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Calcium pyrophosphate dihydrate crystal deposition of the temporomandibular joint: A case report and review of the literature

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ABSTRACT

Calcium pyrophosphate dihydrate crystal deposition disease (CPPDd) induces arthritis in the surrounding joints with symptoms, such as pain, swelling, and restricted joint movement. CPPDd occurs primarily in the major joints; however, the main manifestation of this disease in the temporomandibular joint (TMJ) is relatively rare. We describe a case of CPPDd in the TMJ of a 54-year-old Japanese woman who was referred to our hospital with the complaints of unpainful swelling in the right TMJ region and restricted mouth opening. Computed tomography (CT) and magnetic resonance imaging (MRI) showed 46 × 42-mm and 27 × 18-mm calcified masses in the right TMJ region. The masses were both outside and inside the mandibular condyle, both of which extended to the skull base. Since we suspected a tumor in the TMJ, the lesion was partially excised for biopsy with the patient under general anesthesia. The microscopic examination showed deposits of calcium pyrophosphate dihydrate crystals, which resulted in a histopathologic diagnosis of CPPDd. According to the suggestion of the neurosurgeon, we removed as much as possible of only the CPPDd tissue on the outside of the mandibular condyle via the preauricular approach to avoid damaging the cranial nerves. The patient was satisfied with the postoperative facial symmetry without limited mouth opening. The postoperative follow-up was uneventful.

1. Introduction

Calcium pyrophosphate dihydrate crystal deposition disease (CPPDd) is a disorder characterized by non-infectious joint inflammation with calcification in the joint cartilages, synovial membranes, and surrounding soft tissues [1–3]. In 1960, Zitnan and Sitai initially proposed CPPDd, also known as pseudogout and articular cartilage calcification [4]. In 1976, Pritzker et al. [5] first described CPPDd in the temporomandibular joint (TMJ). CPPDd also has been reported in degenerative or necrotic tissue, resulting from secondary trauma [2,6].

CPPDd in the TMJ is diagnosed according to the following criteria as described by Kohn et al [7]: identification of crystalline components by such means as X-rays analysis and chemical analysis, identification of the appearance and shape of crystals by corrected polarization micro-

scopy, or identification of calcification by X-ray. CPPDd develops most often in the large joints, i.e., knee, wrists, elbows, shoulders, and ankles, but seldom affects the small joints, making CPPDd of the TMJ relatively rare [8].

We report a case of CPPDd involving the TMJ region and summarize the findings of CPPDd reported in the literature.

2. Case report

A 54-year-old woman was referred to our hospital with the complaint of unpainful swelling in the right TMJ region. She first noticed discomfort in the TMJ 3 years previously and the swelling increased in size with time. There was no history of trauma in the orofacial region and no family history of joint disease.

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Fig. 1. The preoperative facial appearance. The arrows indicate extrusion of the right TMJ region.

Physical examinations showed facial asymmetry with localized swelling in the right TMJ region (Fig. 1). On palpation, the swelling was a 50 × 40-mm, tender, hard, immobile mass. The patient had a slight inability to open her mouth fully and could achieve a maximal mouth opening of 30 mm without deviation or ankyloses. There was no malocclusion by intraoral examinations.

Preoperatively, panoramic radiography showed large radiopaque lesions around the right TMJ region (Fig. 2); Computed tomography (CT) showed well-defined 46 × 42 × 37-mm and 27 × 18 × 12-mm calcified masses on the outside and inside of the mandibular condyle, respectively. The outside mass did not connect with the inside mass (Fig. 3A, B, C); Magnetic resonance imaging (MRI) also showed well-defined low-intensity masses in the joint space in the same locations (Fig. 3D) as the CT findings.

