



Received:
9 March 2016
Accepted:
7 June 2016
Disponible en ligne
26 July 2016



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Temporomandibular joint ankylosis after condylar dislocation into the middle cranial fossa: A case report

Ankylose temporo-mandibulaire après luxation du condyle dans la fosse cérébrale moyenne : à propos d'un cas

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Summary

Introduction. Dislocation of the mandibular condyle into the middle cranial fossa after a trauma is a rare event. The lack of appropriate treatment can lead to ankylosis of the temporomandibular joint (TMJ). We report about a case of TMJ ankylosis following intracranial dislocation of the mandibular condyle through the roof of the articular fossa.

Case report. A 9-year-old patient was referred for a severe limitation of mouth opening that began progressively one year before. A history of chin injury due to an accidental fall was found. Preoperative CT scan showed a TMJ ankylosis on the right side combined with a dislocation of the mandibular condyle into the middle cranial fossa. Treatment consisted in an intracranial resection of the mandibular condyle, partial removal of the ankylosis block and TMJ arthroplasty.

Discussion. Our case is the second case of TMJ ankylosis following intracranial dislocation of the mandibular condyle and treated with arthroplasty alone published in the English literature. There is no consensus regarding the pathophysiology of TMJ ankylosis and regarding the attitude towards the ankylosis block. In our case, no

Résumé

Introduction. La luxation du condyle mandibulaire dans la fosse cérébrale moyenne après un traumatisme est rare. En l'absence de prise en charge initiale correcte, une ankylose de l'articulation temporo-mandibulaire (ATM) est possible. Nous présentons un cas d'ankylose temporo-mandibulaire survenue sur un condyle mandibulaire luxé dans la fosse cérébrale moyenne à travers le toit de la fosse articulaire.

Cas clinique. Un patient de 9 ans a consulté pour une limitation sévère de l'ouverture buccale ayant débuté progressivement un an auparavant. L'interrogatoire révèle la notion de traumatisme du menton à l'âge de 6 ans au décours d'une chute accidentelle. Le scanner réalisé dans le cadre du bilan préopératoire a montré l'existence d'une ankylose de l'ATM droite, combinée à une dislocation du condyle homolatéral dans la fosse cérébrale moyenne. Le traitement a consisté en une résection intracrânienne du condyle mandibulaire, associée à une exérèse partielle du bloc d'ankylose et à la création d'une néofosse articulaire.

Discussion. Il s'agit à ce jour du 2^e cas d'ankylose de l'ATM développé après luxation intracrânienne du condyle mandibulaire,

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recurrence was noticed after a one-year follow-up. An interdisciplinary approach is needed, including maxillofacial surgeon, neurosurgeon, physiotherapist and orthodontist.

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Keywords: Intracranial, Dislocation, Mandibular condyle, Middle cranial fossa, Temporomandibular joint ankylosis

traité par arthroplastie isolée et publié dans la littérature anglo-saxonne... Il n'y a pas de consensus sur la physiopathologie de l'ankylose de l'ATM ni sur la conduite à tenir par rapport au bloc d'ankylose. Dans notre cas, il n'y a pas eu de récurrence après un an de surveillance. Une approche interdisciplinaire est nécessaire. Elle inclut le chirurgien maxillofacial, le neurochirurgien, le physiothérapeute et l'orthodontiste.

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Mots clés : Intracrânienne, Luxation, Condyle mandibulaire, Fosse cérébrale moyenne, Ankylose tempo-mandibulaire

Introduction

Dislocation of the mandibular condyle into the middle cranial fossa is extremely rare [1,2]. Most of the time, it is caused by a traumatic impact of great force in the chin with the consequential perforation of the glenoid fossa roof. It can lead to serious complications such as middle meningeal artery damage, cerebrospinal leakage, extradural hematoma, or even meningitis. The clinical features are very similar to those found in a sub-condylar fracture such as decreased height of the ramus on the injured side, mandibular deviation to the ipsilateral side, mandibular function impairment or pain located in the preauricular area [2].

