CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 20 (2016) 133–137

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Synovial chondromatosis of the right side temporomandibular joint extending to the middle cranial fossa: A case report with 7-year postoperative follow up and expression of a biomarker of cell proliferative activity

Hiroyuki Yoshitake[a,*], Kou Kayamori[b], So Wake[a], Fumiaki Sato[a], Koji Kino[c], Kiyoshi Harada[a]

[a] Department of Maxillofacial Surgery, Graduate School, Tokyo Medical and Dental University, Tokyo, Japan
[b] Department of Oral Pathology, Graduate School, Tokyo Medical and Dental University, Japan
[c] Temporomandibular Joint and Oral Function (Temporomandibular Joint Clinic) Graduate School, Tokyo Medical and Dental University, Japan

ARTICLE INFO

Article history:
Received 12 December 2015
Received in revised form 18 January 2016
Accepted 18 January 2016
Available online 25 January 2016

Keywords:
Temporomandibular joint
Synovial chondromatosis
Middle cranial fossa
Cranial dura mater
Cell proliferative activity
Biomarker

ABSTRACT

INTRODUCTION: Synovial chondromatosis of the temporomandibular joint (TMJ) with cranial extension is rare. Here, we report 7-year follow-up of a case with immunohistochemical examination of cell proliferative activity.

PRESENTATION OF CASE: The patient was a 72-year-old man. Severe bone resorption of the glenoid fossa was apparent on CT images. Pathological findings by biopsy led to diagnosis of synovial chondromatosis of the right side TMJ. Extirpation of the tumor was performed via temporopreauricular incision under general anesthesia. PCNA expression was examined by immunohistochemical analysis.

The lesion had penetrated into the middle cranial fossa, but the cranial dura mater was intact. Expression of PCNA was confirmed.

DISCUSSION: The PCNA expression suggested that growth activity caused expansion of the lesion to the skull base.

CONCLUSION: We were able to follow up this case for a long period without recurrence postoperatively.

1. Introduction

Synovial chondromatosis commonly occurs in large joints such as knees, elbows or shoulders, but rarely in the temporomandibular joint (TMJ). Synovial chondromatosis is characterized by formation of cartilage nodules with or without calcification in the synovial membrane or floating free bodies in the synovial fluid of the joint space, and these nodules often grow in the joint space. The etiology and pathogenesis of the disease are unknown, but a correlation with traumatic injury in the joint space has been suggested [1–7]. Synovial chondromatosis extended to the middle cranial fossa is rare in cases in which the lesion occurs in the TMJ [8–10]. Previously, we have reported 6-month postoperative follow-up of a case of synovial chondromatosis of the right side TMJ extending to the middle cranial fossa [11]. In this report, we describe 7-year follow-up of this patient postoperatively.

2. Presentation of case

The patient was a 72-year-old man with complaints of limitation of mouth opening, pain of the right side TMJ and malocclusion of the molar teeth. He had been treated with an occlusal appliance and physical therapy under a diagnosis of temporomandibular disorder (TMD) at a local hospital for several years, but the symptoms did not improve. He was referred to our hospital in April 2005. His medical history included alcoholic hepatitis (LDH: 236 U/l, AST: 55 U/l, ALT: 68 U/l, r-GTP: 236 U/l).

A clinical examination revealed limitation of mouth opening (interincisal: 30 mm) and a shift of the midline of the mandible to the right side by about 4 mm compared with the facial midline. Diffuse swelling and tenderness were present around the right side preauricular region. The right mandibular condyle was displaced anteriorly and a severe bone defect from the glenoid fossa to middle cranial fossa was observed on simple X-ray and CT images (Fig. 1a,b). A large mass lesion near the cranial dura mater and bone resorption of the cranial base were found on MRI, without invasion into the brain. The signal intensity of the tumor mass was intermediate in PDWI and mixed heterogeneous high signal