From the clinical and imaging findings, the provisional diagnoses were a tumor and osteoarthritis in the TMJ region. A biopsy was performed with the patient under general anesthesia to establish a definitive diagnosis. Macroscopically, the excised lesions appeared as white, gritty, chalk-like masses. Microscopically, hematoxylin-eosin staining of the specimen showed a great deal of crystalline materials (Fig. 4A) with positive birefringence by polarized light (Fig. 4B).

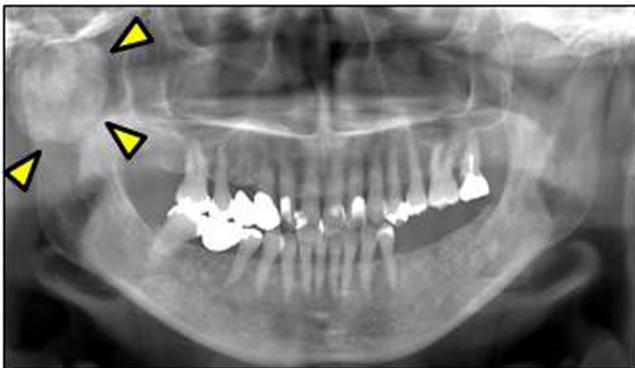


Fig. 2. A panoramic radiograph shows large radiopaque masses (arrows) in the right TMJ region.

Scanning electron microscopy (SEM) showed numerous rod-shaped and tube-like crystals (Fig. 5). Based on the clinical, radiologic, and histologic findings, a diagnosis of CPPDd was established. Preoperatively, a neurosurgical consultation was sought for advice on treating the mass inside the mandibular condyle, because it extended to the skull base. Although the neurosurgeon advised total surgical treatment of the mass, the patient elected to not remove the inside mass to avoid neurologic complications.

A partial excision under general anesthesia was performed to achieve symptomatic relief (Fig. 6A). Only the outside mass, which was a large amount of similar white and gritty material, was surgically separated from the mandibular condyle (Fig. 6B). To confirm the composition in the specimens, X-ray analysis showed diffraction of the crystals, with nearly all peaks corresponding to those of CPPDd (Fig. 7). The clinical outcome was satisfactory without postoperative complications (Fig. 8A). Postoperative CT showed that the outside mass of the mandibular condyle was completely removed by the surgery. Although the size of inside mass of the mandibular condyle was no change compared with preoperative CT, the neurosurgeons recommended a long-term follow-up of the patient for monitoring the growth of the mass to prevent severe neurologic complications (Fig. 8B,C).

3. Discussion

CPPDd has been classified etiologically into four categories: hereditary, sporadic, associated with metabolic disease, and associated with trauma or surgeries [9]. The current case likely was sporadic, because the patient had no relevant medical history. The development of CPPDd in the TMJ region has various clinical symptoms, such as sharp pain, inflammatory swelling, limited joint mobility, restricted mouth opening, abnormal occlusion, and deviation of the mandible due to the crystalline mass [2]. The clinical symptoms resemble other joint diseases with clinical signs similar to those of CPPDd, i.e., gout, osteoarthritis, suppurative arthritis, and rheumatoid arthritis [10], making it difficult to establish a diagnosis based on the clinical symptoms alone.

The provisional diagnosis of CPPDd is based mainly on imaging of the joint and periarticular calcifications and pyrophosphate arthropathy [11]. CT and MRI provide useful assessments of the extension of the lesions into the deeper structures when diagnosing and evaluating the TMJ. In the current case, CT showed large calcified masses around the right TMJ region, and the masses had slight continuity with the mandibular condyle and the skull base. MRI was particularly reliable for obtaining detailed information about the lesions, including the relationship between the lesions and the surrounding tissues. CT and MRI were useful for determining the most advantageous surgical approach, because they pinpointed the accurate locations and sizes of the masses. However, the radiographic features of CPPDd of the TMJ region are non-specific. It was impossible to completely rule out other diseases and provide a definitive preoperative diagnosis by these examinations. Because some cases have been reported in which the preoperative diagnosis was a possible malignancy, i.e., osteosarcoma and bone metastasis [6], a biopsy and histopathological study are recommended to establish the diagnosis. In the current case, a biopsy of the lesions was performed under general anesthesia to obtain a definitive diagnosis.

