clin Orthop*. 1977;124:282–91.
17. Xiang S, Rebellato J, Inwards CY, Keller EE. Malocclusion associated with osteocartilaginous loose bodies of the temporomandibular joint. *JADA*. 2005;36:484–9.
18. Sato J, Segami N, Suzuki T, Yoshitake Y, Nishikawa K. The expression of fibroblast growth factor-2 and fibroblast growth factor receptor-1 in chondrocytes in synovial chondromatosis of the temporomandibular joint. Report of two cases. *Int J Oral Maxillofac Surg*. 2002;31:532–6.
19. Nakanishi S, Sskamoto K, Yoshitake H, Kino K, et al. Bone morphogenetic proteins are involved in the pathobiology of synovial chondromatosis. *Biochem Biophys Res Commun*. 2009;379:914–9.
20. Cai XY, Yang C, Chen MJ, Yun B. Simultaneous pigmented villonodular synovitis and synovial chondromatosis of the temporomandibular joint: case report. *Int J Oral Maxillofac Surg*. 2009;38:1201–25.
21. Petito AR, Bennett J, Assael LA, Carlotti AE Jr. Synovial chondromatosis of the temporomandibular joint: varying presentation in 4 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2000;90:758–64.
22. Thompson K, Schwartz HC, Miles JW. Synovial chondromatosis of the temporomandibular joint presenting as a parotid mass: possibility of confusion with benign mixed tumor. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 1986;62:377–80.
23. Yu Q, Yang J, Wang P, Shi H, Luo J. CT features of synovial chondromatosis in the temporomandibular joint. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2004;97:524–8.
24. Nussenbaum B, Roland PS, Gilcrease MZ, Odell DS. Extra-articular synovial chondromatosis of the temporomandibular joint pitfalls in diagnosis. *Arch Otolaryngol Head Neck Surg*. 1999;125:1394–7.
25. Huh JK, Park JY, Lee S, Lee SH, et al. Synovial chondromatosis of the temporomandibular joint with condylar extension. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2006;101:83–8.
26. Xu WH, Ma XC, Guo CB, Yi B, Bao SD. Synovial Chondromatosis of the temporomandibular joint with middle cranial fossa extension. *Int J Oral Maxillofac Surg*. 2007;36:652–5.
27. Koyama J, Ito J, Hayashi T, Kobayashi F. Synovial Chondromatosis in the temporomandibular joint complicated by displacement and calcification of the articular disk: report of two cases. *Am J Neuroradiol*. 2001;22:1203–6.
28. Sembronio S, Albiero AM, Toro C, Robiony M, et al. Arthroscopy with open surgery for treatment of synovial chondromatosis of the temporomandibular joint. *Br J Oral Maxillofac Surg*. 2008;46:582–4.
29. Mori Y, Kakudo K, Gotoh M, Kubo H, et al. A case of synovial chondromatosis of the temporomandibular joint followed for 17 years. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2011;112:e35–9.
30. Coleman H, Chandraratnam E, Morgan G, Gomes L, Bonar F. Synovial chondrosarcoma arising in synovial chondromatosis of the temporomandibular joint. *Head Neck Pathol*. 2013;7(3):304–9.
31. Sink J, Bell B, Mesa H. Synovial chondromatosis of the temporomandibular joint: clinical, cytologic, histologic, radiologic, therapeutic aspects, and differential diagnosis of an uncommon lesion. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2014;117(3):e269–74.
32. Bertoni F, Unni KK, Beabout JW, et al. Chondrosarcomas of the synovium. *Cancer*. 1991;67:155–62.
33. Merrill RG, Yih W-Y, Shamloo J. Synovial chondrosarcoma of the temporomandibular joint: a case report. *J Oral Maxillofac Surg*. 1997;55:1312–6.
34. Lucas JH, Quinn P, Foote J, Baker S, Bruno J. Recurrent synovial chondromatosis treated with meniscectomy and synovectomy. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 1997;84:253–8.
35. D'Souza B, Dimitroulis G. A case of recurrence of synovial chondromatosis of the temporomandibular joint. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2007;104:59–61.
36. Sato J, Notani KI, Goto J, Shindoh M, Kitagawa Y. Synovial chondromatosis of the temporomandibular joint accompanied by loose bodies in both the superior and inferior joint compartments: case report. *Int J Oral Maxillofac Surg*. 2010;39:86–8.