* Corresponding author at: Maxillofacial Surgery, Tokyo Medical and Dental University, 1-5-45 Yushima, Bunkyo-ku, Tokyo 113-8510, Japan.
E-mail address: h-yoshitake.mfs@tmd.ac.jp (H. Yoshitake).

http://dx.doi.org/10.1016/j.ijscr.2016.01.026

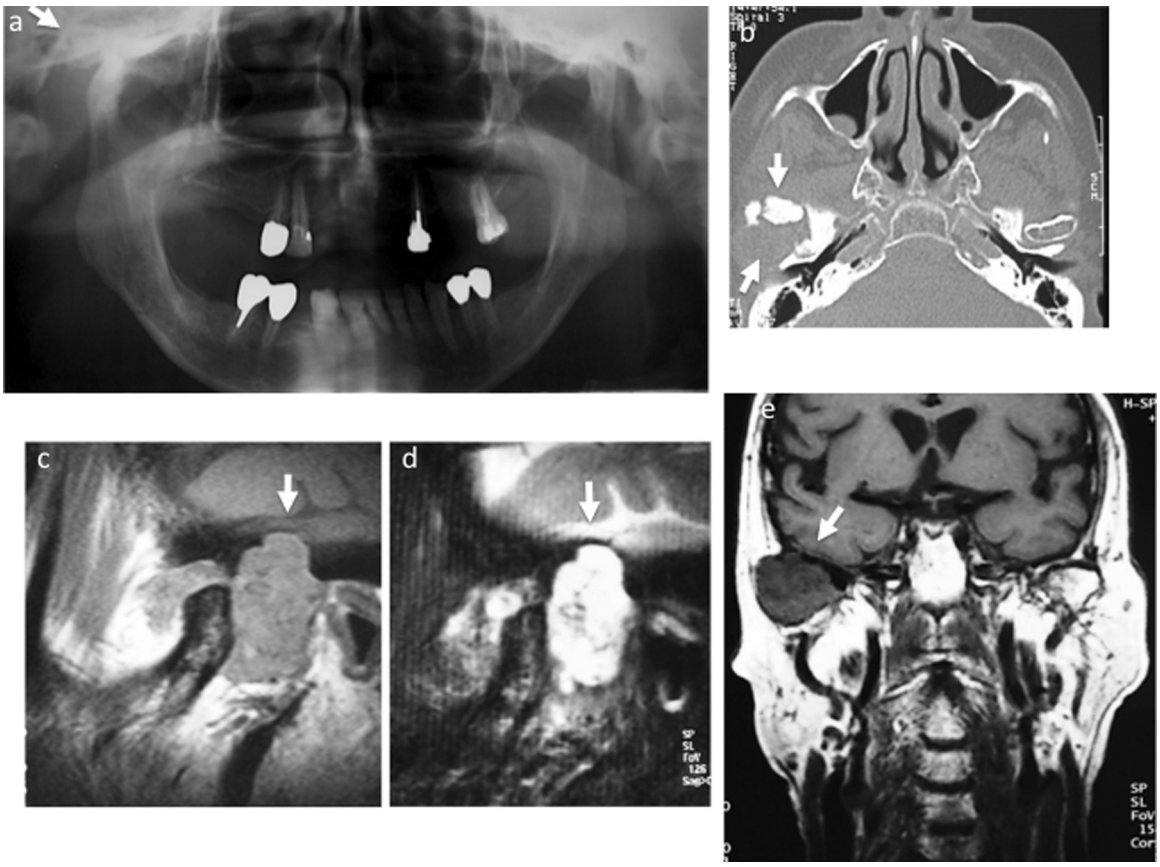

Fig. 1. (a,b) The right-side mandibular condyle was displaced anteriorly by a mass lesion and severe bone resorption from the glenoid fossa to middle cranial fossa was observed on simple X-ray (a) and CT (b). (c–e) A large mass lesion near the cranial dura mater was confirmed on MRI, but invasion into the brain was not observed (c,e) PdWI, (d) T2WI).