Microscopically, a definitive diagnosis of CPPDd requires confirmation of the presence of rhomboidal crystals on polarized light microscopy [12]. We used polarizing microscopy to confirm these crystals, which were strongly birefringent. The histopathological examination

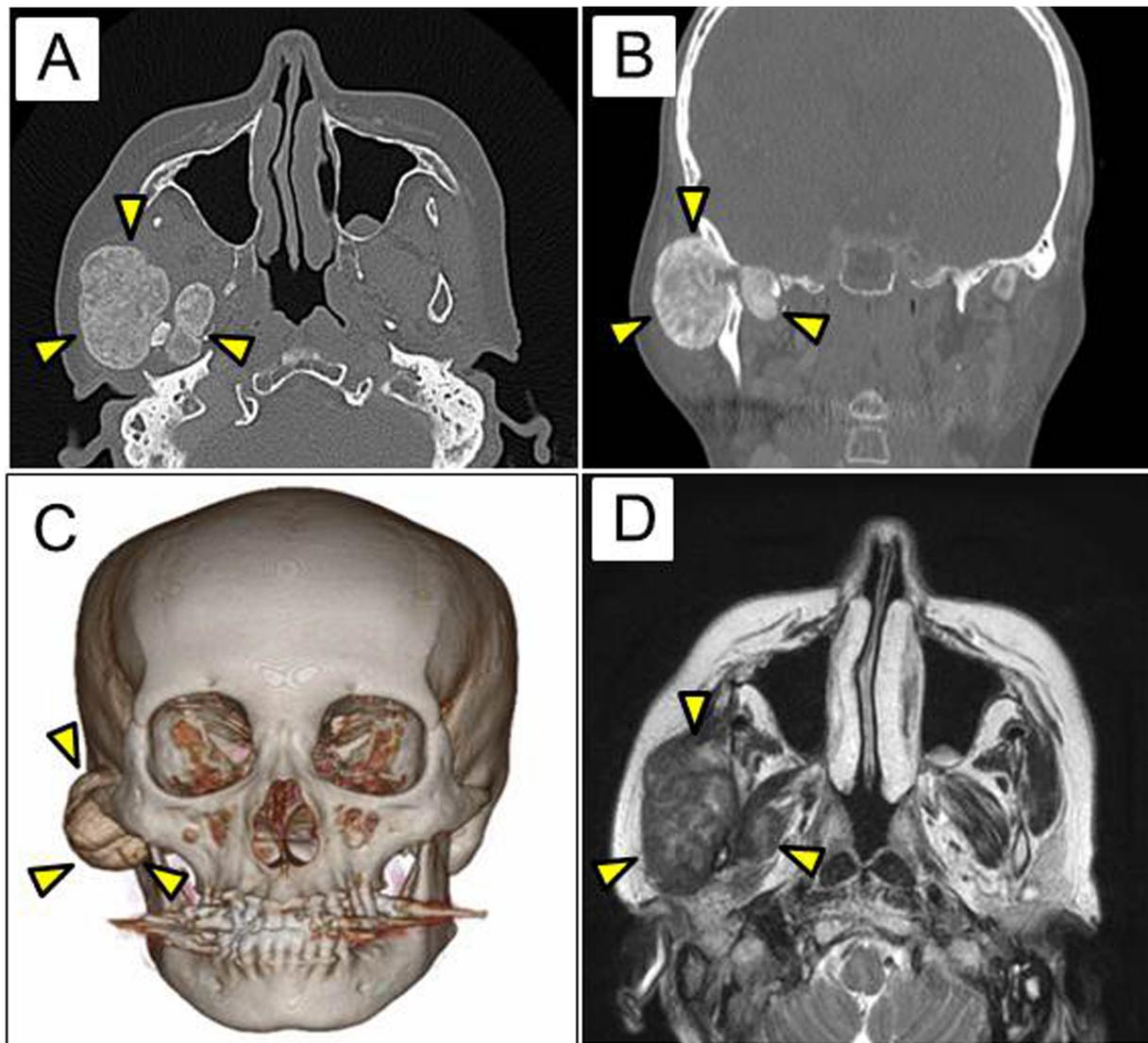


Fig. 3. (A) A preoperative axial CT image. The masses (arrows) are on the outside and inside mandibular condyle. (B) A preoperative coronal CT image. Both masses (arrows) of the mandibular condyle extend to the skull base. (C) The preoperative three-dimensional CT findings. The arrows indicate the mass on the outside of the mandibular condyle. (D) A preoperative axial MR image (T1- weighted image). The masses (arrows) are observed in the same region as on the CT images.

strongly suggested CPPDd in the right TMJ region. However, other crystals, such as synthetic steroids, calcium oxalate, and ethylenediaminetetraacetic acid, also can be birefringent under polarized light [10]. The differential diagnosis should be based on the observation of the crystalline structure or the qualitative and chemical analyses of the crystals. Using SEM, we determined the composition of the crystalline masses, which had numerous rod-shaped and tube-like crystals. Furthermore, the X-ray analysis showed calcium and phosphorus in the specimens. X-ray diffraction was the most dependable of the various analytical methods, because it facilitated identification of many types of crystals. In the current case, we were able to differentiate CPPDd from other diseases, because the peaks corresponded with those of calcium pyrophosphate dihydrate (Fig. 7).

To the best of our knowledge, 49 cases of CPPDd in the TMJ region have been reported [2,6,8,10–45]. The clinical features of the previously published cases and the current case are summarized in Table 1. According to those reports, patients with CPPDd in the TMJ region were typically older adults (35–85 years; mean age, 58.6 years) with a female predilection (male:female, 1:1.5). Regarding the side of the occurrence, lesions on the right side developed in 20 cases and on the left side in 25 cases. Almost all cases were unilateral except for five cases (2 cases were bilateral, and 3 cases were not recorded). The CPPDd was treated surgically in 43 cases (86.0%) of the 50 cases, conservatively in six (12.0%) cases, and untreated in one (2.0%) case. Therefore, according to the review of the previous reports and including the current case, surgical treatment of the CPPDd in the TMJ region has been the primary

