

# Temporomandibular Joint Ankylosis as a Complication of Neonatal Septic Arthritis

## Report of two cases

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## تصلب المفصل الصدغي الفكي كمضاعفة للإلتهاب الإنتاني للمفصل في الأطفال حديثي الولادة

### تقرير عن حالتين

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**ABSTRACT:** Temporomandibular joint (TMJ) ankylosis as a complication of neonatal septic arthritis is rarely reported in the literature. We report two clinical cases of unilateral TMJ ankylosis occurring in paediatric patients subsequent to neonatal septic arthritis. The first case was a 15-month-old male infant who presented to the Sultan Qaboos University Hospital, Muscat, Oman, in May 2010. According to the published English scientific literature, he is the youngest person yet to be diagnosed with this condition. The second case was a five-year-old female who presented to the Al-Nahda Hospital, Muscat, Oman, in October 2011. Both cases presented with facial asymmetry and *trismus*. They subsequently underwent gap arthroplasty and interpositional *temporalis* muscle and *fascia* grafts which resulted in an immediate improvement in mouth opening. Postoperatively, the patients underwent active jaw physiotherapy which was initially successful. Both patients were followed up for a minimum of two years following their surgeries.

**Keywords:** Temporomandibular Joint; Ankylosis; Septic Arthritis; Children; Case Report; Oman.

**المخلص:** قَسَطُ المفصل الصدغي الفكي كمضاعفة للإلتهاب الإنتاني للمفصل الصدغي الفكي في الاطفال حديثي الولادة نادراً ما يُذكر في الأدب الطبي. في هذا التقرير نعرض حالتين من قسط أحادي الجهة للمفصل الصدغي الفكي لطفلين حديثي الولادة كأحد مضاعفات الإلتهاب الإنتاني للمفصل الصدغي الفكي. شخّصت الحالة الأولى لطفل يبلغ من العمر 15 شهراً في مستشفى جامعة السلطان قابوس في مايو 2010، وبمراجعة الأدب الطبي المذكور باللغة الانجليزية يعتبر هذا المريض أصغر طفل يتم تشخيصه بهذه الحالة، أما الحالة الثانية فهي لطفلة تبلغ من العمر 5 سنوات شخّصت في مستشفى النهضة في أكتوبر 2011. كلتا الحالتين عانت من فقدان التناظر بين نصفي الوجه ومن الضرسز. تم إجراء عملية جراحية لفتح القسط الصدغي الفكي لكلتا الحالتين عن طريق رَابِ المفصل واحداث فجوة بين العظمتين الملتحمتين ثم وضع فاصل من العضلة واللفافة الصدغية، مما نتج عنه تحسن واضح في فتح الفم وتحريكه. تم أخضاع المريضين للعلاج الطبيعي مباشرة بعد العملية مع نتائج أولية مُرضية، ثم تمت متابعة كلتا الحالتين لمدة سنتين على الأقل بعد العملية.

