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Pseudotumor in the temporomandibular joint: A case report

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ABSTRACT

INTRODUCTION: Neoplastic disease in the temporomandibular joint (TMJ) is a rare condition and is difficult to differentiate from temporomandibular disorders (TMD) based on symptoms and simple X-ray examinations. Potential differential diagnoses include synovial chondromatosis and pseudotumor, both of which are also relatively rare in the TMJ.

PRESENTATION OF CASE: We report a case of pseudotumor of the TMJ that was difficult to differentiate from synovial chondromatosis in a 71-year-old woman with a chief complaint of pain in the left TMJ. MRI of the right TMJ initially led to diagnosis of synovial chondromatosis. Extirpation of the lesion was performed under general anesthesia. Histopathological findings of the resected specimen revealed inflammatory granulation tissue without cellular atypism.

DISCUSSION: The pathological findings for the resected specimen were compatible with pseudotumor of the TMJ. These findings were not supportive of synovial chondromatosis or other tumor diseases.

CONCLUSION: This case illustrates the importance of careful examination of a mass lesion in the TMJ for differentiation from other TMJ-related diseases.

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1. Introduction

Synovial chondromatosis or other tumors can occur in the temporomandibular joint (TMJ) [1–6], but neoplastic disease of the TMJ is a rare condition. Pseudotumor is identified as a space-occupying lesion with non-specific inflammation that is difficult to differentiate from neoplasms. A few cases of pseudotumor related to the TMJ have been described [7,8]. Here, we report a case of pseudotumor in the right TMJ that was initially diagnosed as synovial chondromatosis.

2. Presentation of case

The patient was a 71-year-old female with a complaint of pain of the TMJ region during mouth opening and closing. Her medical history included gastroesophageal reflux disease, hypothyroidism, hysterosarcoma, hyperlipidemia, and gastric polyp. She had no traumatic history of the craniofacial region. She became aware of right side TMJ pain in 2001. She was treated with occlusal appliance under a diagnosis of temporomandibular disorder (TMD) at a local dental clinic, but her symptoms did not improve.

She was referred to the TMD clinic at the Dental Hospital of Tokyo Medical and Dental University in March 2003. Limitation of mouth opening with TMJ pain was improved by physical therapy and instructions on tooth contact habit (TCH) control, but she relapsed in May 2009. A right side TMJ tumor was suspected based on MRI findings, and she was referred to the Department of Maxillofacial Surgery of Tokyo Medical and Dental University for surgical treatment in July 2009.

Clinical examination revealed limitation of mouth opening with right side TMJ pain including TMJ sound (crepitation). Maximal mouth opening (MMO) without pain was 36 mm (interincisal) with crepitation of the right side TMJ. No osseous changes were observed at the mandibular condyle and mandibular fossa on X-P. MRI revealed that the upper joint space was expanded, with joint effusion in which low signal intensity spots were apparent (Fig. 1a–e). Fibrous adhesion was suspected on arthrography (Fig. 2a and b). Deformity and erosive changes were observed in the lateral part of the mandibular condyle, which was connected to a mass lesion, and the articular disc seemed to be perforated (Figs. 1 d, e and 2 b). These findings led to diagnosis of a tumor of the right side TMJ, which was strongly suspected to be synovial chondromatosis based on the MRI and arthrography findings for the upper joint space (Figs. 1 and 2).

Extirpation of the suspected tumor was performed under general anesthesia in November 2009. Surgery was performed through a temporo-preauricular incision (Figs. 3 a–d and 4 a) and both upper and lower joint spaces were opened for observation. Inconsistent