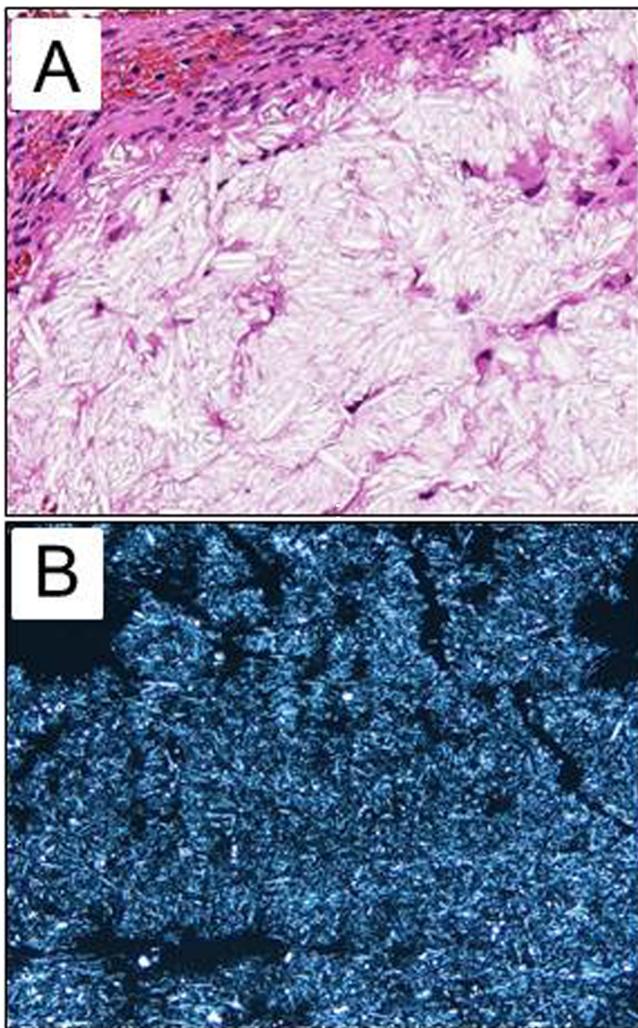


Fig. 4. (A) The histopathologic examination shows abundant crystal depositions (hematoxylin–eosin, x 400). (B) The crystalline deposits have positive birefringence under polarized light (x400).

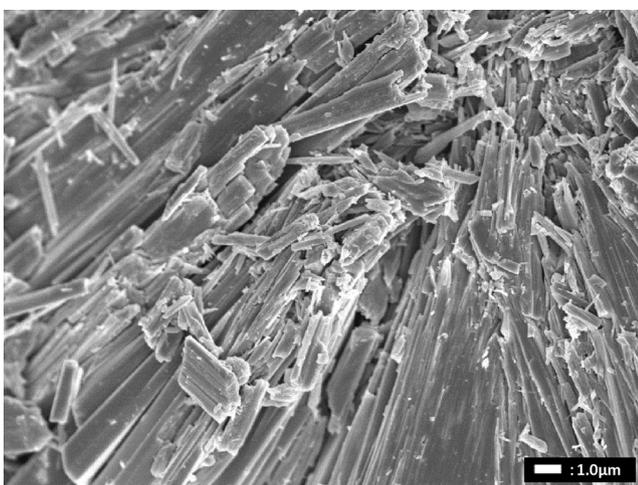


Fig. 5. SEM shows a cluster of tube-like crystals (x5,000). Scale bar, 1.0 μm .