**مفتاح الكلمات:** المفصل الصدغي الفكي؛ القسط؛ التهاب المفصل الإنتاني؛ أطفال؛ تقرير حالة؛ عمان.

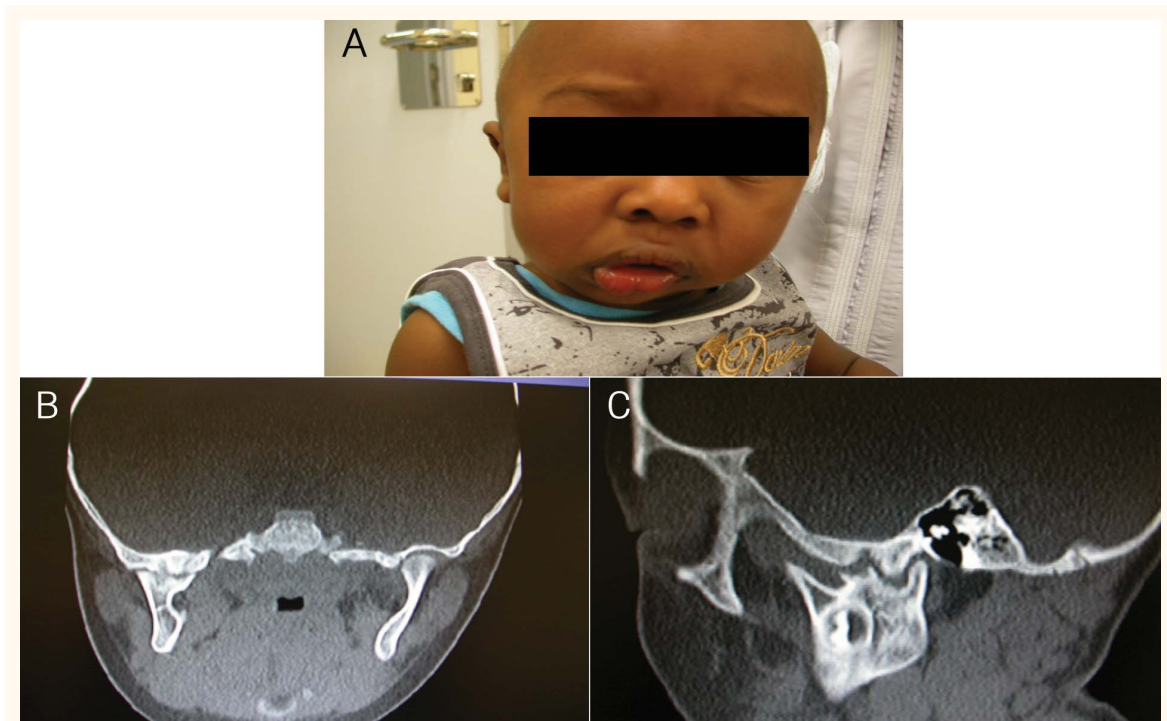
**S**EPTIC ARTHRITIS IS A PURULENT JOINT infection occurring when microorganisms invade the joint space, either through the haematological spread of a distant infection or post-iatrogenic direct joint inoculation.<sup>1,2</sup> The latter can occur either due to traumatic joint exposure to a microorganism invasion or contiguous odontogenic, ear or skin infections.<sup>1,2</sup> As the joint synovium is highly vascular and has no limiting basement membrane, it is more vulnerable to infection by haematological spread.<sup>3</sup> Most infections are monoarticular; however, 10–20% are polyarticular, with the knee being the most commonly involved joint.<sup>1</sup> Septic arthritis usually

affects the knee and the hip; it rarely affects smaller joints, such as the temporomandibular joint (TMJ).<sup>2,4,5</sup>

Septic arthritis of the TMJ is not commonly reported in the literature.<sup>2–4,6</sup> Delayed management of TMJ septic arthritis often leads to irreversible damage of the joint structure with subsequent long-term complications of bony deformity and ankylosis. This report presents two rare cases of TMJ ankylosis following a neonatal septic arthritis infection. Both patients were diagnosed with TMJ ankylosis between one to five years after the infection and presented clinically with facial asymmetry and *trismus*.

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**Figure 1A–C:** A 15-month-old boy presented with (A) *trismus* and chin misalignment. Computed tomography scans in the (B) coronal and (C) sagittal aspects revealed severe bony deformation and ankylosis of the right temporomandibular joint.