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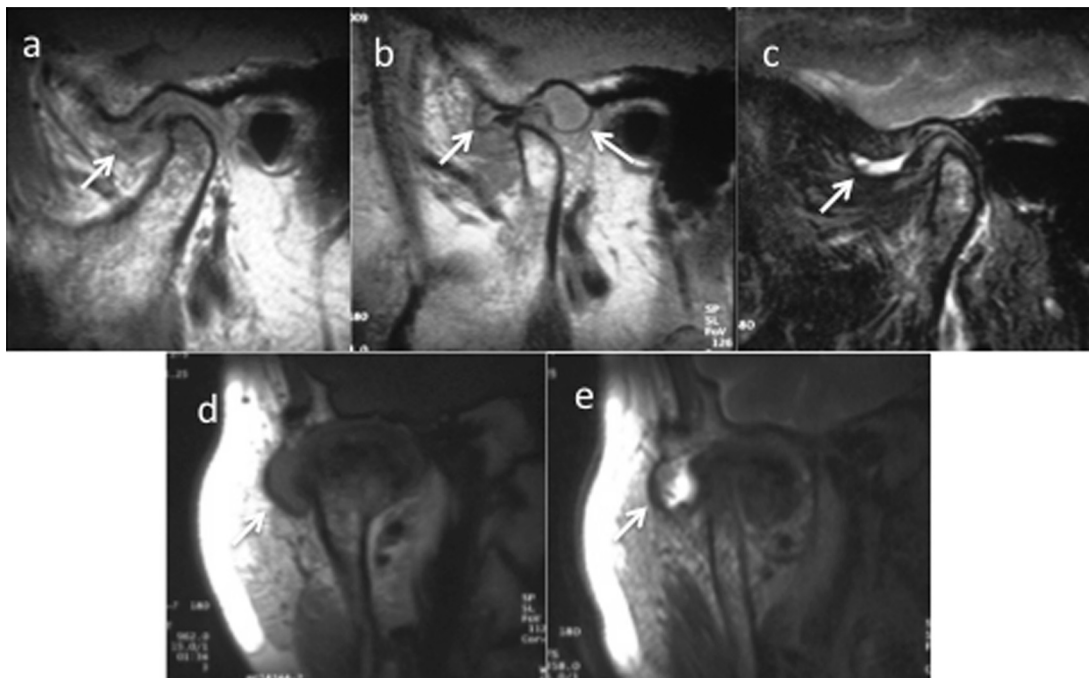


Fig. 1. MRI of the right side TMJ. a: Sagittal PDWI (closed position). b: Sagittal PDWI (open position). c: Sagittal T2WI (closed position). d: Frontal PDWI (closed position). e: Frontal T2WI (closed position). A medium signal intensity area thought to be due to a mass lesion was observed in PDWI and expansion of the upper joint space was suspected (a, b, d, arrows indicate). Joint effusion with hypertrophy of the synovium with low signal intensity spots in the upper joint space was found on T2WI (c, e, arrows indicate).

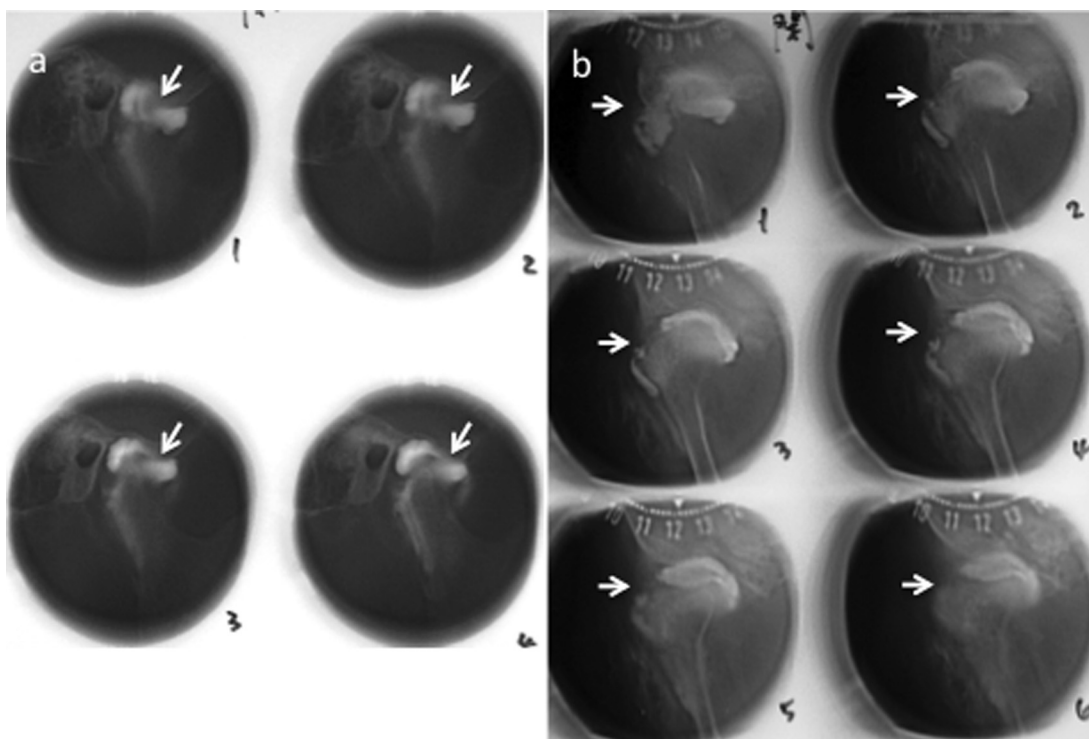


Fig. 2. Arthrography of the right side TMJ. a: Sagittal plane in the closed position. b: Frontal plane in the closed position. Fibrous adhesion was suspected (arrows indicate).

