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Case Report
Clinical Pathology

A case of tophaceous pseudogout of the temporomandibular joint extending to the base of the skull

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Abstract. A case of tophaceous pseudogout (i.e., calcium pyrophosphate dihydrate (CPPD) crystal deposition disease) in the temporomandibular joint (TMJ), extending to the base of the skull, is reported. A 38-year-old man was referred to the hospital with mild pain in the right chin and tip of the tongue. Panoramic radiography showed a large calcified mass around the right TMJ. Computed tomography imaging revealed a large, granular, calcified mass surrounding the right condylar head and extending to the base of the skull. The mass was clinically and radiographically suspected to be a pseudogout lesion. A biopsy specimen was collected under general anaesthesia to confirm the diagnosis. On histology, the mass was found to contain deposits of numerous rod-shaped and rhomboid crystals, which suggested tophaceous pseudogout. The deposits were identified as CPPD crystal deposition, based on analysis by X-ray diffraction and Fourier transform infrared spectroscopy. These two crystallography methods were useful in confirming the diagnosis of CPPD crystal deposition disease in the TMJ.

Key words: calcium pyrophosphate dihydrate; Fourier transform infrared spectroscopy; temporomandibular joint; tophaceous pseudogout; X-ray diffraction.

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McCarty et al. first identified calcium pyrophosphate dihydrate (CPPD) crystals rather than sodium urate crystals in the synovial fluid of patients who had gout-like symptoms; they termed the condition 'pseudogout'.[1] The term 'tophaceous pseudogout' has recently been used to describe lesions that have massive or tumoural CPPD crystal deposition. This variant is one of the rarest forms of CPPD deposition disease; however, it is important because it shares a histological and clinical resemblance to cartilaginous tumours.[2,3]

A case of tophaceous pseudogout in the temporomandibular joint (TMJ) extending to the base of the skull is presented herein. Images of tophaceous pseudogout obtained through X-ray diffraction (XRD) and Fourier transform infrared spectroscopy (FT-IR) are also presented.

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Case report

A 38-year-old man was admitted to the hospital for the diagnosis and treatment of mild pain in the right chin and tip of the tongue. He had first noticed mild pain in the right chin 2 months before admission, and had undergone root canal treatment of his right lower second molar at a neighbouring dentist. However, the pain in the area did not subside and he was admitted to hospital.

The patient's medical history included hyperlipidemia, gout, diabetes, and hypertension. These conditions were all well controlled with medication. It was noted that he had been involved in sumo wrestling since he was 16 years old. He was a high school teacher and coach of the sumo club. As part of his role as a sumo coach, he always received the students' tackles (i.e., *ukemi* ('passiveness')) on the right half of his body, including his right chin.

Clinical examination showed an obvious pre-auricular swelling on the right side, which was tender to the touch. His intrinsic mouth opening was limited to 38 mm. However, he did not have any symptoms that affected his day-to-day function and quality of life.

Computed tomography (CT) scans revealed a calcified mass around the right TMJ, but it was not continuous with the mandibular condyle (Fig. 1A). The calcified mass was pressing on the temporal bone, and erosive bone resorption had occurred at the base of the skull (Fig. 1B). The CT views of the right TMJ showed limited opening positions due to the right condylar opaque mass. In addition, the calcified mass around the mandibular condyle gained mobility and changed shape on translating the joint from the open to the closed position (Fig. 1C). A pseudogout crystal mass was suspected on the basis of these CT views. However, it was not possible to completely confirm that the lesion was not a tumour.

Because the lesion extended to the base of the skull, the case was discussed with neurosurgical specialists. As a result of the discussion, a biopsy of the mass was performed under general anaesthesia to confirm the diagnosis. An intraoral incision was created on the mucosal membrane at the anterior margin of the mandibular ramus. The periosteum of the lateral surface of the mandibular ramus towards the coronoid process was detached. After reaching the mandibular notch, a significant amount of intracapsular calcareous material, which appeared chalky or 'gristlike', was identified between the coronoid process and the articular process (Fig. 2A). The biopsy samples were gently removed from the inside of the mass using a sharp surgical spoon (Fig. 2B).

