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Journal	日本口腔検査学会雑誌, 8(1): 45-51
URL	http://hdl.handle.net/10130/3985
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Case report

Radiological and pathological examination of synovial chondromatosis of the temporomandibular joint: A case report

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Abstract

This report describes a rare case of synovial chondromatosis (SC) in the temporomandibular joint (TMJ) of a patient. SC is a non-neoplastic disease characterized by metaplasia of the connective tissue that leads to chondrogenesis in the synovial membrane. The clinical symptoms of SC affecting the TMJ are often characterized by swelling, pain, headache, crepitation, malocclusion and joint dysfunction. The major challenge concerning SC is to suspect and establish a correct diagnosis. Its nonspecific initial signs and symptoms may mimic other nonspecific TMJ diseases and can easily lead to a delay in diagnosis or a misdiagnosis. This report describes the presentation, diagnosis and surgical management of a SC case involving the right TMJ of a 39-year-old female.

Key words: synovial chondromatosis, temporomandibular joint disorders, diagnosis, Pathology

Received : 1st March 2015 Accepted : 6th April, 2015

Introduction

Synovial chondromatosis (SC) is a rare pathological condition characterized by the development of cartilaginous nodules within the connective synovial tissue of articulating joints. This benign condition is considered to be metaplastic rather than neoplastic. SC is most commonly found in larger joints, with a predilection for the knees, hips and elbows. SC of the knee joint was first reported by Pare in 1558, and

SC of the temporomandibular joint (TMJ) was first reported by Haller in 1764. The first histological description of SC was by Axhousen in 1933¹⁾. This condition is characterized by foci of metaplastic cartilage or osteocartilaginous nodules within the synovial membrane or lying free within the joint space.

Patients with SC will frequently present with symptoms not dissimilar to other pathologic

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conditions of the TMJ²⁾. Therefore, obtaining an accurate diagnosis of SC requires a thorough history-taking, clinical examination and appropriate radiographic study; however, the final diagnosis is confirmed histopathologically. The most common diagnostic modalities for SC include plain film radiography, magnetic resonance imaging (MRI) and computed tomography (CT). Proper selection of radiologic imaging for the diagnosis and study of SC has been previously published²⁾.

SC may be classified as primary or secondary, with primary SC having no known predisposing influence. Primary SC is also considered to be more aggressive in nature³⁾.

In 1977, Milgram⁴⁾ proposed another classification of SC with three Stages. During Stage 1, active metaplasia of the synovium results in the formation of small cartilaginous nodules that are confined entirely within the synovial lining. During Stage 2, some nodules are loose bodies, floating freely within the joint space while others are still attached to the synovium. During Stage 3, the nodules become ossified, and the previously metaplastic synovium becomes inactive. Observation also suggests that this system of staging is progressive, in that Stage 1 disease, if unchecked, will progress to Stage 2 and so on.

Case Report:

A 39 year-old Japanese woman was referred to the Department of Oral and Maxillofacial Surgery at the Tokyo Dental College Chiba Hospital, Japan, on July 17, 2014 with the chief complaint of a right preauricular swelling with TMJ pain and dysfunction.

Upon clinical examination, the patient was found to have a 3 x 3 cm area of right preauricular swelling. This resulted in a noticeable facial asymmetry and an open bite as a dislocation of the jaw from the frontal view. The swelling was firm and slightly tender to palpation. Palpation of the right TMJ revealed crepitus. Her maximal incisal opening was 30 mm.

While the patient's right TMJ symptoms of pain and joint sounds were consistent with another more common diagnosis, the presence of the preauricular swelling warranted immediate imaging of the affected area. A panoramic X-ray showed calcifying changes of the right side around the condylar head (Fig. 1). Subsequently, the patient was sent for a CT and a contrast-enhanced MRI. The CT scan showed the presence of reactive tissue, fluid content in the joint capsule and fragmentation of the cortical bone (Fig. 2). The MR image of the right side showed a large complex joint effusion with mild bony remodeling. There was significant enlargement of