with the preoperative MRI findings, there were no free bodies due to suspected synovial chondromatosis in the joint space. A granulation mass lesion occupied the lateral part of the joint space. The lesion was connected to the articular disc in the medial part of the joint space and adhered to the bone surface of the mid to lateral

aspect of the mandibular condyle (Fig. 4b and c). The mass lesion was resected en bloc with the articular disc (Fig. 4b and c).

Pathological findings of the resected specimen showed granulation tissue with peripheral vasodilatation and inflammatory cell infiltration, including plasma cells and lymphocytes, in synovial tis-

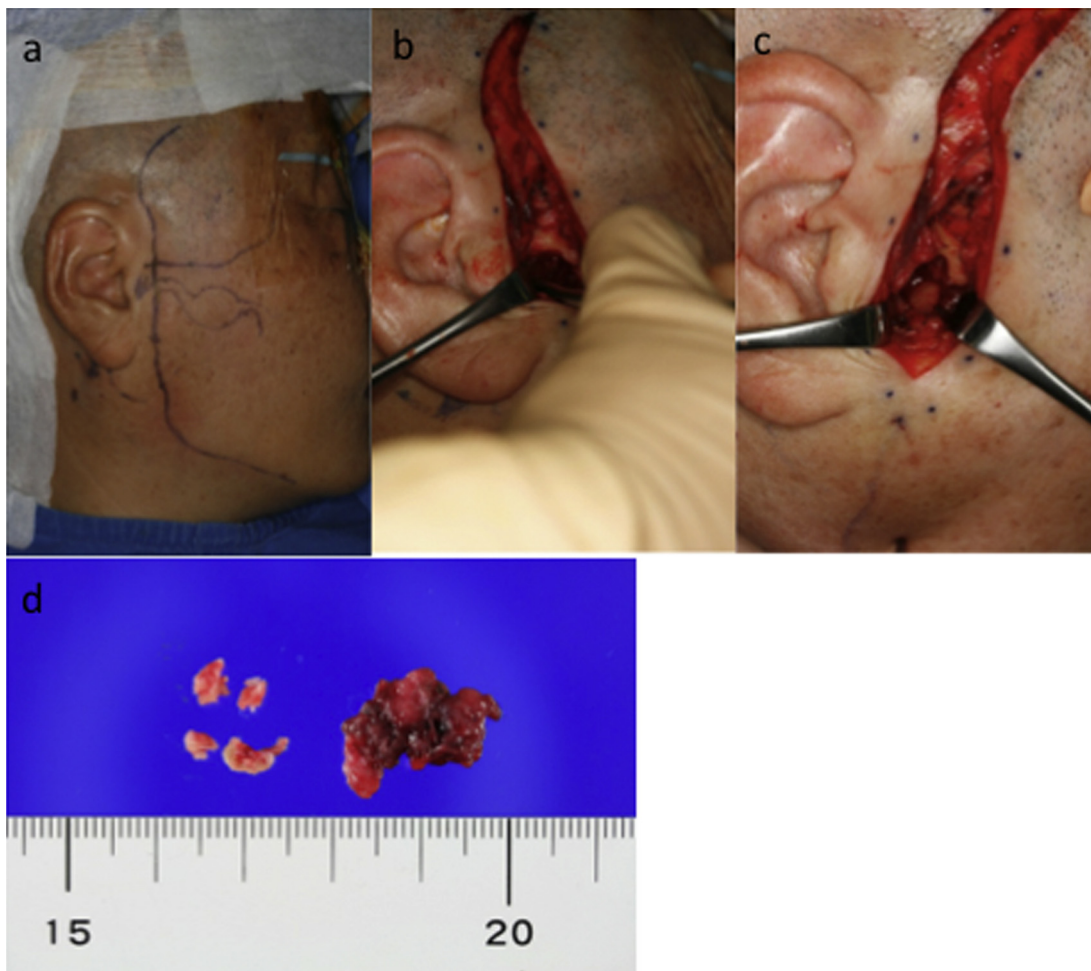


Fig. 3. Intraoperative photographs (a–c) and the resected specimen (d).

sues (Fig. 5a–c) These findings were compatible with inflammatory granulation tissue.