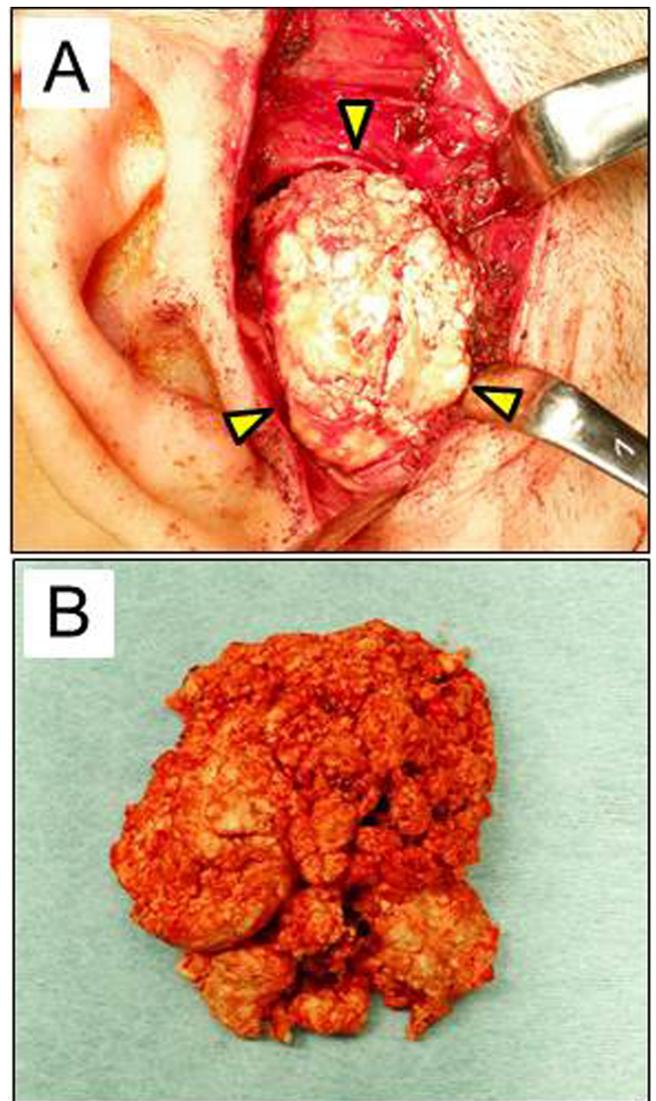


Fig. 6. (A) The intraoperative view of the mass on the outside of the mandibular condyle shows deposition of a chalky calcified material (arrows). (B) A photograph of the postoperative specimen.

approach. The clinical features of the current case were consistent with those of the previous reports.

The treatment of CPPD in the TMJ region varies depending on the extent of the clinical manifestations and symptoms. Various treatment options have been discussed in the literature, including surgery, medication, and conservative therapy [10]. Non-steroidal anti-inflammatory medications have been used to treat the acute symptoms in CPPD of the TMJ region. Aspirin, steroids, and colchicine help alleviate the acute arthritis [24]. Surgical excision of the mass, the main treatment of CPPD in the TMJ region, restores joint function but can result in severe neurologic dysfunction. In the current case, a partial excision was performed via the preauricular approach with the patient under general anesthesia to prevent damaging the cranial nerves. The patient was satisfied with the improved TMJ movement and her postoperative facial symmetry without development of any complications during 10 months follow-up.

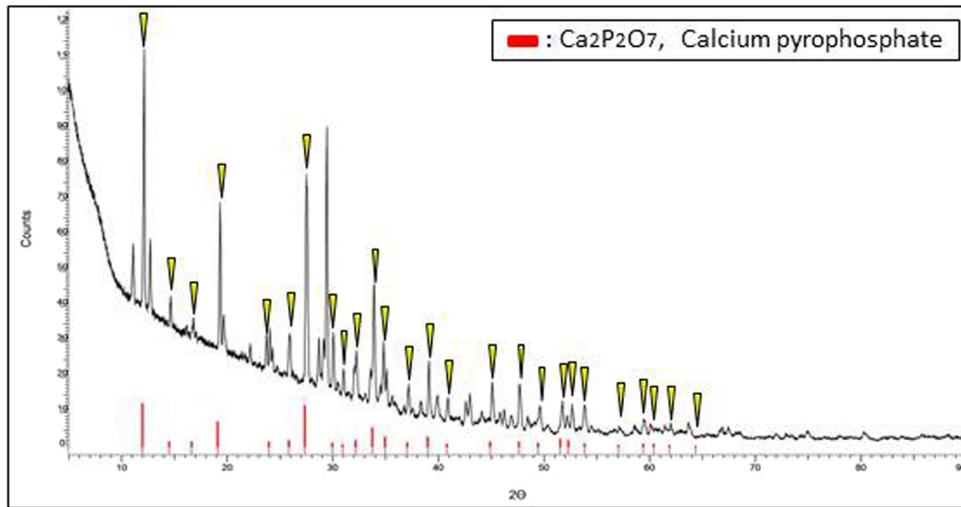


Fig. 7. The X-ray diffraction pattern. The peaks (arrows) correspond to those of calcium pyrophosphate dihydrate.