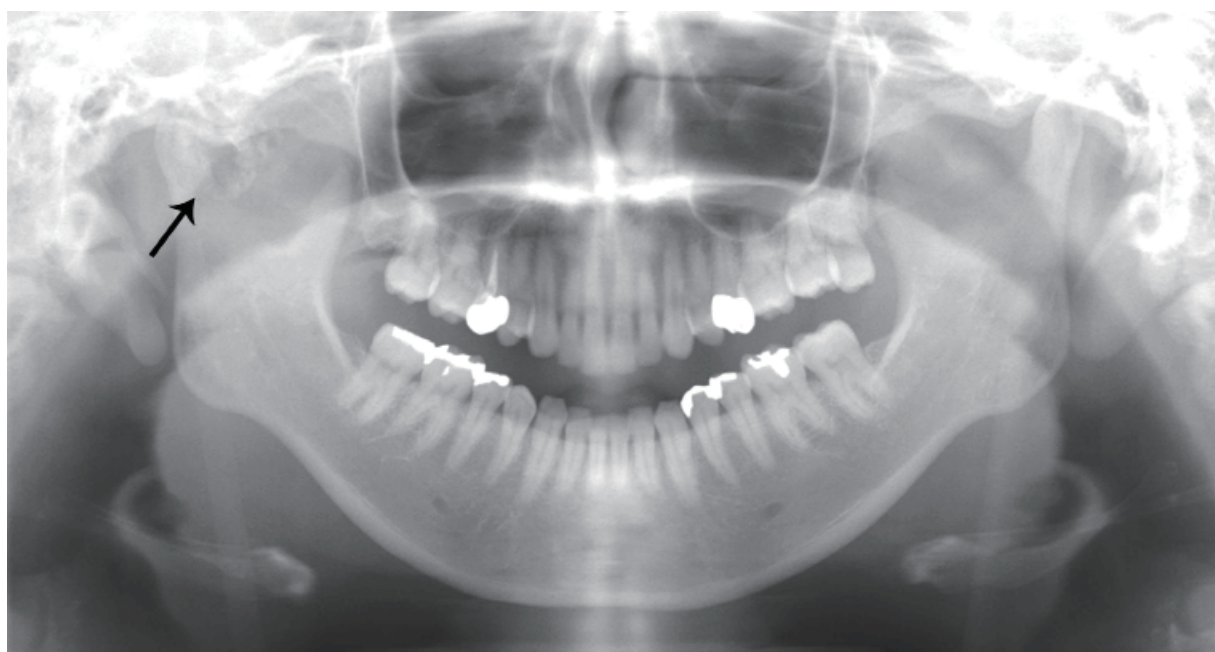


Fig. 1: Panoramic X-ray showing calcifying changes of the right side around the condylar head. (Arrow)

the joint capsule, medially, on the affected side, that resulted in inferior displacement of the mandibular condyle (Fig. 3). The left side was noted as relatively normal. It was still considered as a pre-operative diagnosis along with chondroma and synovial chondromatosis.

The patient's right TMJ was explored through a perauricular approach, and the joint

capsule was opened to expose the upper joint space. Lesions were fused with the articular disk. It was noted to be very friable and was removed in multiple small fragments with the articular disk. The findings correlated with Stage III disease, and the synovial surface was spared. After complete removal of the rice-like lesion (Fig. 4), the materials were sent for histological examination together with a portion of