(a,c–e) Images reproduced from Ref. [11].

intensity and spotted low signal intensity regions were present in T2WI (Fig. 1c–e).

A biopsy under local anesthesia resulted in a pathological diagnosis of synovial chondromatosis of the right TMJ. Extirpation of the tumor of the right TMJ was performed through a temporo-preauricular incision under general anesthesia in September 2005 (Fig. 2a–c). The mass lesion was extirpated completely with the surrounding capsule. Penetration of the middle cranial fossa was observed, but the cranial dura mater was intact (Fig. 2b,c). The mass lesion seemed to be composed of cartilaginous tissues (Fig. 2d). Reconstruction of the bone defect was not performed. The malocclusion of the molar teeth was improved by correct reduction of the condylar position.

Pathological findings showed that the specimen consisted of nodules of cartilage and was surrounded by a capsule of synovial connective tissues, with mineralization of the cartilage (Fig. 2e,f). The clinical and histological findings were consistent with the diagnosis of synovial chondromatosis of the right TMJ made from the biopsy specimen. The specimen was also examined immnohistochemically. Immunostaining with an antibody against proliferating cell nuclear antigen (PCNA) was used to evaluate the growth activity of the lesion. PCNA-positive chondrocyte-like cells were found (Fig. 5a–c).

Malocclusion, limitation of mouth opening and TMJ pain improved after surgery and new bone formation of the glenoid fossa was observed on CT and MRI at 6 months postoperatively (Fig. 3a–d). Further irregular bone resorption was not observed on CT images at 7 years postoperatively and the bone defect of the glenoid fossa was decreased compared with CT images at 6 months postoperatively (Fig. 4a,b). MRI findings at 7 years postoperatively showed a mixed high and low signal intensity area behind the condyle on T2WI (Fig. 4c–e). Malocclusion with displacement of the mandibular condyle, and swelling and tenderness of the TMJ improved postoperatively, but unfortunately the patient died of pancreatic cancer in 2013.

3. Discussion

Synovial chondromatosis in the TMJ with cranial extension is a rare condition and the few reports of such cases have only had short-term postoperative follow-up [9,10]. In contrast, we were able to follow up our case for over 7 years after surgery.

Several metaplastic diseases can occur near the skull base, including acoustic nerve tumor and skull base meningioma. Commonly, the symptoms associated with these diseases include hearing disorder, tinnitus, vertigo, lightheadedness, blurred vision, and diplopia because many important cranial nerves are present near the regions of disease expansion or invasion. However, our case did not show symptoms associated with disorders of cranial nerves. In contrast, the symptoms were limitation of mouth opening, TMJ pain, and malocclusion. These are all TMJ-associated problems and this made it difficult to distinguish the condition from TMD. The size of the mass lesion was very large, but the lesion did not have a bad influence on the cranial nerves because synovial chondromatosis is limited to the joint capsule and is not invasive.

It is difficult to detect synovial chondromatosis in the TMJ on simple X-ray or CT images due to the limited calcification of the lesion. In previous reports, 37–42% of TMJ cases could not be con-

