

## IMAGING HIGHLIGHTS

# Bilateral temporo-mandibular joint dislocation due to complication of oro-facial dyskinesia

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### Abstract

Oro-facial dyskinesia (OFD) is involuntary, abnormal, uncontrolled and stereotyped movements, consisting of forehead frowning, eye opening and closing, smacking and pursing of the lips, lateral deviation and protrusion of the tongue, and occasionally lateral deviation and protrusion of the jaw.<sup>1</sup> OFD is known to have various complications including speech difficulty, chewing and eating disorders, and social embarrassment; facial muscle stiffness, mucosal and gingival traumatic lesions. In addition, it may lead to cranio-mandibular joint (TMJ) complications in the presence of intense and prolonged abnormal movements, with pain and degeneration.<sup>1,2</sup> There is no previous report of TMJ dislocation due to OFD. In this report, we describe a patient who developed bilateral anterior TMJ dislocation due to OFD which occurred following intra-cranial hemorrhage (ICH).

### CASE REPORT

A 68-year-old man underwent surgical management (decompressive occipital craniectomy and hematoma removal, and extra-ventricular drainage) for spontaneous ICH in the cerebellum and intraventricular hemorrhage (Figure 1A). Ten weeks after the onset of ICH, he was transferred to the rehabilitation department of our university hospital. His chief complaints were quadriplegia, and constant and uncontrolled OFD consisting of pursing and lateral deviation of the lips. The patient did not have history of taking any medications that may cause dyskinesia. He underwent a comprehensive rehabilitative management program including medications (amantadine 200mg/day, levetracetam 1500mg/day, clonazepam 1.5mg/day, and pyridoxine 100mg/day). After one month of rehabilitation, the OFD symptoms improved by about 80 percent, and he was discharged to a local medical rehabilitation center. When he was admitted to the local medical rehabilitation center, the medications for OFD were changed as follows: clonazepam 2mg/day, quetiapine 125mg/day. At 4 months after the onset of ICH, he was admitted to the cardiology department at our university for stress induced cardiomyopathy, and the patient had worsened symptoms of OFD. After management

of cardiomyopathy, he was discharged to another local medical center. The medications to control OFD were reduced to quetiapine 100mg/day. Six months after onset of ICH, he was re-admitted to the rehabilitation department of our university hospital for the management of the worsened OFD. His lips were persistently pursed, forehead was continuously in furrow, and his tongue and jaw had intermittent lateral deviation and protrusion. He was very anxious. On physical examination, there was prominent right pre-auricular protuberance and the patient could not close his mouth. There was no history of TMJ trauma after previous discharge from our hospital. Three dimensional computed tomography showed bilateral anteriorly displaced mandibular condyles well beyond the articular eminences. (Figure 1B)

### DISCUSSION

Dislocation of the TMJ represents three percent of all reported dislocated joints. The majority of the cases are non-traumatic and is often precipitated by yawning, eating, dental treatment, endoscopy, or oral intubation. We believe that the uncontrolled OFD during the 6 months period after the onset of ICH was the most probable cause of bilateral TMJ dislocation in this patient, because he had no previous history of TMJ problem and we