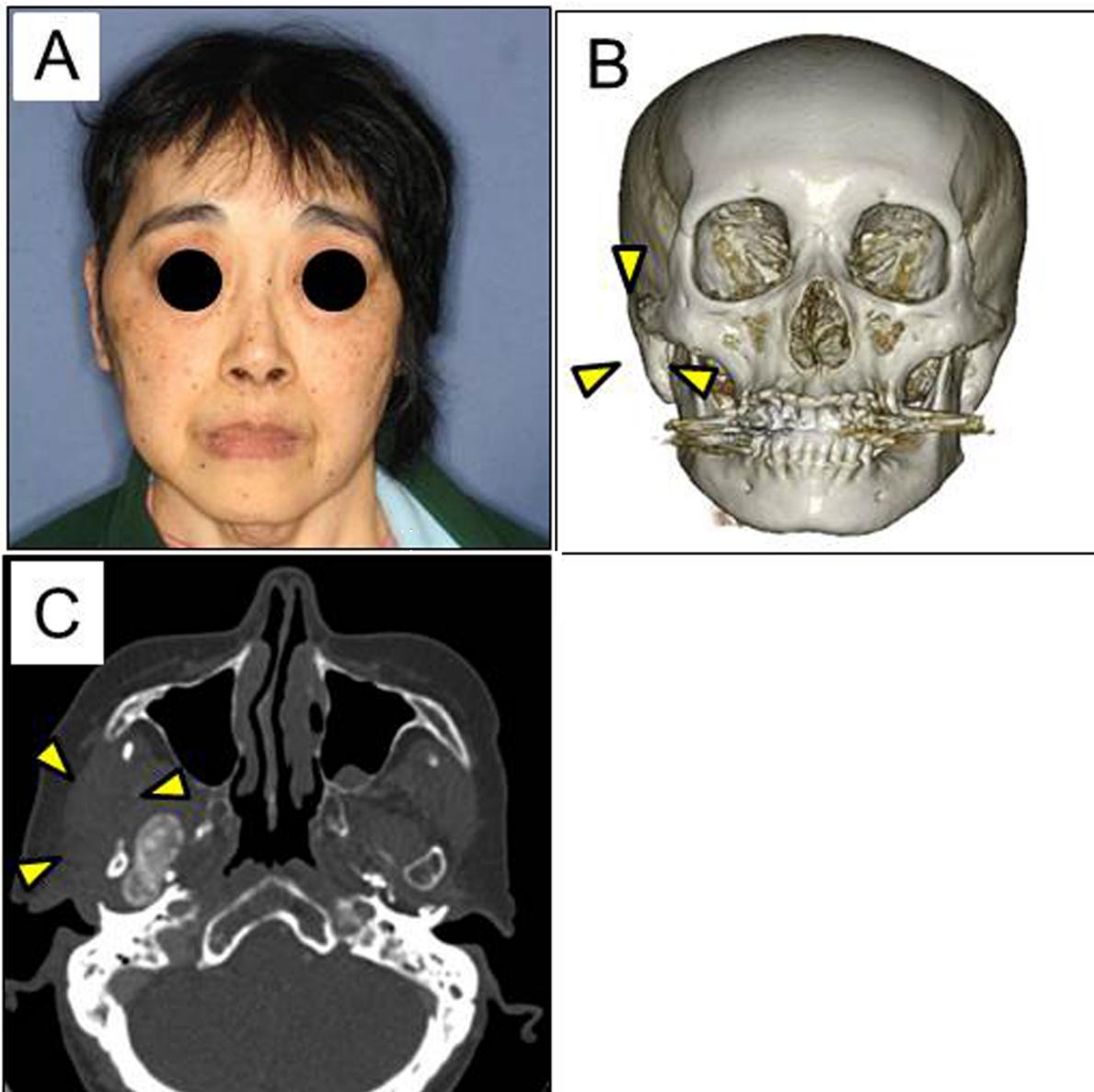


Fig. 8. (A) The postoperative facial appearance. (B) A postoperative three-dimensional CT findings. (C) A postoperative axial CT image. Only the outside masses (arrows) are removed.

Table 1
Summary of cases of previously reported CPPD.