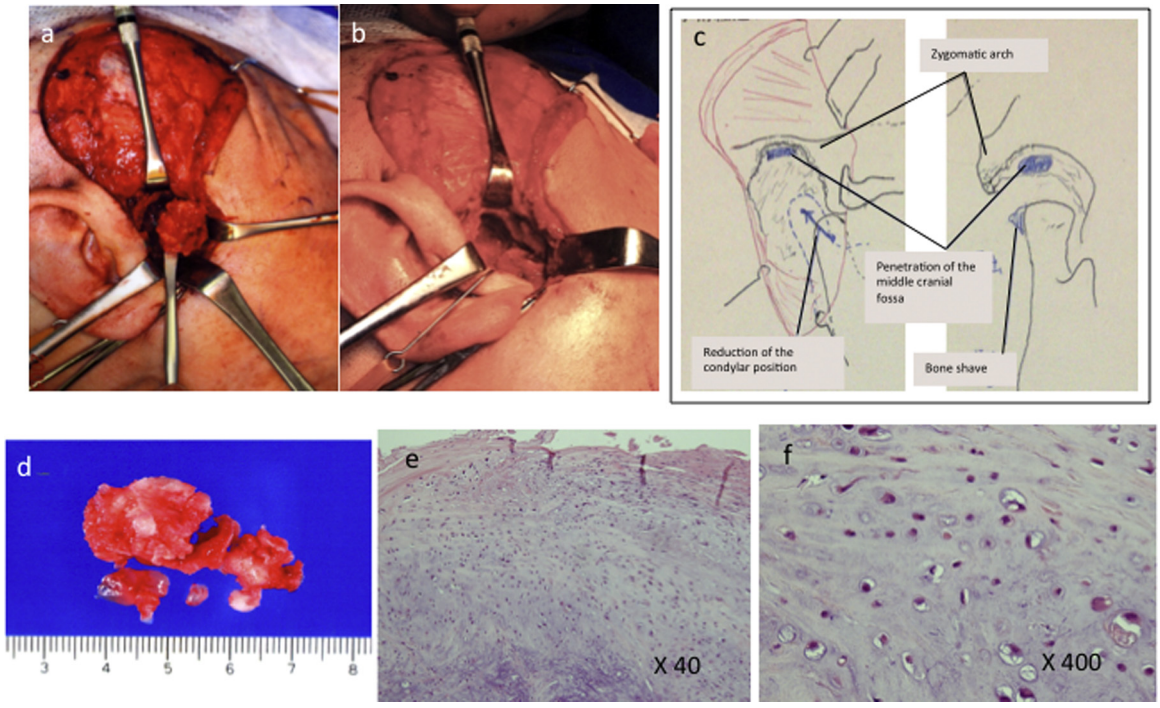

Fig. 2. (a,b) Intraoperative photographs. A bone defect of about 1 cm diameter in the gleinoid fossa was observed after extirpation. (c) Intraoperative findings. The illustrations are taken from the operative notes for the case. The annotations were translated into English by the author. (d–f) The specimen consisted of nodules of cartilage and was surrounded by a capsule of the synovial connective tissues. Mineralization of the cartilage was observed.
(a,d) Images reproduced from Ref. [11].

