Tophaceous pseudogout of the temporomandibular joint extending into the cranium: a case report with literature review

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Abstract

Pseudogout is a disease characterized by calcium pyrophosphate crystal deposition. Involvement of the temporomandibular joint (TMJ) is rare. We herein report a case of tophaceous pseudogout of the TMJ with cranial extension. An 83-year-old woman was referred to our institution for treatment of right TMJ pain. The patient's medical and family histories were unremarkable. Magnetic resonance imaging showed a mass of about 35 mm in diameter compressing the bottom of the right temporal lobe of the brain. Based on a clinical diagnosis of a right TMJ tumour, biopsy was performed under general anaesthesia. The histopathological diagnosis was pseudogout. Considering the risk of surgically induced brain damage, the patient's advanced age and her relatively good quality of life, the treatment plan simply involved the observation of the lesion. Fourteen months after biopsy, the patient's activities of daily living remained unchanged and she had no TMJ pain.

INTRODUCTION

Pseudogout is a disease characterized by calcium pyrophosphate crystal deposition, and it usually occurs in individuals older than 50 years. The most frequently involved joint is the knee, followed by the wrists, elbows, shoulders and ankles [1]. Involvement of the temporomandibular joint (TMJ) is rare; furthermore, only nine cases of pseudogout of the TMJ extending into the skull base have been reported in the English-language literature.
literature [2–9]. Because of the rarity of this condition, the
treatment outcome of such cases was not elucidated.

We herein report a case of large pseudogout of the
TMJ with cranial extension and present a review of the
literature.

CASE REPORT
An 83-year-old woman was referred to our institution for
treatment of right TMJ pain. She had first noticed the
pain ~3 years earlier. The patient’s medical and family
histories were unremarkable. Intra-oral examination
revealed no abnormal findings associated with
the right TMJ pain. Occlusal deviation was not observed.
The maximum mouth opening was 28 mm, and trans-
sient pain occurred during mouth opening. A hard
protrusion was observed in the right TMJ region. There
was no evidence of cranial nerve paralysis or cervical
lymphadenopathy. On T1- and T2-weighted magnetic
resonance coronal images, the mass showed low signal
intensity and compressed the bottom of the right
temporal lobe of the brain. The mass about 35 mm in
diameter was inhomogeneously enhanced by gadolinium
(Fig. 1). The patient’s calcium, phosphate and uric acid
concentrations were within the reference range.

Based on a clinical diagnosis of a right TMJ tumour,
biopsy was performed under general anaesthesia (Fig. 2).
Histopathological examination revealed lobular basophilic
materials surrounded by fibrous tissue. Various sized
rectangular or parallelogram-shaped crystals were irreg-
ularly present within the basophilic materials (Fig. 3).
These crystals were identified under polarized light.

The result of X-ray diffraction analysis of the biopsy
specimen was consistent with the pattern of calcium
pyrophosphate crystals (Fig. 4).

The final diagnosis of tophaceous pseudogout of the
right TMJ was made based on these findings. A neurosur-
gical consultation in our hospital was performed. Con-
sidering the risk of surgically induced brain damage, the
patient’s advanced age and her relatively good quality of

![Figure 4. The result of X-ray diffraction analysis of the biopsy specimen showing the pattern of calcium pyrophosphate crystals.](image-url)
life, the treatment plan simply involved the observation of the lesion with pain control. Fourteen months after biopsy, the patient was pain-free and her activities of daily living had remained unchanged.

**DISCUSSION**

To the best of our knowledge, 10 cases (including the present case) of pseudogout of TMJ extending into the skull base have been reported in the English-language literature (Table 1). The most frequent clinical symptoms in these cases were swelling and pain in five patients and trismus in three patients. No patients had symptoms related to a central nervous system disorder. Pseudogout was pre-operatively diagnosed in 8 of the 10 patients. The remaining two patients were diagnosed with a neoplastic lesion or synovial osteochondromatosis [5] and synovial chondromatosis [7], respectively. General treatment of pseudogout is supportive to minimize symptoms [1]; however, tophaceous pseudogout that destroys surrounding structures sometimes requires surgery. Surgery was performed in six (75%) of the eight reported cases correctly diagnosed as pseudogout pre-operatively. Cerebrospinal fluid leakage occurred as an intra-operative complication in one patient [8]. Exposure of the dura mater was overlaid using a flap of temporal muscle [2, 5, 9], temporalis fascia [7], harvested fat [2] and bone wax [8]. No patients developed brain damage as a post-operative complication. However, conductive hearing loss was reported in one 83-year-old patient [9]. To avoid possible surgically induced complications, observation was selected in two patients, including ours. These patients experienced no deterioration of clinical symptoms within the follow-up period [4]. Because our patient’s oral dysfunction in daily life was mild, observation with pain control was selected.

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**CONFLICT OF INTEREST STATEMENT**

None.

**ETHICS STATEMENT/CONFIRMATION OF PATIENT’S PERMISSION**

Ethics approval was not required. Written consent was obtained by patient for use of the photographs in this report.

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