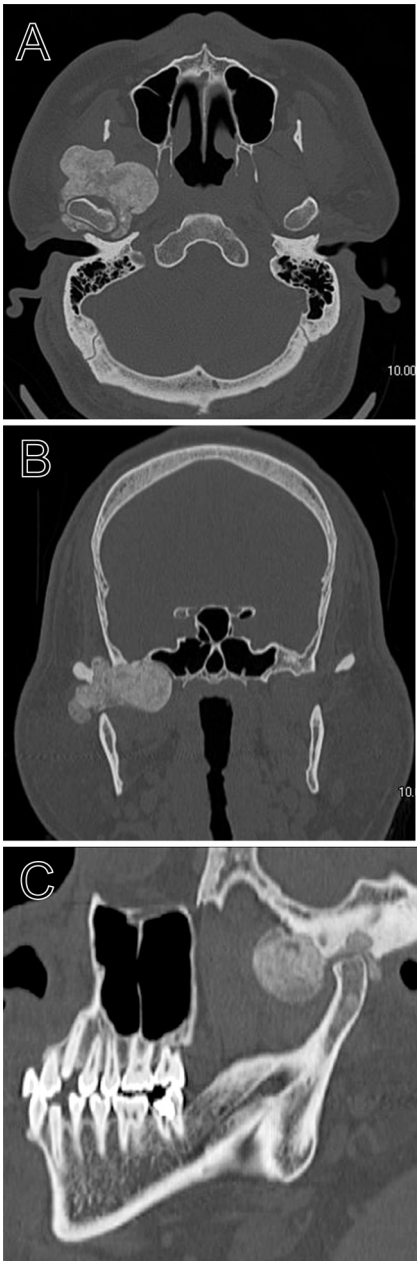

Fig. 1. Computed tomography (CT) imaging showing a large calcified mass around the right temporomandibular joint (TMJ). (A) Axial CT scan showing a ring-shaped calcified mass around the condylar process of the right TMJ; the mass is not continuous with the mandibular condyle. (B) Coronal CT scan revealing a calcified mass in the joint space; bone resorption and thinning of the middle cranial base are present and the lesion appears to extend into the middle cranial fossa. (C) Sagittal CT scan of the right TMJ; the calcified mass limits condylar head movement.

One-half of the biopsy specimen was immersed in 10% formalin solution and used for the pathological examination. This examination of the specimen revealed that the mass contained numerous deposits of rod-shaped or rhomboid crystals. The background of the crystals was cellular fibrous tissue and foreign body-type giant cells (Fig. 2C). These views suggested CPPD crystal deposition.

For the differential diagnosis, the remaining half of the specimen was examined by XRD and FT-IR spectroscopy using the potassium bromide (KBr) disc method. The XRD profile was recorded using a D8 Advance diffractometer (Bruker AXS, Karlsruhe, Germany) operated at 40 kV–40 mA acceleration using copper–potassium alpha (Cu Kα) radiation. The patterns obtained by XRD showed that nearly all peaks corresponded to those of CPPD (JCPDS-ICDD-PDF#00-041-0488), although some peaks with an unknown pattern were also observed (Fig. 3A). The FT-IR spectrum was recorded on an FT/IR-6700 spectrometer (JASCO, Tokyo, Japan) at a resolution of 4 cm^{-1}. The FT-IR spectrum of calcium pyrophosphate tetrahydrate (CPPT) crystals has been reported previously,[4] and it was found to be similar to the FT-IR spectrum obtained in this study (Fig. 3B). In brief, the following were recorded: the O–P–O bending vibrations at 509 cm^{-1} and 568 cm^{-1}; P–O stretching vibrations at 923 cm^{-1}, 990 cm^{-1}, 1037 cm^{-1}, and 1089 cm^{-1}; O–H plane bending vibration at 1659 cm^{-1}; and broad peak around 3300 cm^{-1} due to the absorption of water. These data provided further support for the histological diagnosis of CPPD deposition disease, including tophaceous pseudogout.

After the diagnosis was confirmed, the treatment plan was discussed with the neurosurgeons. The patient was not experiencing any disruption to his daily function or everyday life as a result of the lesion, aside from the mild pain in the joint. Furthermore, the lesion was not neoplastic. Based on the situation and the clinical and pathological findings, the neurosurgeons advised against resection, because severe neurological dysfunction could result. The patient ultimately did not wish to have the mass resected. He continued to coach sumo wrestling, but decided that he should not receive student tackles to the right chin in the future. Three years after the first visit, the mass showed virtually no change in size.