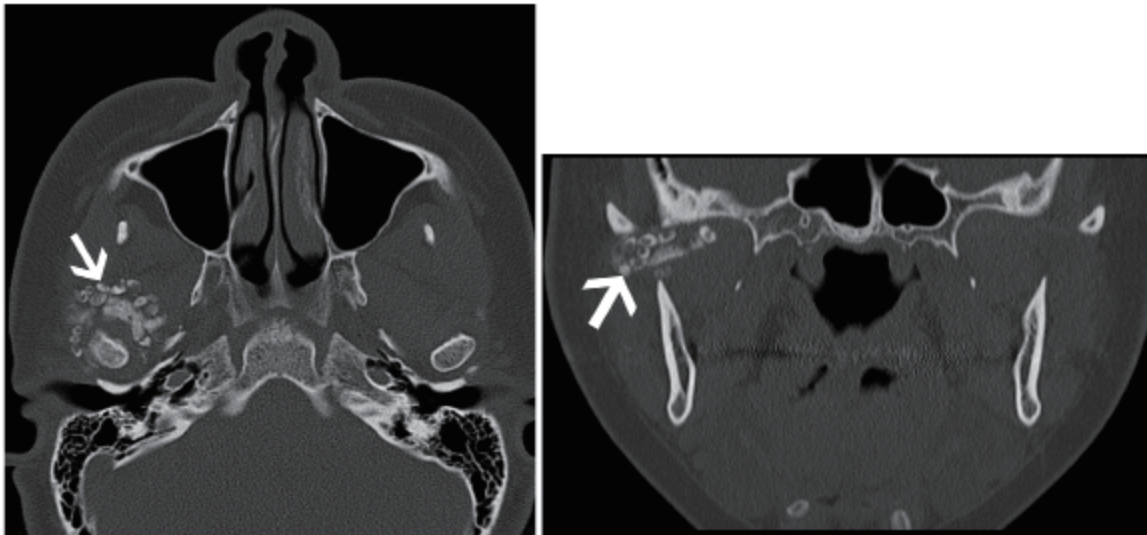


Fig. 2: CT-scan image showing calcification of the lesion in the axial and coronal planes. (Arrows)

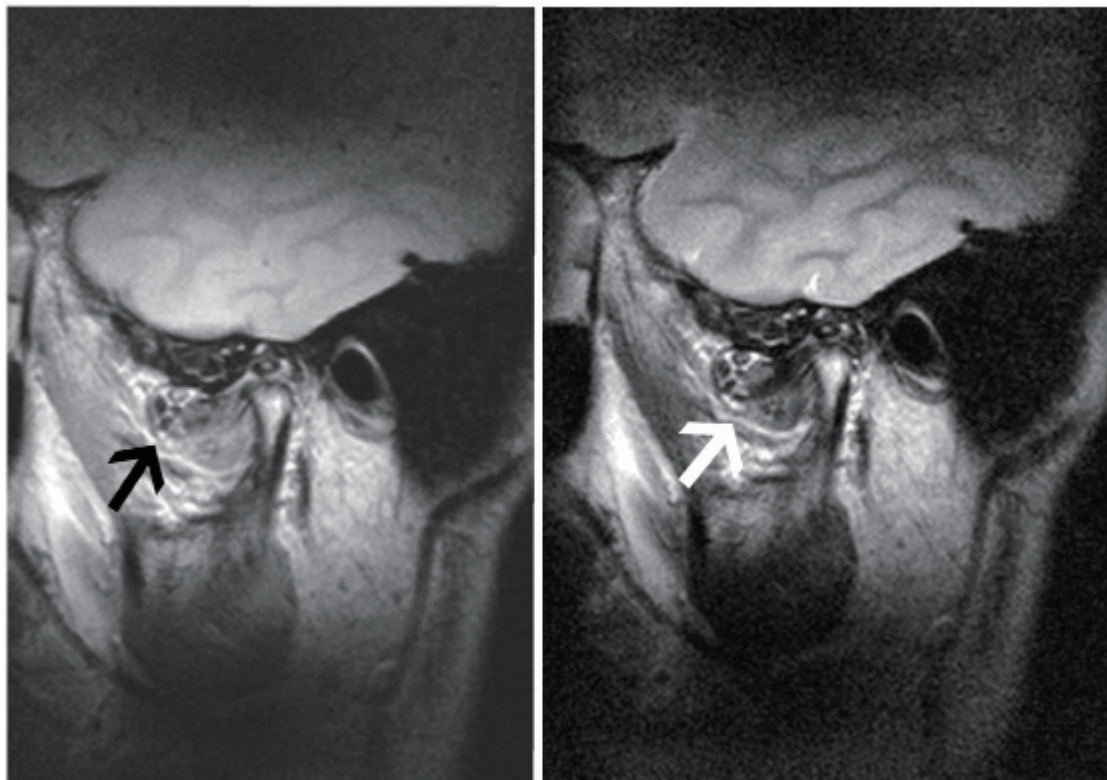


Fig. 3: Pre-operative sagittal MRI of the right TMJ (left: T2 weighted, right: H+ weighted) showing an expanding articular cavity and indicating a high signaling lesion. (Arrows)