In some cases, there are no clinical signs or symptoms of middle cranial fossa damage and less often no clinical symptoms of temporomandibular joint dysfunction, which is why the patient does not seek medical care; or where proper complementary studies such CT scan or MRI are lacking, these kind of injuries may go undiagnosed [1]. Delayed diagnosis can lead to long-term temporomandibular joint ankylosis with a dislocated condyle in the middle cranial fossa. This last phenomenon has only been reported once in the English literature [3].

The case of a 9-year-old male patient suffering a temporomandibular joint ankylosis with mandibular condyle dislocated into the middle cranial fossa is reported. The treatment options and physiopathology of this injury are discussed.

Case report

A healthy 9-year-old male patient was referred to the San Juan de Dios Hospital Maxillofacial Service with a chief complaint of severe limitation of mouth opening. This limitation has gradually started a year before, but his parents sought medical care when he reached the point when he could not eat properly. His medical history revealed a chin blow due to an accidental fall when he was 6 years old. Because of the lack of clinical signs, severe symptoms or functional impairment at the time of the traumatic event, no emergency care consultation was requested.

On general physical examination, patient's height and weight were below average. Facial and oral examination showed a noticeable facial asymmetry with a mandibular deviation to the right side, micrognathia and excessive maxillary vertical growth with significant gingival exposure. Regarding the dynamics of the TMJ, left side lateral movements were abolished whereas the right side was diminished and a maximum mouth opening of 6 mm with mandibular deviation to the right side was observed (fig. 1).

The initial CT scan showed an anatomic osseous remodeling of the right condyle compared to its contralateral counterpart, showing an increase in size and irregular articular surface, an increase osteosclerosis and a reduced bone marrow cavity, a fracture of the glenoid fossa roof and an intrusion of the mandibular right condyle into the middle cranial fossa towards cranial, medial and anterior direction. There was a radiolucent peripheral area between the condyle and surrounding bone structures. These images were consistent with fibrous ankylosis and dislocation into the middle cranial fossa of the right mandibular condyle (figs. 2-4).

A surgery was performed. A flap of the temporal region extended into the preauricular region was performed. The vascular and the neurological structures have been identified (fig. 5). A craniotomy was performed exposing the middle cranial fossa and the mandibular condyle preserving the dura mater throughout the entire procedure.

A condylectomy was performed despite the absence of neurological signs to allow proper brain development. The floor of the middle cranial fossa floor was remodeled. The ankylosed joint was subsequently released. A new condyle was carved in the lower part of the condylar process. The mandibular fossa was remodeled and a temporal muscle flap was interposed. Finally, a bilateral coronoidectomy was performed using an intraoral approach. Maximum mouth opening of 25 mm was achieved in the operating room. The patient started with physiotherapy on the third day after surgery (fig. 6).

A postoperative CT scan showed evidence of condylectomy and coronoidectomy of the right mandibular side and coronoidectomy of the left mandibular side. There were signs of

