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Abstract: Degenerative changes of the temporomandibular joint (TMJ) present with a broad spectrum of morphological alterations. However, erosions leading to a glenoid fossa defect and condylar impingement of the temporal lobe are a rare finding. A 77-year-old female patient presented with limited mouth opening and pre-auricular pain during mastication on the left side. She denied any neurological dysfunction. Her medical history included poliomyelitis, multiple cancers, and osteonecrosis of the left tibial plateau. Computed tomography revealed advanced degeneration of both TMJs. On the left side, a glenoid fossa fragment was elevated towards the left middle cranial fossa. Real-time dynamic magnetic resonance imaging (MRI) showed repetitive intracranial condylar dislocation during mouth closure. She declined surgery and received instructions for self-management. At the 12-month follow-up, she reported resolution of the pain and normal masticatory function. A control MRI showed a stable radiographic appearance. This report illustrates that intermittent dislocation of the mandibular condyle into the middle cranial fossa can be successfully managed conservatively. The self-limiting nature of the TMJ degenerative joint disease, patient preference, and the patient's general health status require consideration when advising patients on the therapeutic strategy.

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Intermittent intracranial condylar dislocation with minimal functional sequelae

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Abstract. Degenerative changes of the temporomandibular joint (TMJ) present with a broad spectrum of morphological alterations. However, erosions leading to a glenoid fossa defect and condylar impingement of the temporal lobe are a rare finding. A 77-year-old female patient presented with limited mouth opening and pre-auricular pain during mastication on the left side. She denied any neurological dysfunction. Her medical history included poliomyelitis, multiple cancers, and osteonecrosis of the left tibial plateau. Computed tomography revealed advanced degeneration of both TMJs. On the left side, a glenoid fossa fragment was elevated towards the left middle cranial fossa. Real-time dynamic magnetic resonance imaging (MRI) showed repetitive intracranial condylar dislocation during mouth closure. She declined surgery and received instructions for self-management. At the 12-month follow-up, she reported resolution of the pain and normal masticatory function. A control MRI showed a stable radiographic appearance. This report illustrates that intermittent dislocation of the mandibular condyle into the middle cranial fossa can be successfully managed conservatively. The self-limiting nature of the TMJ degenerative joint disease, patient preference, and the patient's general health status require consideration when advising patients on the therapeutic strategy.

Key words: temporomandibular joint disorders; joint diseases; diphosphonates; middle cranial fossa; skull base.

Accepted for publication

The temporomandibular joints (TMJs) are located in direct proximity to vital structures such as cranial nerves V and VII (auriculotemporal nerve), branches of the internal maxillary artery (most important—the middle meningeal artery) and

superficial temporal artery, the parotid gland, and external auditory canal (EAC). The mandibular condyle is separated from the brain by a thin bony layer forming the glenoid fossa, which measures between 0.6 mm and 0.85 mm in healthy

joints¹. The roof of the glenoid fossa belongs to the floor of the middle cranial fossa, with the inferior temporal gyrus abutting the TMJ and dura. The anterior border of the glenoid fossa forms the articular tubercle, which is covered with

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fibrous cartilage and articulates with the mandibular condyle. The tympanic plate bounds the glenoid fossa posteriorly and separates it from the EAC.

Degenerative joint disease (DJD) of the TMJ is common². Since the terms osteoarthritis and osteoarthritis are inconsistently applied in medicine, use of the term DJD is currently endorsed by international consensus³. Joint space narrowing and erosion of the articular surfaces are early imaging criteria of DJD⁴.

A rare case of advanced glenoid fossa erosion that resulted in intermittent unilateral intracranial condylar dislocation is presented here.

Case presentation

A 77-year-old female presented to the Orofacial Pain Unit, University of Zurich, complaining of limited mouth opening and pre-auricular pain during mastication, predominantly on the left side. Her symptoms had developed spontaneously, with gradual progression over approximately 2 years, and had persisted despite multiple physical therapy sessions. She denied any history of trauma and her cognitive state was excellent. There were no signs or symptoms of any neurological dysfunction other than residual paresis of the right leg due to poliomyelitis in early childhood.

Multiple cancers (uterus, breast, and colon) had been diagnosed and managed successfully during the preceding 10 years. In the wake of 10 years of diphosphonate therapy for metastatic breast cancer, she had developed osteonecrosis of the left tibial plateau, leading to discontinuation of this medication at the time of initial presentation to the clinic.

Clinical examination revealed tenderness upon palpation of the left TMJ, whereas the right joint was painless. The masticatory muscles were mildly tender. Her edentulous jaws had been rehabilitated with full upper and lower dentures. Maximum mouth opening was subjectively unrestricted and the distance between the edentulous alveolar ridges measured 40 mm.

Computed tomography (CT) revealed bilateral advanced degenerative condylar changes with flattening of the articular surface, anterior directed osteophytes and sclerosis, and elevation of a glenoid fossa fragment towards the middle cranial fossa on the left side (Fig. 1). Real-time dynamic magnetic resonance imaging (MRI) confirmed an osseous defect of the skull base with intermittent intracranial dislocation of the left condyle upon jaw

closing (Supplementary Material Video 1). Metastatic jaw lesions were excluded.