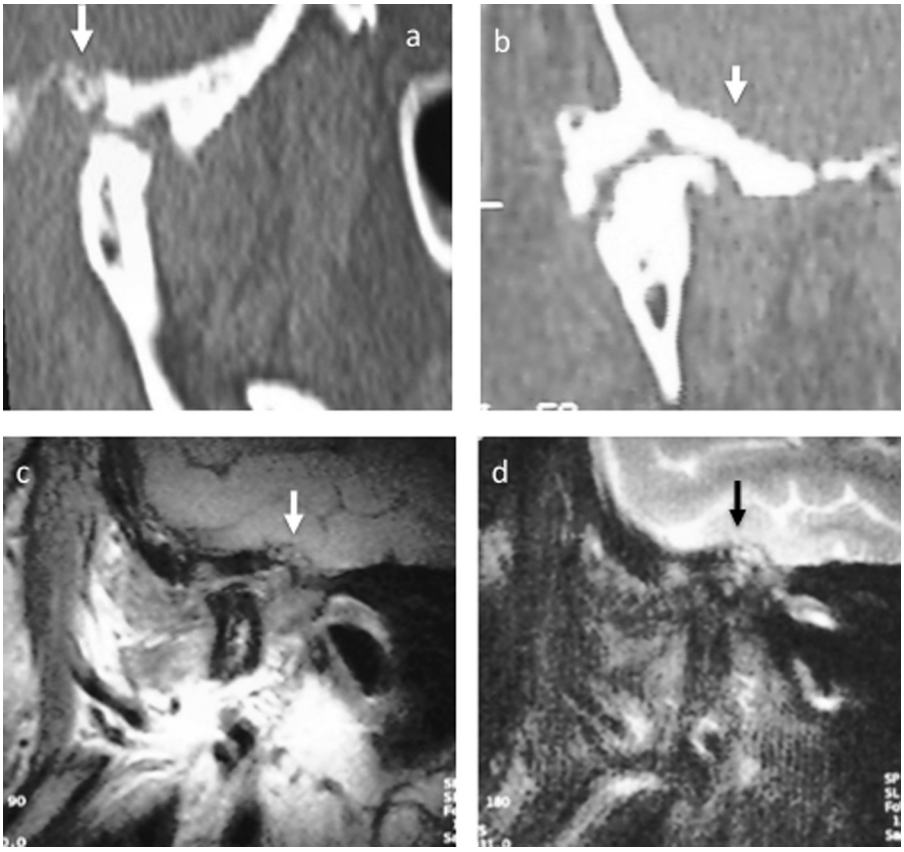

Fig. 3. (a–d) CT (a,b) and MRI (c,d) at 6 months after surgery. Arrows indicate new bone formation of the glenoid fossa.
(a,b) Images reproduced from Ref. [11].