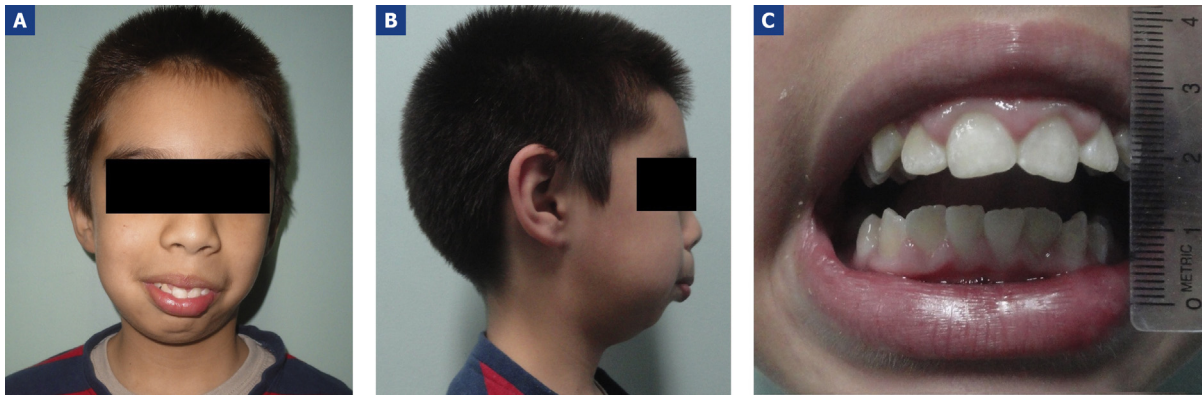


Figure 1. Patient at the moment of first examination; **A:** frontal view with closed mouth showing lip incompetence; **B:** lateral view showing micrognathia and lower lip eversion; **C:** frontal view of maximum mouth opening of 6 mm and mandibular deviation to the right side.

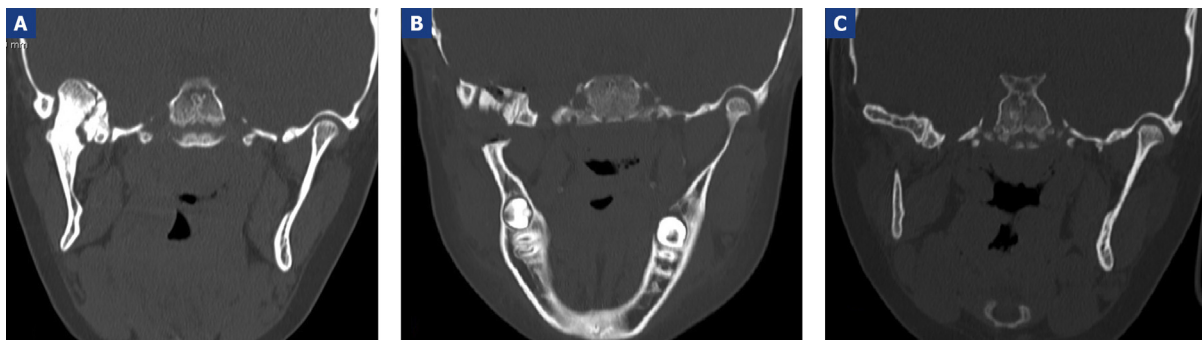


Figure 2. CT scan, frontal view; **A:** before surgery scan, showing an increased size and irregular anatomy of the right mandibular condyle compared to the left; **B:** after surgery CT scan, showing removal of the deformed condyle intruded in the middle cranial fossa with ankylosed mass remnant as a new roof of the right glenoid fossa; **C:** one year after surgery scan, showing a new roof of the glenoid fossa remodeled without any growth of the ankylosed mass remnant.

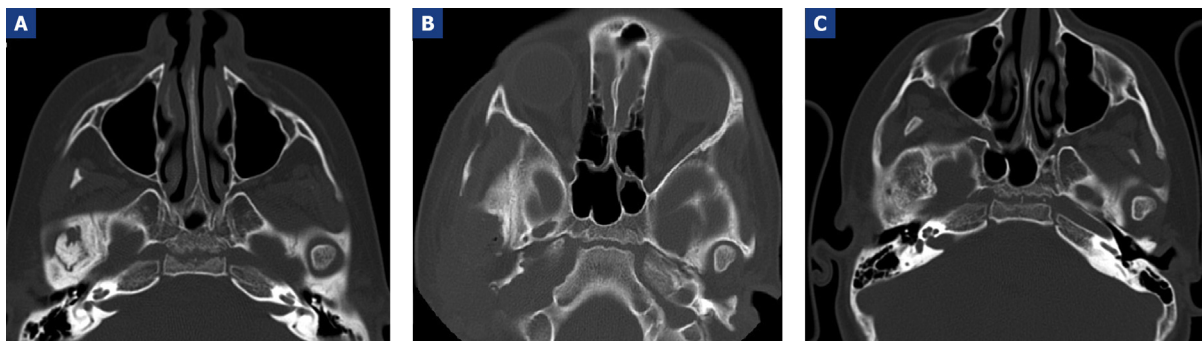


Figure 3. CT scan, axial view; **A:** before surgery scan showing the radiolucent peripheral area between the condyle and surrounding bone structures; **B:** after surgery scan where showing the absence of the right condyle in the glenoid fossa; **C:** one year after surgery scan, showing completely remodeled floor of the middle cranial fossa.