## Case 1

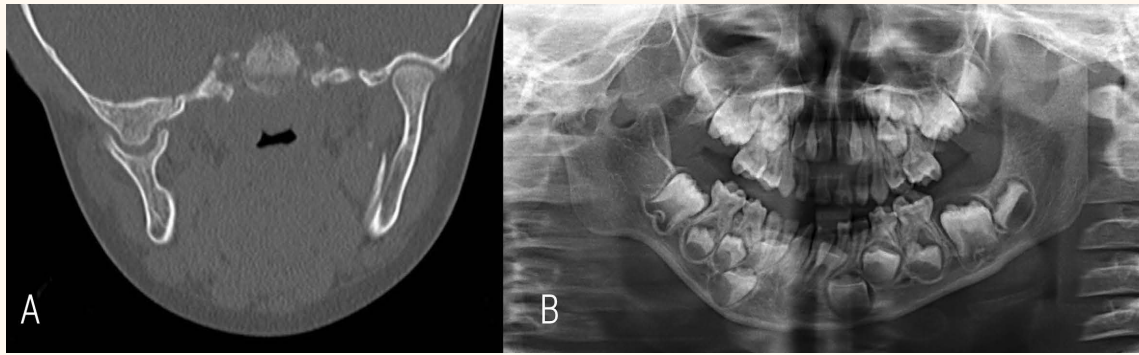
A 15-month-old boy presented to the Oral Health Department at Sultan Qaboos University Hospital, Muscat, Oman, in May 2010 with *trismus* and a chin misalignment [Figure 1A]. His medical history indicated that he had developed neonatal septic arthritis due to an umbilical cord infection at the age of 10 days old. At the time, the neonate had multiple joint swelling and pain involving both knees and hand joints. In addition, there were multiple subcutaneous infections at various sites, including the skin of the right pre-auricular region. Microbiological tests revealed the growth of *Staphylococcus aureus*. The infant recovered well following treatment with intravenous antibiotics. The patient presented again at the age of 10 months due to a recurrence of the infection in both knees and in multiple subcutaneous areas. He again responded well to intravenous antibiotics. There was no reported history of facial trauma or a fall at this time.

A clinical examination showed that the 15-month-old child had a maximum mouth opening of 8 mm with facial asymmetry and chin deviation to the right. Maxillofacial computed tomography (CT) showed gross enlargement and deformation of the right condylar head with evidence of fusion with the base of the skull. This led to a diagnosis of right TMJ ankylosis [Figures 1B & C].

At the age of 16 months, the child underwent gap arthroplasty with interpositional *temporalis* muscle and *fascia* grafts with removal of the ipsilateral coronoid process under general anaesthesia to release the right TMJ ankylosis. The mouth opening was increased intraoperatively from 8 mm to 24 mm. Recovery during the postoperative period was uneventful and the patient immediately began jaw exercises and physiotherapy. At a six-month postoperative review, the patient was able to maintain a mouth opening of 30 mm with satisfactory lateral movement.

When he was three years old, the patient was found to have a maximum mouth opening of 22 mm. A CT scan revealed growth of the condylar head on the mediolateral aspect and a U-shaped condylar head on the coronal aspect. A bony overgrowth was seen extending from the cranial aspect into the condylar depression [Figure 2A]. His parents were reluctant to proceed with any further surgical interventions, so the patient was closely monitored with continuous physiotherapy and jaw exercises.

At five years of age, the *trismus* had worsened with a maximum mouth opening of 13 mm. An orthopantomogram showed that the bony mass of the deformed right condyle had increased and the gap space had decreased, indicating a recurrence of the right TMJ ankylosis [Figure 2B]. The patient was operated on again to release the ankylosis under



**Figure 2A & B:** A: Coronal computed tomography scan of the patient at three years old showing mediolateral temporomandibular joint (TMJ) bony overgrowth resulting in a U-shaped deformity, shortening of the mandibular ramus and downward overgrowth of the cranial bone into the deformed U-shaped condyle. B: Orthopantomogram of the patient at five years old showing right TMJ bony deformity and a reduced gap with a short ramus in comparison to the normal left side.

general anaesthesia and the intraoperative mouth opening was increased from 13 mm to 32 mm. The postoperative recovery period was uneventful and the patient again began jaw exercises and physiotherapy.

## Case 2

A five-year-old girl presented to the Oral & Maxillofacial Surgery Unit of Al-Nahda Hospital, Muscat, Oman, in October 2011 with a complaint of longstanding *trismus* [Figure 3A]. Her medical history indicated neonatal septic arthritis occurring on the second day post-delivery and manifesting as a high fever with painful swelling of the left knee. There was no record of the causative microorganism for the infection. The neonate was treated with intravenous antibiotics for three weeks, followed by a one-month oral antibacterial course. She recovered well but suffered a recurrence of the knee swelling one year later which was again successfully treated with antibiotics.