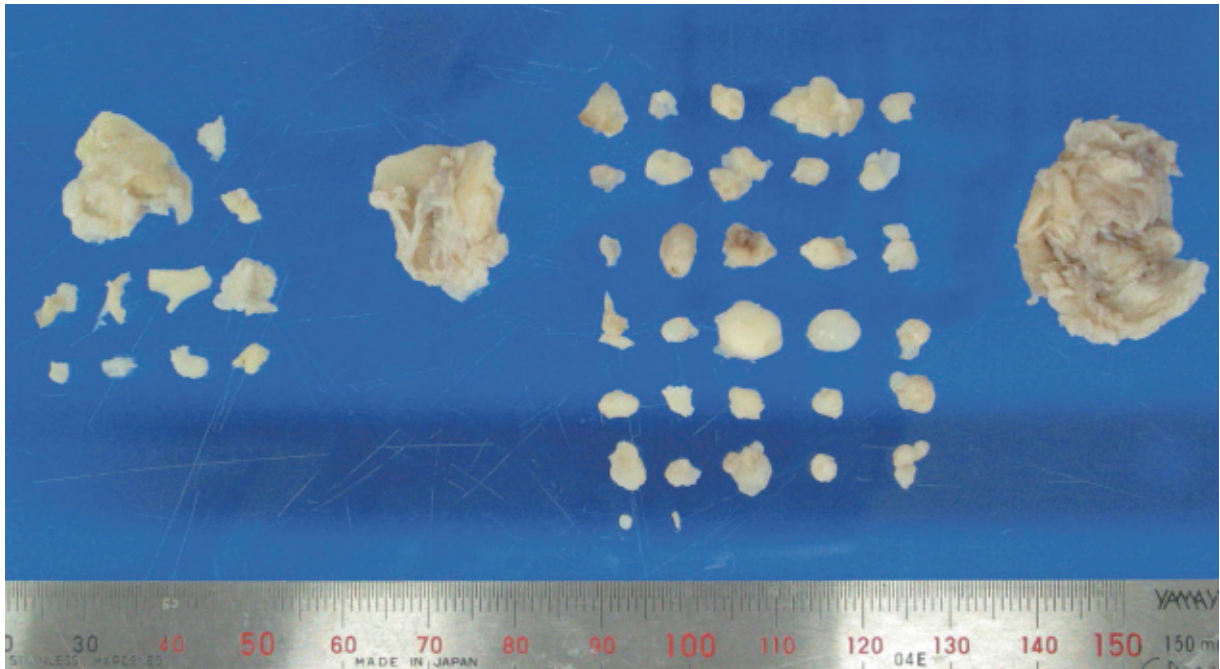


Fig. 4: Cartilaginous deposits, known as joint rice, after removal by surgical extraction.

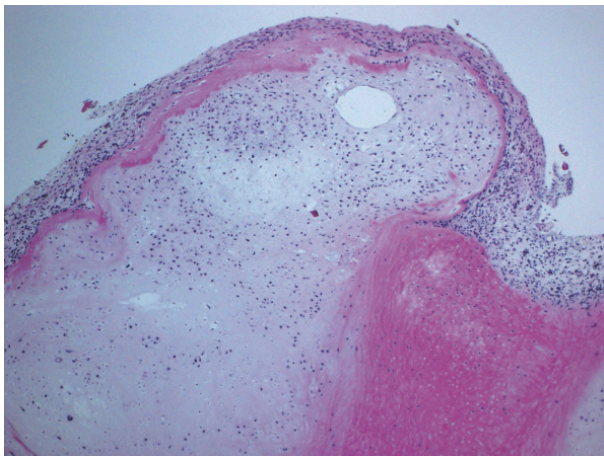


Fig. 5: Low magnification photomicrograph showing a nodule of hyaline cartilage exhibiting clustering of chondrocytes within the synovial tissue.