osseous remodeling and reduction of bone fragments including the right condylar fragment as part of the cranial fossa floor repairing bone tissue (figs. 2-4). The patient was discharged home few days later.

One year after the surgical procedure, the clinical results have been exceptional: the maximum mouth opening was 40 mm and the mandibular function had partially recovered. The left mandibular movement was slightly limited due to asymmetric

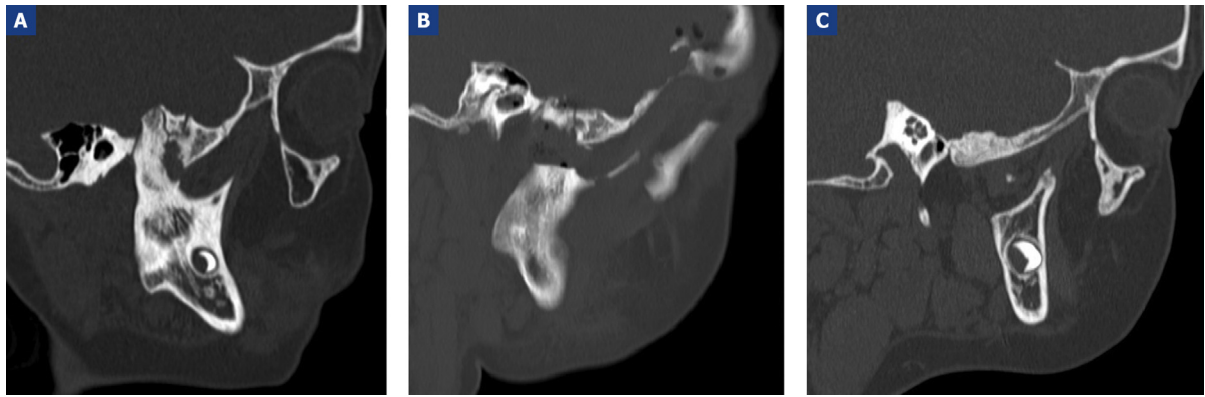


Figure 4. CT scan, sagittal view; **A:** before surgery CT scan, showing the irregular shape of the right condyle dislocated in the middle cranial fossa; **B:** after surgery, showing the extent of the right condyle removed; **C:** one year after surgery, showing completely remodeled floor of the middle cranial fossa without continuity between the two regions.