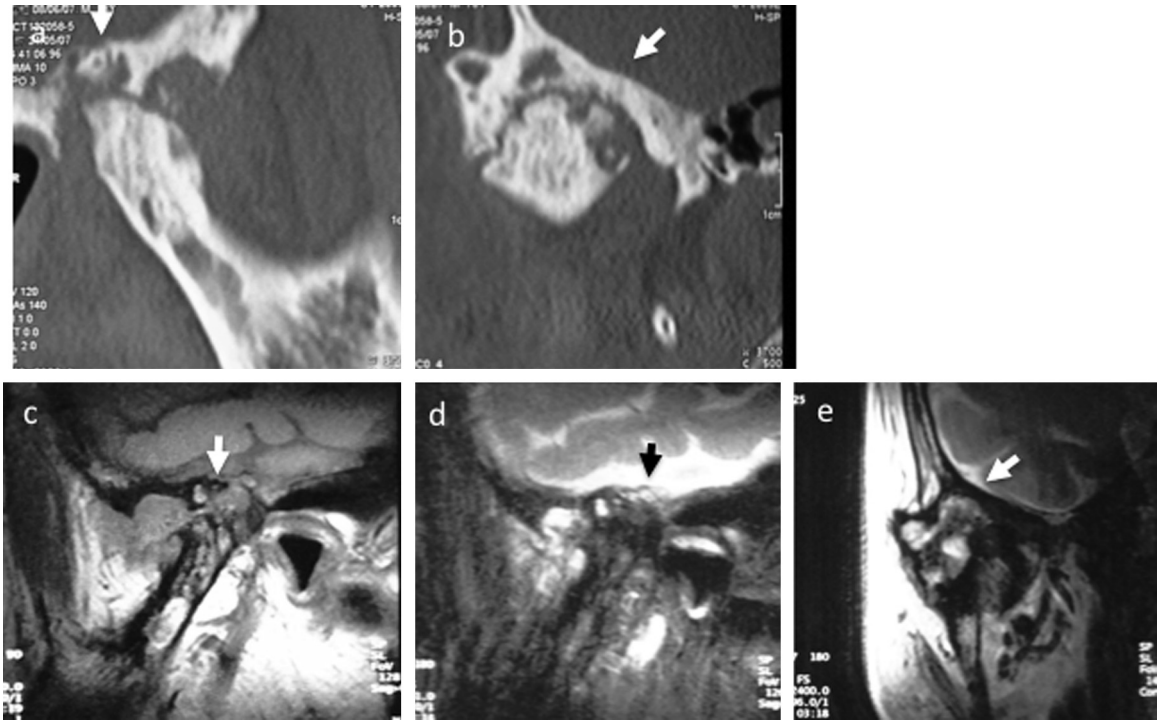

Fig. 4. (a,b) CT at 7 years after surgery (a,b). The bone defect of the glenoid fossa had decreased compared with CT at 6 months after surgery ((a,c) sagittal plane, (b,d) frontal plane). (c–e) MRI at 7 years after surgery ((a) sagittal plane on PdWI, (b) sagittal plane on T2WI, (c) frontal plane on T2WI). A mixed high and low signal intensity area was observed behind the condyle on T2WI (arrows).

firmed by simple X-ray [2–4]. MRI is useful for detection of this disease. The etiology and pathogenesis of synovial chondromatosis involving the TMJ is unclear, but there may be a correlation with traumatic injury of the synovial tissue in the joint space and most cases occur in the upper joint space. However, our patient did not have a history of a traumatic episode and the lesion extended to the skull base. CT scans performed just after MRI were also similar at 6 months, indicating no long-term bone degeneration (Fig. 2g,h).

In this case, there are two major points to consider with regard to penetration of synovial chondromatosis into the middle cranial fossa. The first is the time course. The patient was treated under a diagnosis of TMD for a long period. We hypothesize that synovial chondromatosis might have been present in the TMJ and that the lesion might even have been relatively large at that time. Examinations such as MRI performed at an earlier time may have permitted detection of the lesion around the TMJ, even if it was difficult to distinguish this disease from TMD. However, it actually took more than 7 years from the time the patient first noticed symptoms to make a correct diagnosis and provide appropriate treatment. During this period, the lesion had grown larger and penetrated into the middle cranial fossa.

The second important point is that the lesion had growth activity, but was not invasive. This may be a characteristic of synovial chondromatosis in the TMJ. In a previous report, little Ki-67 expression was detected in loose bodies of the syonovial chondromatosis [12]. PCNA is a biomarker of cell proliferative activity that is similar to Ki-67 [13]. In the current case, expression of PCNA indicated that the lesion maintained growth activity at the time of the operation, and was in stage 3 in the classification proposed by Gerard et al. [2]. This result shows the different character of the disease compared to typical TMJ-synovial chondromatosis [12,13]. This may be the most important reason why the lesion expanded to the skull base. However, the lesion was surrounded by a capsule of synovial connective tissue and was limited to the joint capsule, with invasion not observed. Therefore, the cranial dura mater was intact despite the severe bone resorption in the skull base. These conditions made complete tumor extirpation possible and led to a good outcome, despite the growth activity.

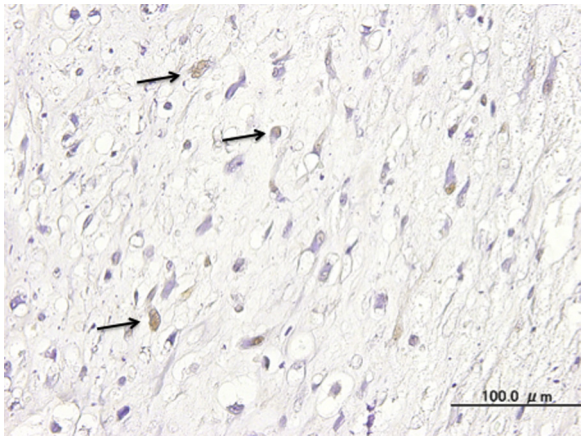

Fig. 5. Immnohistochemical detection of PCNA in chondrocytes in the resected specimen. Arrows indicate typical PCNA-positive cells. Original magnifications, ×200.