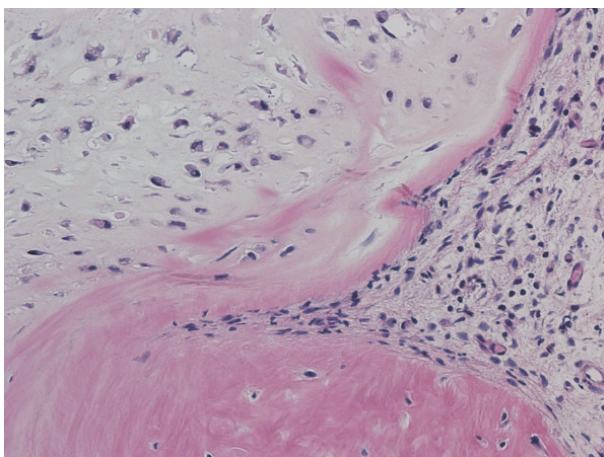


Fig. 6: High magnification photomicrograph showing a marked degree of atypical chondrocytes in the peripheral chondroid cells.

the articular capsule. The surgical site was copiously irrigated and a definitive closure was performed.

Histological examination of the material revealed variably sized nodules of hyaline cartilage exhibiting a clustering of chondrocytes within the subsynovial tissue (Fig. 5). In several areas, peripheral cellularity was present in which the peripheral chondroid cells showed a marked degree of atypical chondrocytes (Fig. 6). A diagnosis was made of SC with no evidence of atypia. Confirmation of that diagnosis was also obtained by an outside consulting pathology service.

The patient's postoperative course was unremarkable. Her maximal incisal opening increased to 49 mm. No recurrence was apparent six months after the operation. She was scheduled for continued follow-up in the future to monitor for recurrence.

Discussion

SC is most likely caused by metaplasia of the mesenchymal tissue rests within the synovial membrane⁵. Whether or not a true etiology exists as the root cause of most cases of SC is largely unknown. Potential etiologies have been speculated to be mechanical and/or traumatic in origin, although

Table 1: Summary of data from studies included in review (2011-2014)

No.	First author	Year	Age (range)	gender	R/L	Clinical signs and symptoms	Diagnostic imaging technique	Ref. No.
1	Gonzalez-Perez L.M.	2011	50	M	L	pain, preauricular swelling, limitation of mouth-opening	CT, MRI, Histological examination	15
2	Varol A.	2011	46	F	L	pain, preauricular swelling, limitation of mouth-opening	OPT, CT, MRI, Histological examination	11
3	Lim S.W.	2011	49	M	R	pain, preauricular swelling, limitation of mouth-opening	OPT, CT, MRI, Histological examination	16
4	Mori Y.	2011	52	F	L	strange sensation	OPT, CT, MRI, Histological examination	17
5	Campbell D.I.	2011	59	M	L	pain, limitation of mouth-opening	MRI, Histological examination	18
6	Guijarro-Martinez. R.	2011	38	F	L R	pain, preauricular swelling,	CT, MRI, Histological examination	19
7	Chen M-J.	2011	50	M	L	pain, preauricular swelling, limitation of mouth-opening	OPT, CT, MRI, Arthroscopy, Histological examination	20
8	Matsumura Y.	2012	46	M	R	pain, preauricular swelling, limitation of mouth-opening	OPT, CT, MRI, Histological examination	21
9	Mikami T.	2012	30 51 59	F3	L3	pain, preauricular swelling, limitation of mouth-opening	CT, MRI, Histological examination	22
10	Wang P.	2012	45.3 (17-64)	F15 M7	L10 R12		MRI	23
11	Cai X.Y.	2012	43 (21-62)	F26 M7	L15 R18	pain, limitation of mouth-opening, joint sounds(33/33),	MRI, Arthroscopy, Histological examination	12
12	Cho Y.A.	2013	2	M	L	pain, limitation of mouth-opening	CT, Histological examination	24
13	Poveda-Roda R.	2013	45.5 (8-81)	F93 M33		pain, preauricular swelling,	CT, MRI, Arthroscopy, Histological examination	25
14	Wake M.	2013	62.3 (32-81)	F6, M4		pain, limitation of mouth-opening		26
15	Matsumoto K.	2013	34	F	R	pain, preauricular swelling, limitation of mouth-opening	OPT, CT, MRI, Arthroscopy, Histological examination	27
16	Reed L. S.	2013	31	F	R	pain, preauricular swelling, limitation of mouth-opening	MRI, Histological examination	13
17	Valentini V.	2013	60	F	L	preauricular swelling,	OPT, MRI, Histological examination	28
18	Colean H.	2013	63	F	L	pain, preauricular swelling, limitation of mouth-opening	CT, MRI, Histological examination	29
19	Pau M.	2013	70	M	R	pain, preauricular swelling, limitation of mouth-opening	CT, MRI, Histological examination	14
20	Sink J.	2014	82	F	L	Alzheimer's disease, epilepsy	MRI, Histological examination	30
21	Kim S.M.	2014	49	F	L	pain, preauricular swelling, limitation of mouth-opening	CT, MRI, Histological examination	31
22	Li Y.	2014	14 27 20	F2 M1	L1 R2			32
23	Peyrot H.	2014	42	F	L	pain, preauricular swelling, limitation of mouth-opening	MRI, Histological examination	33