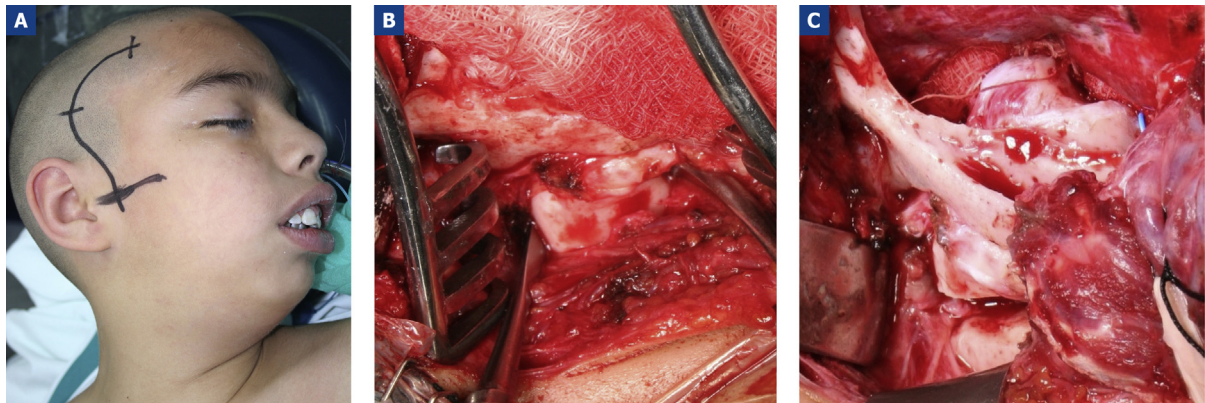


Figure 5. Intraoperative pictures; **A:** flap design; **B:** temporomandibular joint exposed; **C:** upper circle: deformed condyle head in the middle cranial fossa with surrounding soft tissue, probably articular disc remnant. Lower circle: ankylosed joint freed.

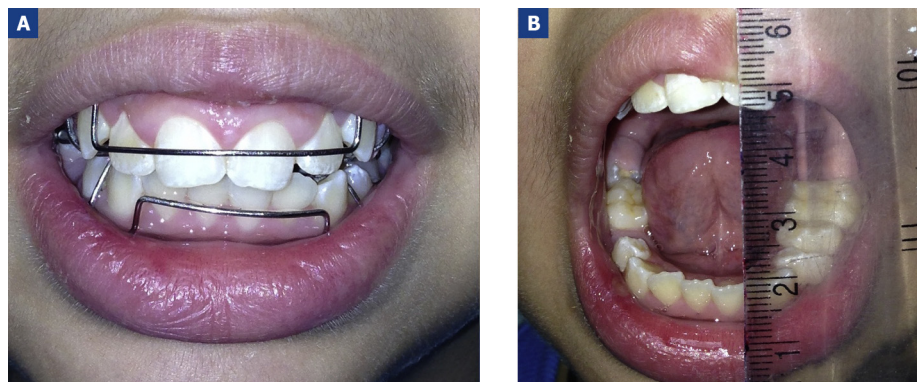


Figure 6. Postoperative pictures; **A:** frontal occlusion view showing midlines alignment; **B:** maximum mouth opening of 26 mm after surgery.



Figure 7. One year postoperative pictures; **A:** frontal view with closed mouth showing lip incompetence improved; **B:** lateral view showing micrognathia and surgical scar; **C:** frontal view of maximum mouth opening of 40 mm.

anatomical development. Further orthodontic and surgical treatment will be necessary (fig. 7).

Discussion

Dislocation of the mandibular condyle into the middle cranial fossa is a very rare phenomenon. Since the first case described by Dingman and Crabb in 1963 [4] and until 2014, only 50 cases were reported in the English literature [5]. This injury is most common in children and young adults [2]. This can be explained by anatomical features of children's condyles when compared to those of adults.

The condylar neck is thicker in children than in adults. Thus, when during trauma, instead of a fracture and a posterior displacement, the condyle is dislocated superiorly and passes through the floor of the middle cranial fossa [2].

Anatomical features that predispose the condyle to dislocation have been proposed by several authors: increase pneumatization of the temporal bone, absence of posterior dentition and small rounded condyle [2]. Other authors propose that this phenomenon occurs when the mandible is hit posteriorly and superiorly while the mouth is open. This pushes the condylar head through the roof of the glenoid fossa into the middle cranial fossa [1,2]. In the case presented, the left condyle is currently very thin and round in shape, but it is impossible to assume that these features were present at the time of the accident, or the mouth was open at the time of the trauma.

Signs and symptoms of a condylar dislocation in the middle cranial fossa are similar to those found in a subcondylar neck fracture [1,2]: mandibular deviation to the dislocated side, reducing the height of the lower jaw on the same side, limited mandibular movements, only ipsilateral occlusion, open bite in the contralateral side, and preauricular pain [2]. In some patients, there auditory deficits, cerebrospinal fluid leak from

the external auditory canal (otorrhea), facial nerve paralysis, consciousness alterations from cerebral contusions, extradural or intradural hematoma, damage to the middle meningeal artery and even meningitis may occur [2].

