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IMAGING

Boering's Arthrosis of the Temporomandibular Joint in a 9-Year-Old Girl; a Case Report

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Abstract

Boering's arthrosis is a rare disorder of the temporomandibular joints that usually occurs in the second decade and seldom after age 20. It was first described by G. Boering in 1966. It is more common in females and can be unilateral or bilateral. The disease results in resorption of the condyle with elongation of the articular surface resulting in the characteristics toadstool appearance observed on panoramic images. Condylar changes are often accompanied by anterior displacement of the disc and remodeling of the glenoid fossa. Facial asymmetry is almost always present. We present a case of a 9-year-old female with Boering's arthrosis. She presented with facial asymmetry and bite abnormality but did not report symptoms typically associated with temporomandibular disorders.

Keywords Arthrosis · TMJ · CBCT · MRI · Facial asymmetry

Introduction

Boering's arthrosis (BA) is a rare disorder of the temporomandibular joints (TMJs) that mainly affects the condyles in children. It was first described by G. Boering in 1966 and has been seldom reported in the literature since then. Both, developmental and degenerative, origins have been proposed. Terms including juvenile arthrosis, arthrosis deformans juvenilis, and condylysis have been used to describe this condition [1]. The onset of the disease is typically in the second decade. It is significantly more common in females with a 9:1 female to male ratio and could be unilateral or bilateral [2]. The disease process results in an alteration of the normal condyle morphology to "toadstool" or "mushroom" appearance. The disease is further characterized by a decrease in the vertical height of the condyle, an abnormal backward bend of the condylar head and neck and mandibular hypoplasia. Boering described the classic features of the disease based on condyle changes observed in panoramic images taken over periods of time. With cone beam computed tomography (CBCT) TMJs can be studied without superimposition and distortion inherent in panoramic images. This presents an opportunity for a more accurate analysis of the osseous changes occurring due to BA.

We report a case of BA in a 9-year-old girl who underwent investigations including panoramic, cephalometric, CBCT, magnetic resonance imaging (MRI), and blood tests for evaluation of the disease process affecting her TMJs. Written informed consent was obtained from patient's mother for use of protected health information for this publication.

Case Presentation

A 9-year-old girl presented for orthodontic treatment with the chief complaint that "the way my daughter bites doesn't seem right" as stated by the mother. The patient did not report any previous medical history, including trauma or tenderness or abnormalities in other joints. Clinically, she presented with lower jaw deviation to the right side (Fig. 1) and slightly convex soft tissue profile (Fig. 2). Intraoral examination revealed an Angle's class III subdivision left malocclusion, characterized by malalignment in both dental arches, unilateral crossbite on the right side from the lower right lateral incisor to the 1st molar (Figs. 3 and 4) and lower dental midline shift of 3 mm to the right, in relation to the upper dental midline.

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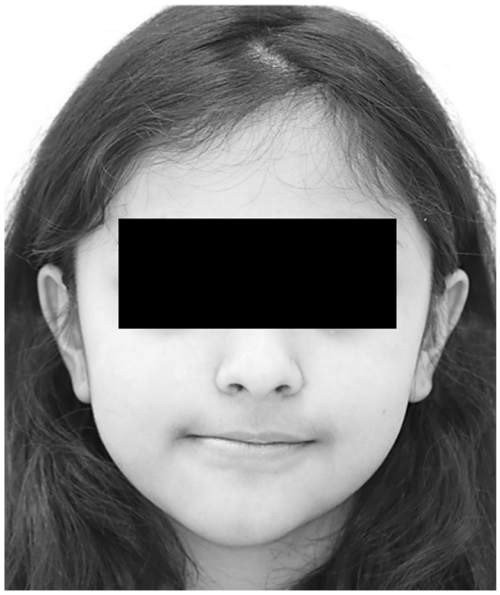

Fig. 1 Facial asymmetry due to right deviation of the mandible

Interincisal distance with maximum mouth opening was 30 mm.

Panoramic image (Fig. 5) revealed the presence of all permanent dentition with the 3rd molars in various stages of development. The right condyle was significantly shorter in height than the left, exhibited erosion of the superior surface, and subchondral sclerosis. The lateral and posteroanterior cephalometric images (Figs. 6 and 7) showed skeletal class III relationship and asymmetry.

CBCT exam (closed-mouth) of the right TMJ (Fig. 8) showed anterior and distal osteophytosis, flattening, and

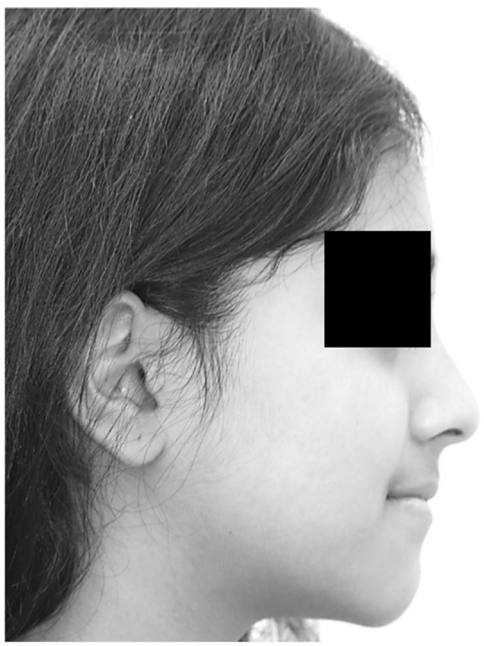

Fig. 2 Profile of head (right)