analysis shows that a known history of facial trauma is rare⁶⁾. When the disease lacks a known cause, it is classified as primary; conversely when the patient reports a history of pre-existing TMJ disease and/or trauma, it is classified as secondary³⁾. However, this classification system may be equivocal since patients may be unaware of previous subclinical microtrauma and/or joint disease and may thus be classified as primary.

The histological staging of SC was published by Milgram in 1977, and this classification system has been used in all studies published in the literature. Over 80% of patients are already at Milgram's Stage III when the diagnosis of SC is made⁷⁾. The histological (Milgram's) staging is based on disease activity and development¹⁾. Stage I is the early stage and represents intrasynovial metaplasia without loose bodies; perifocal macrophages may

Table 2. Summary of data from the present systematic review of the literature and the one by Guarda-Nardini et al., 2010³⁾

	Review by Guarda-Nardini		
	This review	et al, 2010	Total
Nr.of studies reviewed	7	102	109
Single case reports	16	87	103
Total cases	215	154	369
Male:female ratio	59:155	43:107	102:262 (1:2.6)
Left:right joint ratio	41:36	68:77	109:113 (1:1.04)

be present⁸⁾. Stage II is the transitional phase, in which synovial involvement and detached bodies are present. Stage III shows no more active intrasynovial disease (metaplasia), but the joint space is full of multiple detached bodies, and secondary calcification or ossification may be present. The secondary calcification is called Henderson-Jones syndrome. Fujita et al. also described metaplastic activity in Stage III patient⁹⁾. The loose bodies in the joint space might be surrounded by a layer of synovial membrane¹⁰⁾.

SC of the TMJ symptoms can include pain, clicking, tenderness, functional limitations and swelling⁵⁾. The differential diagnoses include osteoarthritis, osteochondroma, chondrocalcinosis (pseudogout), pigmented villonodular synovitis and osteochondritis dissecans²⁾. Taking a thorough history, including histories of facial trauma and previous TMJ pathologies, clinical examination and radiographic studies are thus paramount in making accurate preliminary diagnosis and ultimately prescribing proper treatment. Due to the lack of response with conservative measures, surgical removal is usually required. Treatment options consist of arthroscopy, arthrotomy with synovectomy, excision of cartilaginous bodies and/or possible discectomy⁵⁾. In review process, all articles pertaining to SC of TMJ were searched systemically in the National Library of Medicine's PubMed database. The search strategy consisted of searching the word synovial chondromatosis and Temporomandibular joint within the PubMed database. The summary of data from the present review, which systematically assessed all available publications from 2011 to the present (Table 1), and

that by Guarda-Nardini et al., which covered most papers published before 2010³⁾. Advance imaging techniques, such as CT and MRI, be helpful for depicting joint changes and the presence of loose bodies, suggesting the diagnosis of SC¹¹⁾. MRI may be useful for depicting the nodules in the early stages of formation, before ossification, and for planning surgery in the early stages. Surgery has always been recommended as the therapy of choice, but recently, 33 cases of SC of the TMJ were reported to have been treated with arthroscopy in patients with separate mass lesions and no extra-articular extension with encouraging results¹²⁾. Reed et al. and Pau et al. reported the importance of diagnosing SC of the TMJ using arthroscopy^{13, 14)}. In conclusion, we present a rare case of SC in the TMJ. And a systematic assessment of the literature on SC of the TMJ over the past four years allowed the identification of 215 new cases since the last comprehensive review by Guarda-Nardini et al.³⁾. 369 cases have been describe in 109 publication (Table 2).