TMJ pain and limitation of mouth opening improved postoperatively. MMO increased to 41 mm at one month postoperatively and finally increased to 47 mm without TMJ pain. TMJ crepitation and pain completely disappeared, and malocclusion due to the change of condylar position and abnormal bone change of the TMJ were not observed 2 years postoperatively.

3. Discussion

Neoplasms in the TMJ are rare. Mass lesions in the TMJ can limit mouth opening and cause TMJ sounds and pain. However, these symptoms are similar to those in TMD. Thus, it is important to differentiate such lesions from TMD, but this is difficult based on clinical symptoms and simple X-P examinations [2–8]. CT images also cannot be used because TMJ diseases generally involve soft tissue without bone changes. In contrast, MRI can provide important information on TMJ conditions [7].

In our case, the patient was initially diagnosed and treated for TMD at a local clinic, but was ultimately found to have pseudotumor of the TMJ. This case illustrates the importance of multiple examinations for a mass lesion in the TMJ, and the need for differentiation from other TMJ-related diseases. Mass lesions or free bodies in the TMJ are often due to synovial chondromatosis [2,3] and preoperative examinations in our patient suggested synovial

chondromatosis in the upper joint space of the right side TMJ. Therefore, extirpation of the TMJ lesion was performed under general anesthesia. However, free bodies or tumors isolated in joint spaces were not observed, and the lesion was found to consist of inflammatory granulation tissue that connected to the lateral part of the articular disc.

The patient had no history of maxillofacial injuries, and the etiology of the mass lesion with granulation tissues was unclear. Pathological findings for the resected specimen revealed inflammatory granulation tissues without cellular atypism or invasive bone resorption. These findings allowed rejection of diagnosis of synovial chondromatosis or another tumor, but were compatible with so-called pseudotumor.

The etiology of pseudotumor is unclear, but is thought to be related to inflammatory changes evoked by the wound healing process after injury or infection. The first description of inflammatory pseudotumor was given by Umiker et al. in 1954, in a case report of a mass lesion in the lung [9]. Pseudotumor can occur in organs such as skin, lung, upper respiratory tract and the digestive apparatus. In the craniofacial region, there are reports of pseudotumors in the orbit [10], nasal cavity, paranasal sinus, larynx and thyroid gland. Fibro-osseous pseudotumors of the finger with swelling and pain in soft tissues around bones have also been described [11,12]. These cases illustrate that pseudotumor can occur in many organs.