Diagnosis of intracranial dislocation of the mandibular condyle is very challenging. According to the literature, only half of the cases are successfully diagnosed at first examination. Other cases are only diagnosed when the initial treatment has failed [1]. Difficulties in the diagnosis are based to the fact that there are no particular signs or symptoms exclusively associated with this injury [1]. In addition, conventional radiography is not useful because of the overlap of surrounding structures. CT and MRI are more accurate to reveal the dislocation [1,4] but such exams are not always available. Delayed treatment of this injury may lead to ankylosis of the mandibular condyle dislocated in the middle cranial fossa, which is the case of our report.

To date, only one case of condylar ankylosis in the middle cranial fossa has been published in the English literature [3]. The reported case has very similar clinical and radiographic features to the case presented here. The authors promote the hypothesis raised by Pirok and Merrill [6], in which the temporomandibular joint ankylosis occurred because of the ossification of the intracapsular haematoma in a condylar fracture. This hypothesis is based on in vitro experiments and may be different in living subjects. In addition, if ankylosis is only a simple organization with ossification of an intracapsular haematoma, it should be similar to a normal fracture healing. Progression of an ankylosis is much longer in time and there are still radiolucent zones between the bone fusion area in an ankylosis [7]. In addition, an experimental study by Oztan et al. concluded that intra-articular hemorrhage in the joint space following a trauma does not always progress into new bone formation [8].

In the case presented, it is supposed that when the trauma occurred, there might have been a small dislocation of the

mandibular condyle into the cranial vault, and a minor condyle head fracture, without major morbidity at the time, which explains the lack of clinical signs or symptoms. As a development and growth center, the condyle continued to grow, with an intrusion through the roof of the glenoid fossa. This phenomenon resulted in a deformed and hypertrophic condyle and fixes to the surrounding tissue. Nevertheless, this adhesion is incomplete. Preoperative CT scans reveal that there are radiolucent zones between the condyle and the surrounding bone tissue. This last phenomenon is consistent with Yan's theory of pathogenesis of ankylosis that behaves like a hypertrophic non-union [7].

Many surgical techniques have been described for the treatment of both dislocation of the mandibular condyle into de middle fossa and temporomandibular joint ankylosis [9,10]. Treating the dislocation with an open craniotomy has been proposed by many authors, but it is reserved in cases of neurological damage or when there is a delayed diagnosis [1]. In the case presented by de Oliveira et al. [3], the intracranial ankylosed mass was not removed because of the absence of neurological damage. The intra-articular ankylosed mass was removed using a preauricular flap. In the current case, the neurosurgeon considered that despite the absence of neurological damage, the size of the ankylosed mass in the cranial vault was large, and considering the patient's age and the growth potential, the possible damage to the nearby structures was unpredictable.

This argument is consistent with theories proposed by Salins [9]: despite the absence of neoplastic process, the bone is able to grow back because it acts as a reparative process similar to that found in an exuberant callus or the hypertrophic non-union seen in children's fractures or in fractures that have not been immobilized adequately [7,9].

Temporomandibular joint arthroplasty consisted in a partial removal of the ankylosed mass in the glenoid cavity, leaving a bony remnant as a new roof. This procedure is in partial agreement with Kaban et al.'s [10] theory of radical removal and contradicts Salins's [9] hypothesis of leaving the ankylosed mass intact. No regrowth of the ankylosed mass remnant or recurrence of TMJ ankylosis has been observed after a year. This type of case represents a challenge because of its low frequency. An individual approach and a multidisciplinary work are required to restore mandibular function, esthetics

and to avoid further neurological damage. A CT scan is essential for diagnosis, treatment and follow-up, even when no initial neurological damage is present. When there is a blow in the chin, especially in children, besides suspecting mandibular condyle fracture, it is essential to always look for any neurological signs or symptoms, and consider a possible dislocation of the condyle into the middle cranial fossa.

Disclosure of interest

The authors declare that they have no competing interest.

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