References

- 1) Linger O, Zix J, Stauffer-Brauch EJ, Iizuka T: Synovial chondromatosis of the temporomandibular joint with cranial examination: a case report and literature review. *J Oral Maxillofac Surg*, 65: 2073-2080, 2007.
- 2) Balasundaram A, Geist JR, Gordon SC, Klasser GD: Radiographic diagnosis of synovial chondromatosis of the temporomandibular joint: a case report. *J Can Dent Assoc*, 75: 711-714, 2009.
- 3) Guarda-Nardini L, Piccotti F, Ferrontato G, Manfredini D: Synovial chondromatosis of the temporomandibular joint: a case description with systematic literature review. *Int J Oral Maxillofac Surg*, 39: 745-755, 2010.
- 4) Milgram JW: The classification of loose bodies in human joint. *Clin Orthop*, 124: 282-291, 1977.
- 5) Boffano P, Viterbo S, Bosco GF: Diagnosis and surgical management of synovial chondromatosis of the temporomandibular joint. *J Craniofac Surg*, 21: 157-159, 2010.
- 6) Zaw W, Zhao YF, Liu Y, Jiang L: A case of synovial chondromatosis of the temporomandibular joint secondary to preauricular trauma. *Int J Oral Maxillofac*

- Surg, 38: 1212-1215, 2009.
- 7) Shah SB, Ramanojam S, Ggadre PK, Gadre KS: Synovial chondromatosis of temporomandibular joint: journey through 25 decades and a case report, *J Oral Maxillofac Surg*, 69: 2795-2814, 2011.
 - 8) Blankestijn J, Panders AK, Vermey A, Scherpbier AJ: Synovial chondromatosis of the temporo-mandibular joint: report of three cases and review of the literature, *Cancer*, 86: 664-666, 1998.
 - 9) Fujita S, Yoshida H, Tojyo I, Wada T, Murakami K, Iisuka T: Synovial chondromatosis of the temporomandibular joint: clinical and immunohistopathological consideration, *Br J Oral Maxillofac Surg*, 42: 259-260, 2004.
 - 10) von Lindern JJ, Theuerkauf I, Niederhagen B, Berge S, Appel T, Reich RT: Synovial chondromatosis of the temporomandibular joint: clinical, diagnosis, and histomorphologic findings. *Oral Surg Oral Med Oral Pathol, Oral Radiol Endod*, 94: 31-38, 2002.
 - 11) Varol A, Sencimen M, Gurses A, Altug HA, Dumlu A, Kurt B: Diagnostic Importance of MRI and CT Scans for Synovial Osteochondromatosis of the Temporomandibular Joint, *J Craniomandibular Practice*, 29: 313-317, 2011.
 - 12) Cai XY, Yang C, Chen MJ, Jiang B, Zhou Q, Jin JM, Yun B, Chen ZZ: Arthroscopic management for synovial chondromatosis of the temporomandibular joint: a retrospective review of 33 cases, *Amer Assoc Oral Maxillofac Surg*, 70: 2106-2113, 2012.
 - 13) Reed LS, Michael D, Foster DF, John WH: Synovial Chondromatosis of the temporomandibular joint: a case report and literature review, *J Cra and Sleep Prac*, 31: 309-313, 2013.
 - 14) Pau M, Bicsak A, Reinbacher KE, Feichtinger M, Karcher H: Surgical treatment of synovial chondromatosis of the temporomandibular joint with erosion of the skull base: a case report and review of the literature, *Int J Oral Maxillofac Surg*, 43: 600-605, 2014.
 - 15) Gonzalez-Perez LM, Congregado-Cordoba J, Salinas-Martin MV: Temporomandibular joint synovial chondromatosis with a traumatic etiology, *Int J Oral Maxillofac Surg*, 40: 330-334, 2011.
 - 16) Lim S.W., Jeon S.J., Choi S.S., Choi K.H. Synovial chondromatosis in the temporomandibular joint: a case with typical imaging features and pathological findings, *British J Radiology*, 84: e215-e218, 2011.
 - 17) Mori Y, Kakudo K, Gotoh M, Kubo H, Akiyama H, Yotsui Y, Shimizutani K: A case of synovial chondromatosis of the temporomandibular joint followed for 17 years, *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*, 112: e35-e39, 2011.