4. Conclusion

We have described a rare case of synovial chondromatosis in the TMJ that extended to the middle cranial fossa with a large bone defect produced by tumor mass expansion, despite the absence of progressive ossification or calcification. Immunohistochemical analysis indicated that the lesion maintained cell proliferative activity, but complete extirpation of the lesion led to a good outcome. We were able to follow-up the case for a relatively long period of over 7 years without recurrence.

Conflict of interest

None.

Funding

None.

Ethical approval

None.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Authors contribution

Hiroyuki Yoshitake, Fumiaki Sato, So Wake, Koji Kino and Kiyosi Hrada were the oral surgeons. Kou Kayamori was the oral pathologist for the study. Hiroyuki Yoshitake was responsible for writing the article and responsible for the manuscript preparation.

Guarantor

Hiroyuki Yoshitake.

References

[1] J.W. Milgram, Synovial osteochondromatosis: histopathological study of thirty cases, J. Bone Joint Surg. 59 (1977) 792–801.

[2] M.A. Aydin, A. Kurtay, S. Celebioglu, A case of synovial chondromatosis of the TMJ: treatment based on stage of the disease, J. Craniofac. Surg. 13 (2002) 670–675.

[3] Q. Yu, J. Yang, P. Wang, H. Shi, J. Luo, CT features of synovial chondromatosis in the temporomandibular joint, Oral Surg. Oral Med. Oral Pathol. Oral Radiol. Endod. 97 (2004) 524–528.

[4] M. Ida, H. Yoshitake, H. Yoshitake, K. Okoch, A. Tetsumura, N. Ohbayashi, et al., An investigation of magnetic resonance imaging features in 14 patients with synovial chondromatosis of the temporomandibular joint, Dentomaxillofac. Radiol. 37 (2008) 213–219.

[5] L.M. González-Pérez, J. Congregado-Córdoba, M.V. Salinas-Martín, Temporomandibular joint synovial chondromatosis with a traumatic etiology, Int. J. Oral Maxillofac. Surg. 40 (2011) 330–334.

[6] W. Zha, Y.F. Zhao, Y. Liu, L. Jiang, A case of synovial chondromatosis of the temporomandibular joint secondary to preauricular trauma, Int. J. Oral Maxillofac. Surg. 38 (2009) 1201–1225.

[7] E.L. Reinish, S.E. Feinberg, K. Devaney, Primary synovial chondromatosis of the temporomandibular joint with suspected traumatic etiology report of a case, Int. J. Oral Maxillofac. Surg. 76 (1997) 419–422.

[8] L. Guarda-Nardini, F. Piccotti, G. Ferronato, D. Manfredini, Synovial chondromatosis of the temporomandibular joint: a case description with systematic literature review, Int. J. Oral. Maxillofac. Surg. 39 (2010) 745–755.

[9] N. Yokota, C. Inenaga, T. Tokuyama, S. Nishizawa, K. Miura, H. Namba, Synovial chondromatosis of the temporomandibular joint with intracranial extension, Neurol. Med. Chir. (Tokyo) (48) (2008) 266–270.

[10] W.H. Xu, X.C. Ma, C.B. Guo, B. Yi, S.D. Bao, Synovial chondromatosis of the temporomandibular joint with middle cranial fossa extension, Int. J. Oral. Maxillofac. Surg. 36 (2007) 652–655.

[11] H. Yoshitake, F. Sato, K. Kino, Synovial shondromatosis of the temporomandibular joint extending to the middle cranial fossa: a case report, J. Jpn. Soc. TMJ 24 (2012) 17–21 (in Japanese).

[12] H. Yoshida, K. Tsuji, N. Oshiro, M. Wato, S. Morita, Preliminary report of Ki-67 reactivity in synovial chondromatosis of the temporomandibular joint: an immnohistochemical study, J. Craniomaxillofac. Surg. 41 (2013) 473–475.

[13] B. Mattews, R.M. Bernstein, B.R. Franza Jr, et al., Identify of the proliferating cell nuclear antigen and cyclin, Nature 309 (1984) 374–376.