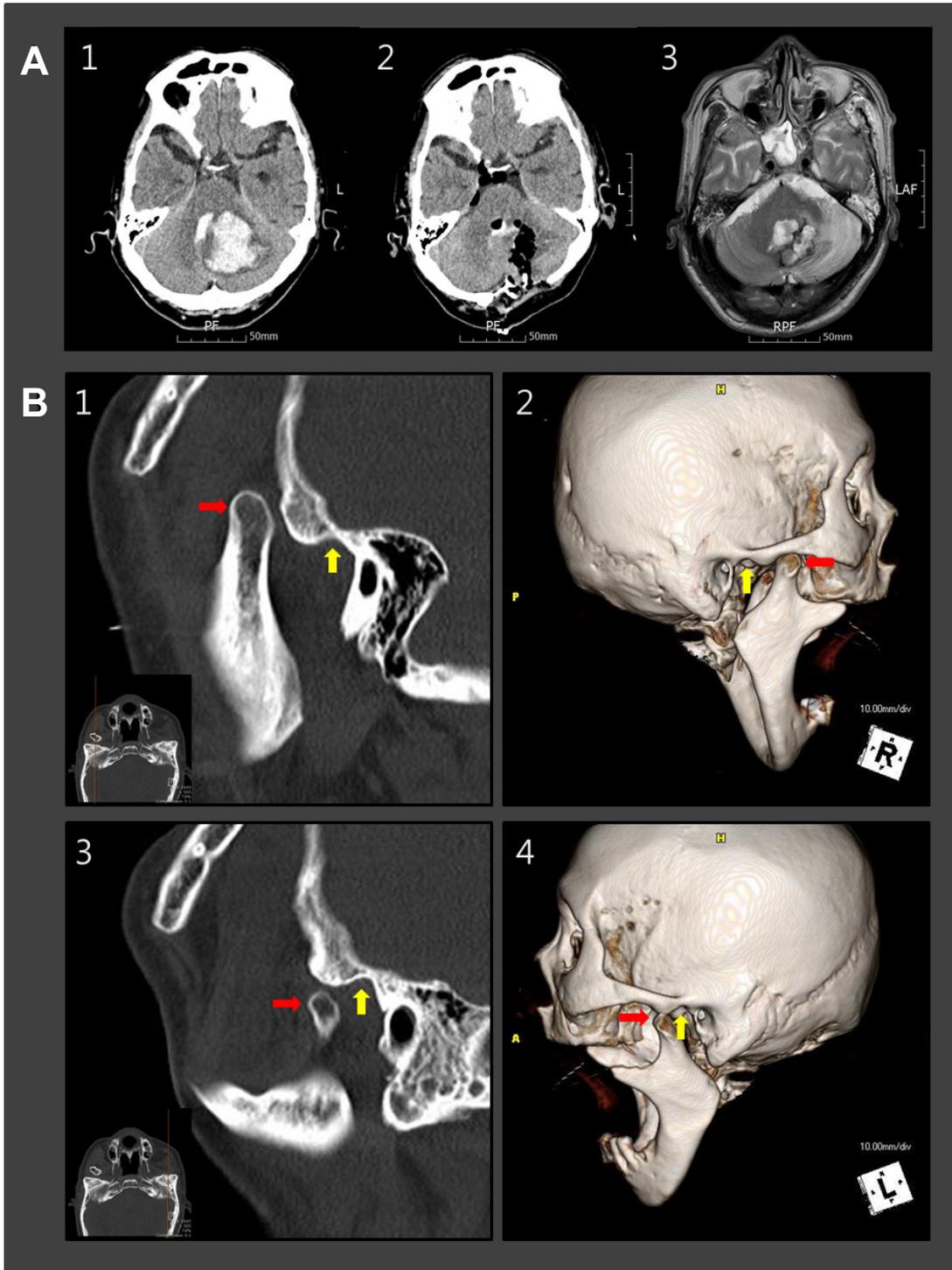


Figure 1. (A) Brain computed tomography scan at onset showing hemorrhage in the cerebellum and fourth ventricle (1), postoperative computed tomography scan after decompressive occipital craniectomy and hematoma removal (2), and T2-weighted MRI image at 71 days after hemorrhage in the cerebellum and fourth ventricle (3). (B) Anteriorly dislocated right mandibular condyle (red arrow) in sagittal computed tomography (CT) image (1), and in 3 dimensional reconstructed CT image (2). Anteriorly dislocated left mandibular condyle (red arrow) in sagittal CT image (3), and in 3 dimensional reconstructed CT image (4). Yellow arrow indicates proper position of mandibular condyle.

could not find any other possible causes for TMJ dislocation except for OFD. With aging population and increased use of polypharmacy, some of which can induce OFD, there is likely to be increased incidence of OFD. It is also common for OFD to be inadequately treated resulting in various complications.<sup>1</sup>

As for the previous complications of TMJ from OFD, in 1989, Osborn *et al.* reported a patient who developed severe degenerative joint disease of both TMJ after mandible dyskinesia.<sup>2</sup> In 2010, Pekkan *et al.* reported a patient with masticatory muscle pain and TMJ osteoarthritis due to dyskinesia.<sup>3</sup> Although there has been several reports of TMJ dislocation as a complication of oro-facial dystonia after using neuroleptic drugs such as haloperidol, benztropine and aripiprazole<sup>4-6</sup>, to the best of our knowledge, this is the first report of TMJ dislocation due to OFD.

In conclusion, we report a patient who had bilateral anterior TMJ dislocation due to OFD following ICH. This case demonstrate that clinicians should try to control OFD more aggressively to prevent serious complication such as TMJ dislocation.

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