At the age of five years, the patient had a maximum mouth opening of 5 mm, restriction of the lateral jaw movements and clinically apparent facial asymmetry with chin misalignment [Figure 3B]. CT scans showed a bony deformation of the left TMJ with reduced joint space and partial fusion of the condylar head with the base of the skull, which confirmed the diagnosis of left TMJ ankylosis [Figures 3C & D].

The patient underwent a gap arthroplasty with interpositional *temporalis* muscle and *fascia* grafts. This improved her mouth opening ability from 5 mm to 27 mm. Postoperatively, the recovery period was uneventful and was followed by immediate jaw exercises and physiotherapy. At a six-month postoperative review, the patient maintained satisfactory mouth opening and lateral mandibular movements. At a two-year follow-up, the mouth opening had further

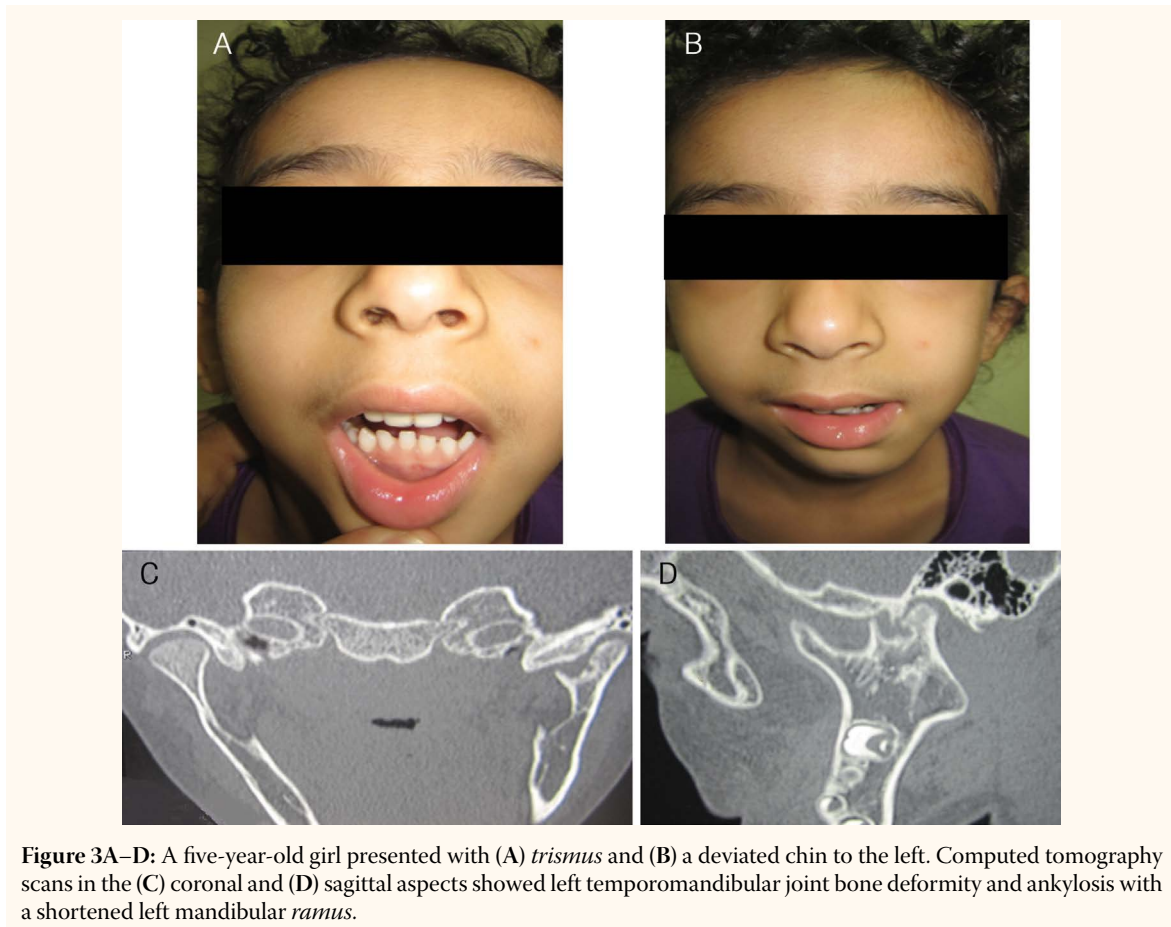
improved and the patient could open her mouth to 40 mm.