Based on clinical and radiographic findings, a diagnosis of advanced TMJ DJD was postulated. The fact that the patient had received diphosphonate injections for approximately 10 years raised the hypothesis that diphosphonates might have contributed to the unusual glenoid fossa bony defect.

The patient declined surgery and received instructions for self-management focusing on conscious relaxation of the masticatory muscles, topical non-steroidal anti-inflammatory drugs, and avoidance of hard food in order to reduce the mechanical loading of the joint. In particular, the patient was instructed to avoid parafunctions such as teeth clenching during the daytime. Upon re-examination after 3, 6, and 12 months, the patient reported resolution of the pain,

normal masticatory function, and an absence of neurological symptoms. A control MRI performed at the 12-month follow-up showed no disease progression (Supplementary Material Video 2; Fig. 2) and the patient was satisfied with the conservative management strategy.

Discussion

It is widely recognized that the degree of TMJ DJD as depicted by imaging often does not correlate with the presence and intensity of joint pain unless an additional inflammatory process is initiated, sometimes referred to as 'activated osteoarthritis'². Consequently, TMJ pain (e.g. arthralgia) has been qualified as a separate diagnostic entity that differs from DJD³. Furthermore, arthralgia associated with TMJ DJD is characteristically

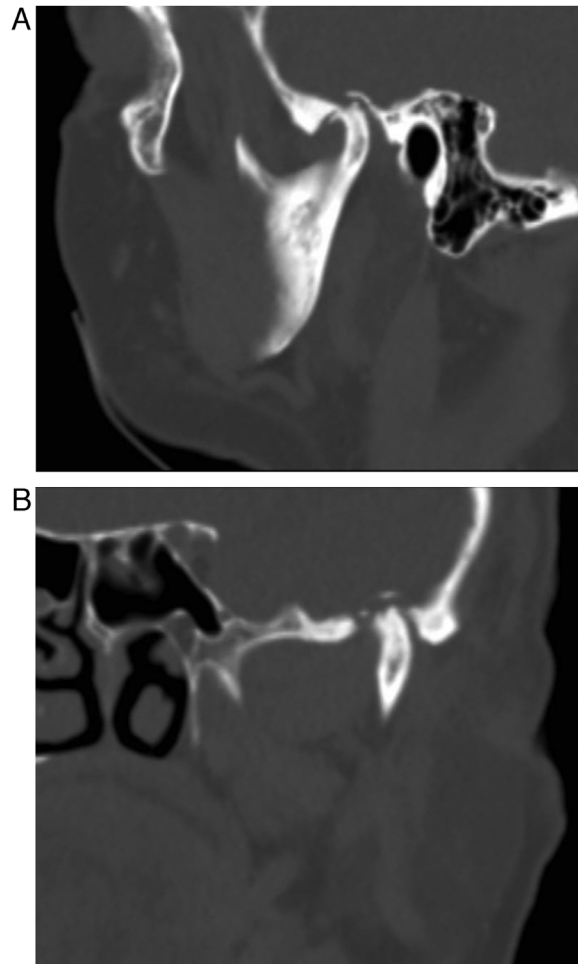


Fig. 1. Computed tomography of the left temporomandibular joint depicting advanced degenerative joint disease. (A) Sagittal view: flattening of the articular surface, anteriorly directed osteophyte, and sclerosis, as well as a vertical intracranial displacement of the condyle in conjunction with an elevated bony shell. (B) Coronal view: elevation of a glenoid fossa fragment and left condyle towards the middle cranial fossa.