 - 18) Campbell DI, Da Silca RK, Da Silva H, Simon SH, Rich AM: Temporomandibular Joint Synovial Chondromatosis With Intracranial Extensin: A Review and Observations of Patient Observed for 4 Years, *J.Oral Maxillofac Surg*, 69: 2247-2252, 2011.
 - 19) Guijarro-Martinez R, Torres MP, Mateo MM, Garcia IS, Alba LM, Gimilio MEI, Hermosa GPH, Gli JVP: Bilateral synovial chondromatosis of the temporomandibular joint, *J Cranio-Maxillo- Facial Surgery*, 39: 261-265, 2011.
 - 20) Chen MJ, Yang C, Zhang XH, Qiu YT: Synovial chondromatosis originally arising in the lower compartment of temporomandibular joint: A case report and literature review, *J Cranio-Maxillo-Facial Surg*, 39: 459-462, 2011.
 - 21) Matsumura Y, Nomura J, Nakanishi K, Yanase S, Kato H, Tagawa T: Synovial chondromatosis of the temporomandibular joint with calcium pyrophosphate dihydrate crystal deposition disease, *Dentomaxillofacial Radiology*, 41: 703-707, 2012.
 - 22) Mikami T, Aomura T, Ohira A, Kumagai A, Hoshi H, Takeda Y: Three Case Reports of Synovial Chondromatosis of Temporomandibular Joint: Histopathologic Analyses of Minute Cartilaginous Loose Bodies From Joint Lavage Fluid and Comparison With Phase II and III Cases, *J Oral Maxillofac Surg*, 70: 2099-2105, 2012.
 - 23) Wang P, Tian Z, Yang J, Yu Q: Synovial chondromatosis of the temporomandibular joint: MRI finding with pathological comparison, *Dentomaxillofacial Radiology*, 41: 110-116, 2012.
 - 24) Cho Y.A., Yoon HJ, Hong SD, Lee JI, Hong SP: A giant radiopaque mass in the masticatory space, *Oral Surg Oral Med Oral Pathol Oral Radiol*, 115: 566-570, 2013.
 - 25) Poceda-Roda R, Bagan JV, Sanchis JM, Margaix M: Pseudotumors and tumors of the temporomandibular joint. A review, *Med Oral Pathol Cir Bucal*, 18: e392-402, 2013.
 - 26) Wake M, Hamada Y, Kumagai K, Tanaka N, Ikeda Y, Nakatani Y, Suzuki R, Fukui N: Up-regulation of interleukin-6 and vascular endothelial growth factor-A in the synovial fluid of temporomandibular joints affected by synovial chondromatosis, *British J Oral and Maxillofacial Surg*, 51: 164-169, 2013.
 - 27) Matsumoto K, Sato T, Iwanari S, Kameoka S, Oki H, Komiyama K, Honda K: The use of arthrography in the diagnosis of temporomandibular joint synovial chondromatosis, *Dentomaxillofacial Radiology*, 42: 15388284, 2013.
 - 28) Valentini V, Arangio P, Egidi S, Capriotti M, Vellone V, Costrehini M, Boschi G, Cascone P, Calafati V, Torre U, Ricciardi I: Diagnosis and treatment of synovial chondromatosis of the TMJ: a clinical case, *Annali di Stomatologia*, IV: 269-272, 2013.
 - 29) Coleman H, Chandraratnam E, Morgan G, Gomes L, Bonar F: Synovial Chondrosarcoma Arising in Synovial Chondromatosis of the Temporomandibular Joint, *Head and Neck Pathol*, 7: 304-309, 2013.
 - 30) 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*, 117: e269-274, 2014.
 - 31) Kim SM, Hong SW, Ryu DJ, Huh JK: Chondroblastoma of the temporomandibular joint lateral capsule: a case report. *J Craniomandibular and Sleep Practice*, DOI10.1179, 2014.
 - 32) Li Y, El Mozen LA, Cai H, Fang W, Meng Q, Li J, Deng M, Long X: Transforming growth factor beta 3 involved in the pathogenesis of synovial chondromatosis of temporomandibular joint, *Scientific Reports*, 5:8843/ DOI:10.1038, 2015.
 - 33) Peyrot H, Montoriol RF, Beziat JL, Barthelemy I: Synovial chondromatosis of the temporomandibular joint: CT and MRI findings, *Diagnostic Interventional Imaging*, 95: 613-614, 2014.