## Discussion

TMJ septic arthritis is an infection of the joint space that can lead to many complications, including intracranial abscesses, growth impairment and deformity of the condyle and destruction of the joint. Subsequent TMJ ankylosis can be a long-term complication.<sup>4</sup> TMJ ankylosis is the replacement of normal joint articulation with fibrous, fibro-osseous or bony tissues which results in partial or complete fusion of the mandibular condyle to the base of the skull.<sup>5</sup> It is the most severe long-term complication of septic arthritis and has a devastating presentation. The long-term effect of joint deformity and ankylosis is unpredictable, depending on the degree of joint damage, type of ankylosis and age of the patient.<sup>5</sup> As such, each case is unique and should be assessed individually. In the paediatric population, patients with bilateral TMJ ankylosis usually present with facial deformity, *trismus* and *dysmasesis*.<sup>5</sup> Deviation of the jaw when the mouth is open and facial asymmetry are the usual presentations of unilateral TMJ.<sup>5,7</sup>

Owing to the gradual nature of the joint deformity and ankylosis, it may take several months or years before the facial deformity is noticed and medical treatment is sought, as was the case for the patients described in the current report. Both cases presented with facial asymmetry due to the TMJ deformity, growth deficiency of the affected condyle and ramus, misalignment of the chin and mandible to the affected side and *trismus*. These findings are in line with previously published cases.<sup>5,7</sup>

According to the available English literature, there were 29 reported cases of septic arthritis involving



**Figure 3A–D:** A five-year-old girl presented with (A) *trismus* and (B) a deviated chin to the left. Computed tomography scans in the (C) coronal and (D) sagittal aspects showed left temporomandibular joint bone deformity and ankylosis with a shortened left mandibular *ramus*.

the TMJ from 1931–2011, with only four cases of ankylosis subsequent to septic arthritis in neonates.<sup>7,8</sup> The current report adds two rare cases of neonatal septic arthritis that resulted in TMJ ankylosis after the haematogenous spread of a distant systemic infection involving multiple joints. In addition, to the best of the authors' knowledge, the first case represents the youngest child so far to be diagnosed and treated for TMJ ankylosis due to neonatal septic arthritis.

Septic arthritis can be caused by bacteria, viruses, fungi or parasites. The most common isolated causative bacteria are Gram-positive *S. aureus* and *Streptococcus*.<sup>1,4</sup> Gram-negative *bacilli* are found in 20–50% of neonates, the elderly, intravenous drug users and the immunocompromised.<sup>1,4</sup> *S. aureus* was found to be the causative agent in the first case of the current report. Unfortunately, the microorganism in the second case was unknown.

Joint infections initiate an inflammatory response in the host which is responsible for much of the joint destruction. Inflammation leads to synovial hyperplasia and subsequent intra-articular abscess formation. This, in turn, leads to increased intra-articular pressure with subsequent diminished joint blood flow, ischaemia and necrosis of the cartilage.<sup>1,6</sup> Intra-articular cartilage damage may be seen after

only three days, with irreversible changes in joint structure occurring as early as seven days.<sup>1,6</sup> Acute TMJ infections manifest clinically as severe joint pain, *trismus*, acute malocclusion, contralateral deviation on opening, preauricular erythema and swelling.<sup>4,6</sup>

Radiographical assessment is essential in evaluating the extent and nature of the TMJ ankylosis following septic arthritis. The ankylosis will usually appear as an ill-defined radiopaque mass with partial or complete fusion between the mandibular condyle, glenoid *fossa*, articular eminence and base of the skull with an irregular overgrowth. A plain radiograph can underestimate the extent of the ankylotic mass as it gives no information about the anatomy medial to the condyle.<sup>9</sup> A CT scan is more diagnostically useful and was hence used to evaluate the extent of ankylosis in both of the cases reported here.