Case	Age	Gender	Side	Treatment	Year	Study
1	55	Male	Right	Excision	1976	Pritzker et al. [5]
2	51	Female	Left	Excision	1981	de Vos et al. [13]
3	56	Male	Left	Medication	1982	Good and Upton [14]
4	51	Female	Left	Excision	1985	Zemplenyi and Calcaterra [15]
5	57	Male	Left	Excision	1987	Kamatani et al. [16]
6	59	Female	Left	Excision	1987	Gross et al. [17]
7	54	Female	Right	Excision	1987	Mogi et al. [18]
8	78	Female	Right	None	1987	Hutton et al. [19]
9	76	Female	Right	Medication	1987	Hutton et al. [19]
10	68	Female	Right	Medication	1987	Hutton et al. [19]
11	41	Male	Right	Excision	1990	Lambert et al. [20]
12	53	Female	Left	Excision	1992	Magno et al. [21]
13	53	Female	Left	Excision	1995	Dijkgraaf et al. [22]
14	65	Female	Bilateral	Excision	1995	Chuong and Piper [23]
15	58	Male	Left	Excision	1995	Pynn et al. [24]
16	47	Female	–	Excision	1995	Ishida et al. [25]
17	50	Female	–	Excision	1995	Ishida et al. [25]
18	55	Female	–	Excision	1995	Ishida et al. [25]
19	48	Female	Left	Excision	1997	Onodera et al. [26]
20	85	Male	Right	Excision	1997	Kurihara et al. [27]
21	80	Male	Right	Excision	1998	Jordan et al. [28]
22	51	Female	Left	Excision	1998	Strobl et al. [29]
23	63	Female	Left	Excision	1999	Goudot et al. [30]
24	60	Female	Right	Excision	1999	Nakagawa et al. [8]
25	45	Female	Left	Medication	1999	Nakagawa et al. [8]
26	76	Male	Right	Irrigation	1999	Nakagawa et al. [8]
27	45	Female	Left	Excision	2000	Aoyama et al. [10]
28	72	Male	Right	Excision	2001	Eriksson et al. [12]
29	51	Female	Left	Excision	2001	Olin et al. [31]
30	56	Male	Bilateral	Medication	2002	Greaves and Fordyce [32]
31	68	Female	Right	Excision	2002	Cottrel et al. [33]
32	40	Male	Left	Excision	2003	Osano et al. [34]
33	70	Male	Right	Biopsy	2004	Marsot-Dupuch et al. [35]
34	53	Female	Left	Biopsy	2004	Marsot-Dupuch et al. [35]
35	57	Male	Right	Medication	2004	Goldblatt et al. [36]
36	44	Female	Left	Excision	2004	Dimitroulis [37]
37	74	Female	Left	Excision	2005	Smolka et al. [38]
38	64	Male	Left	Excision	2006	Cascone et al. [39]
39	35	Male	Left	Excision	2008	Naqvi et al. [40]
40	52	Female	Left	Excision	2008	Reynolds et al. [41]
41	72	Female	Right	Excision	2008	Ascani et al. [42]
42	71	Female	Left	Excision	2010	Kalish et al. [43]
43	64	Female	Left	Excision	2011	Meng et al. [6]
44	46	Male	Right	Excision	2012	Matsumura et al. [44]
45	62	Male	Right	Excision	2013	Lv et al. [11]
46	60	Male	Left	Excision	2014	Abdelsayed et al. [2]
47	75	Male	Right	Excision	2014	Abdelsayed et al. [2]
48	74	Female	Left	Excision	2014	Abdelsayed et al. [2]
49	38	Male	Right	Excision	2017	Kudoh et al. [45]
50	54	Female	Right	Excision	2018	Current case

4. Conclusion

We describe a rare case of CPPD in the TMJ that had a good outcome as the result of appropriate examinations, diagnosis, and treatments.

Ethical approval

The Institutional Review Board reviewed and approved this study.

Conflict of interest

No authors have any conflicts of interest to report.

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