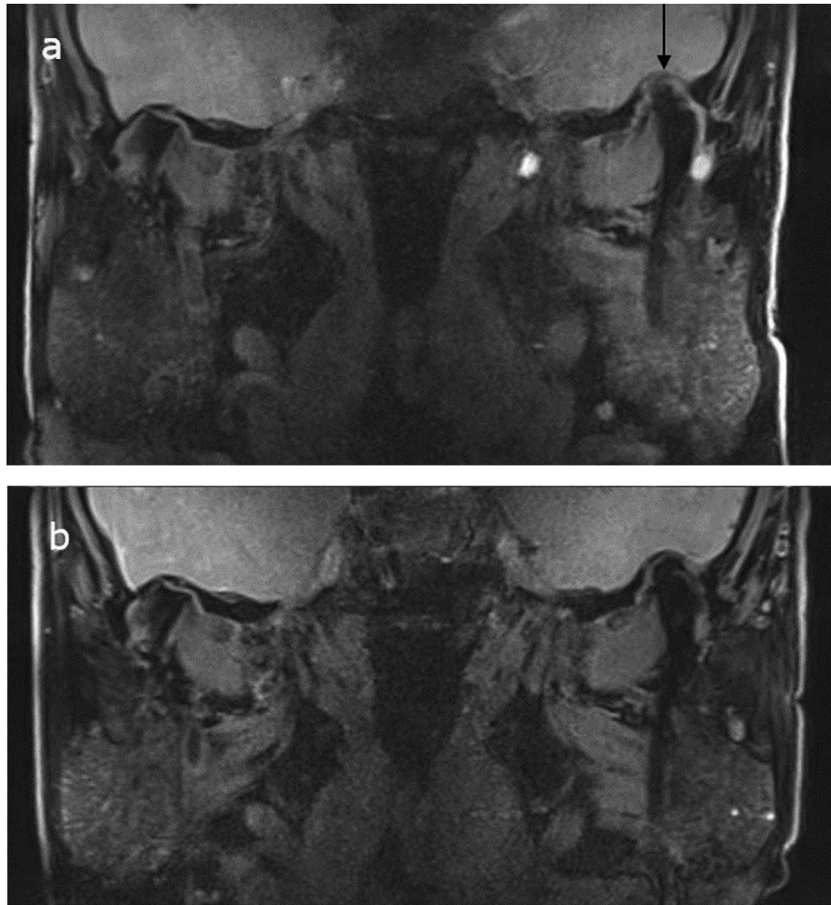


Fig. 2. Coronal DESS (Double Echo Steady State) MR sequence displays a difference in height of the upper condylar contour due to intracranial subluxation on the left while the condylar shape is well maintained as opposed to the flattened right side. Marked indentation of the floor of the left middle cranial fossa (arrow): (A) at initial presentation; (B) 12-month follow-up.

self-limiting. Elderly patients are unlikely to report TMJ pain in spite of marked morphological degenerative changes⁵. This observation is possibly related to the weakening of masticatory muscles in those of advanced age and the associated reduction in joint load. Hence, the primary therapeutic goal in this case was to reduce the pain and not the recovery of the radiological changes. Information therapy (self-management instructions, masticatory muscle relaxation training), topical non-steroidal anti-inflammatory drugs, and advice to avoid hard food proved to be sufficient to alleviate the pain and keep the radiological findings stable.

DJD of the TMJ is usually associated with thickening of the glenoid fossa¹. We are aware of only one reported case of glenoid fossa perforation due to erosive TMJ disease⁶. The most common causes of condylar dislocation into the middle cranial fossa are trauma and surgical complications⁷. Under such circumstances,

characteristic symptoms are pre-auricular pain, limited mouth opening, and malocclusion with an anterior open bite, as well as facial paralysis due to facial nerve damage. Complications such as epidural or subdural haemorrhage and cerebrospinal fluid leaks require surgical management. Therapy of an intracranial condylar dislocation may include closed or open treatment strategies. The latter may consist of surgical reduction, condylotomy, condylectomy, and TMJ reconstruction with alloplastic implants⁸. Independent of the surgical treatment modality, persistent mandibular deviation during opening is a common sequela, yet this is mostly functionally irrelevant to patients. Rare complications include facial paralysis, hearing loss, cerebral contusion, and pneumocephalus. Our patient was not willing to undergo a surgical procedure due to her advanced age and compromised general health. Hence, a conservative treatment approach was pursued.

This patient was at high risk of developing medication-related osteonecrosis of the jaw (MRONJ). The risk factors were female sex, age, long-term therapy with alendronate, spontaneous occurrence of pain, and comorbid hypertension⁹. A potential association of diphosphonates with TMJ ankylosis has been postulated previously¹⁰. The dense, well-maintained bone contours may support the effect of anti-resorptive agents as a contributory factor in this case. However, it appears that no cases of MRONJ with isolated TMJ involvement have been published.

Since the patient had suffered from several cancers in the past, a further differential diagnosis included neoplastic destruction of the glenoid fossa. However, bilateral TMJ involvement, the smooth contour of the widened glenoid fossa, dense bone structure, and elevated bone fragment are in favour of a degenerative fossa destruction.

There are multiple possible explanations for the positive clinical outcome in this case. Conceivable contributory factors included, among others, cessation of the antiresorptive medication, reduction of mechanical loading, and the natural, self-limiting course of TMJ DJD. We have no definitive explanation for the long-lasting symptom resolution by conservative management and hope that this case may open a discussion of what readers may speculate.

This report illustrates that intermittent dislocation of the mandibular condyle into the middle cranial fossa can be managed conservatively without adverse long-term sequelae. When advising patients on the appropriate therapeutic strategy, the self-limiting nature of TMJ DJD, patient preference, and the patient's general health status require consideration.

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Competing interests

The authors declare that they have no competing interests.

Ethical approval

In Switzerland, ethical approval is required for research projects that are subject to the Human Research Act. Individual case reports are not defined as research projects, as their results cannot be generalized, and therefore do not

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require an approval. Hence, for this case report, no ethical approval was required.

Patient consent

Written consent for publication was obtained from the patient.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:<https://doi.org/10.1016/j.ijom.2020.09.007>.

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