Surgical management of TMJ ankylosis continues to be a challenge in children. Different surgical techniques have been reported in the literature, including gap or interpositional arthroplasty and wide excision of the ankylotic mass with varying methods of reconstruction.<sup>5,7</sup> However, no single method has yet reported complete success. This is likely due to the challenging nature of this condition, poor compliance with postoperative physiotherapy and the unavoidable

complication of relapse and recurrence of the ankylosis, as with the first case reported here. Most published reports of TMJ ankylosis following septic arthritis only provide short-term follow-up periods of less than one year.<sup>7</sup> However, the patients in the present case report were followed up for a minimum of two years following surgical release of the TMJ ankylosis. Long-term follow-up will continue with these patients until they reach early adulthood in order to monitor facial growth and to detect any further complications.

## Conclusion

TMJ neonatal septic arthritis is uncommon and early recognition is crucial. Delayed diagnosis and management can lead to devastating functional and aesthetic complications of facial deformity, *trismus* and *dysmasesis* due to the progressive and permanent joint distortion, overgrowth and gradual fusion with the base of the skull. The management of such complications is challenging with low success rates and a high likelihood of relapse and recurrence of the ankylosis. Rapid diagnosis and treatment of the TMJ septic

arthritis is critical in preventing and avoiding such complications.

## References

1. Ohl CA, Forster D. Infectious arthritis of native joints. In: Bennett E, Dolin R, Blaser M, eds. *Mandell, Douglas, and Bennett's Principles and Practice of Infectious Diseases*. 8th ed. Philadelphia, Pennsylvania, USA: Saunders, 2015. Pp. 1302–17.
2. Cai XY, Yang C, Zhang ZY, Qiu WL, Chen MJ, Zhang SY. Septic arthritis of the temporomandibular joint: A retrospective review of 40 cases. *J Oral Maxillofac Surg* 2010; 68:731–8. doi: 10.1016/j.joms.2009.07.060.
3. Lynn MM, Mathews CJ. Advances in the management of bacterial septic arthritis. *Int J Clin Rheumatol* 2012; 7:335–42. doi: 10.2217/ijr.12.11.
4. Leighty SM, Spach DH, Myall RW, Burns JL. Septic arthritis of the temporomandibular joint: Review of the literature and report of two cases in children. *Int J Oral Maxillofac Surg* 1993; 22:292–7. doi: 10.1016/S0901-5027(05)80519-3.
5. Kaban LB, Bouchard C, Troulis MJ. A protocol for management of temporomandibular joint ankylosis in children. *J Oral Maxillofac Surg* 2009; 67:1966–78. doi: 10.1016/j.joms.2009.03.071.
6. Goldschmidt MJ, Butterfield KJ, Goracy ES, Goldberg MH. Streptococcal infection of the temporomandibular joint of hematogenous origin: A case report and contemporary therapy. *J Oral Maxillofac Surg* 2002; 60:1347–53. doi: 10.1053/joms.2002.35736.
7. Chaves Netto HD, Nascimento FF, Chaves Md, Chaves LM, Negreiros Lyrio MC, Mazzone R. TMJ ankylosis after neonatal septic arthritis: Literature review and two case reports. *Oral Maxillofac Surg* 2011; 15:113–19. doi: 10.1007/s10006-010-0210-4.
8. Sembronio S, Albiero AM, Robiony M, Costa F, Toro C, Politi M. Septic arthritis of the temporomandibular joint successfully treated with arthroscopic lysis and lavage: Case report and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2007; 103:e1–6. doi: 10.1016/j.tripleo.2006.08.028.
9. Sanders R, MacEwen CJ, McCulloch AS. The value of skull radiography in ophthalmology. *Acta Radiol* 1994; 35:429–33. doi: 10.1177/028418519403500506.