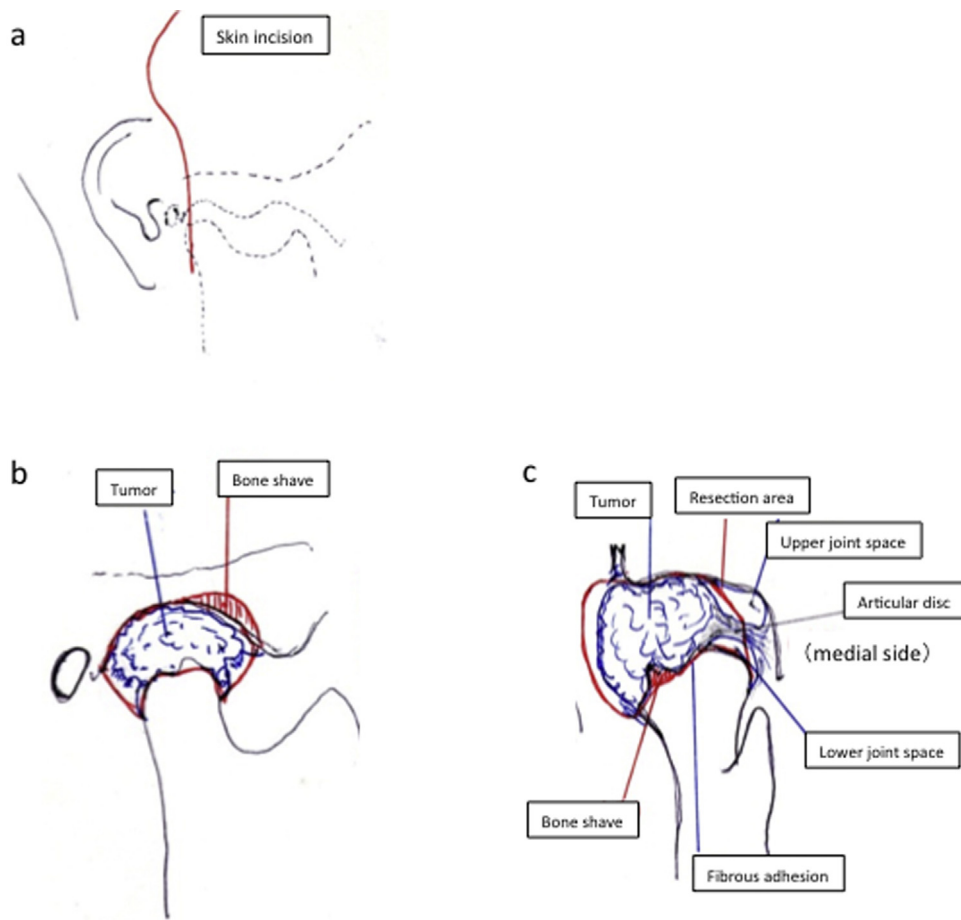


Fig. 4. Intraoperative findings. These illustrations are taken from the operative notes for this case. The annotations were translated into English by the author.

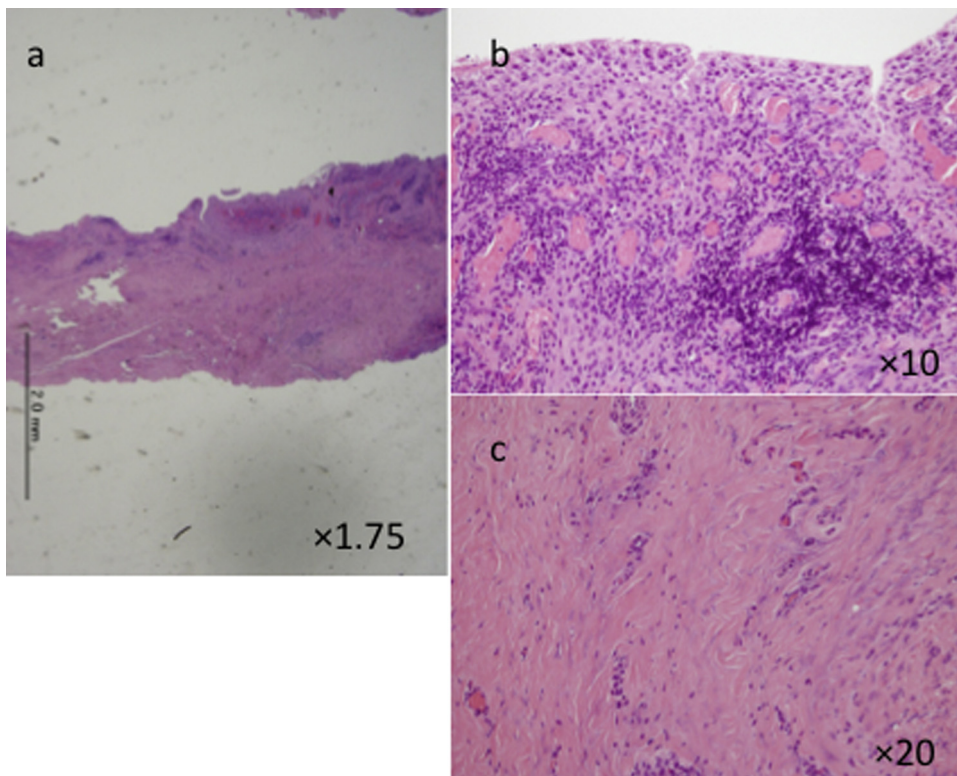


Fig. 5. Pathological findings showed granulation tissue with peripheral vasodilation and inflammatory cell infiltration, including plasma cells and lymphocytes, in synovial tissues.

4. Conclusion

In our case, the mass lesion was connected to the synovial membrane and articular disc, and consisted of granulation tissue with fibrous connecting tissue and inflammatory cell infiltration. Clinical and pathological findings indicated a pseudotumor in the TMJ induced by micro-injuries in the upper or lower joint space.

Conflict of interest

None.

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None.

Ehtical approval

None.

Consent

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

Author contributions

Hiroyuki Yoshitake, Ryousuke Nkamura, So Wake 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.

Research registry

N/A.

Guarantor

Hiroyuki Yoshitake.

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