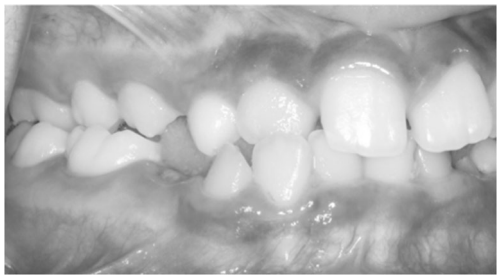

Fig. 3 Crossbite on the right

erosion of the anterosuperior and posterosuperior surfaces, subchondral sclerosis, and subchondral cystic degeneration (Ely cyst) in the condyle. These changes caused volume loss resulting in reduced vertical height, elongation of the articulating surface and characteristic "toadstool" appearance of the condyle. Condyle was positioned posterior to the center of significantly modified glenoid fossa with reduction in posterior and an increase in superior joint space. There was effacement of the posterior slope of articular eminence. The roof of the glenoid fossa exhibited subchondral sclerosis. Left TMJ (Fig. 9) showed anterior osteophytosis and flattening of the posterosuperior surface of the condyle. Condyle was positioned posterior to center of glenoid fossa with reduction in superior joint space. Significant flattening was accompanied with effacement and subchondral sclerosis in the posterior slope of articular eminence. A follow-up CBCT exam, acquired after 16 months, showed no improvement in the right condyle; In addition, significant subchondral sclerosis was present in the posterior slope of articular eminence (Fig. 8b). Left condyle showed an increased flattening and erosion of the posterosuperior surface and flattening, erosion, and subchondral sclerosis in the posterior slope of articular eminence (Fig. 9b). Findings in the follow-up CBCT exam suggest an increase in the intensity and continued progression of the disease in both joints.

An MRI exam (3-mm slice thickness) was acquired 3 weeks after the initial CBCT exam. The MR images exhibited anterior displacement of the discs in both joints (Figs. 10 and 11). A 4.4-mm thick layer of

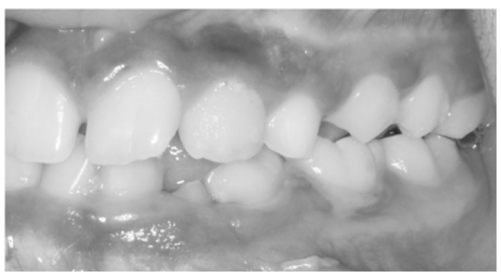

Fig. 4 Normal bite on the left

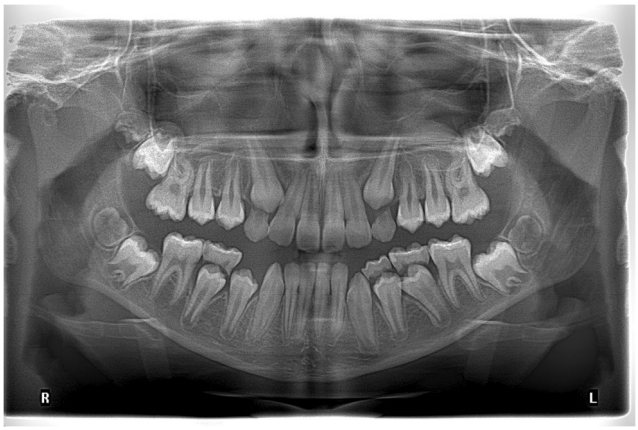

Fig. 5 Panoramic: flattening, subchondral sclerosis, and reduction of the vertical height in the right condyle

pannus was present in the right joint (Fig. 10) resulting in an increase in the joint space. Fluid was seen in the inferior compartment of the right joint (Fig. 10b).

Her blood tests including creatinine, liver function test (LFT), c-reactive protein, white cell count, platelet count, erythrocyte sedimentation rate (ESR), and rheumatoid factor (RF) were within normal range. Only mean corpuscular hemoglobin (MCH) and mean corpuscular hemoglobin concentration (MCHC) were borderline low.

BA is primarily managed conservatively including reduction in joint loading during active phase of the disease, using NSAIDs to treat joint inflammation (if present) and management of muscle hyperactivity and parafunction. Orthodontic therapy with or without orthognathic surgery may be needed for correction of facial deformity. However, orthodontic therapy may cause further joint damage due to additional stress and should be undertaken with caution after the disease has been in remission for 6–12 months [1, 2]. Since the initial and follow-up, clinical and imaging exams (CBCT and MRI) suggest an active disease process, our patient is currently under observation of a team of healthcare providers consisting of an orthodontist, an oral and maxillofacial radiologist (OMR), an oral and maxillofacial surgeon, and a pediatrician with the orthodontist and the OMR taking the lead.

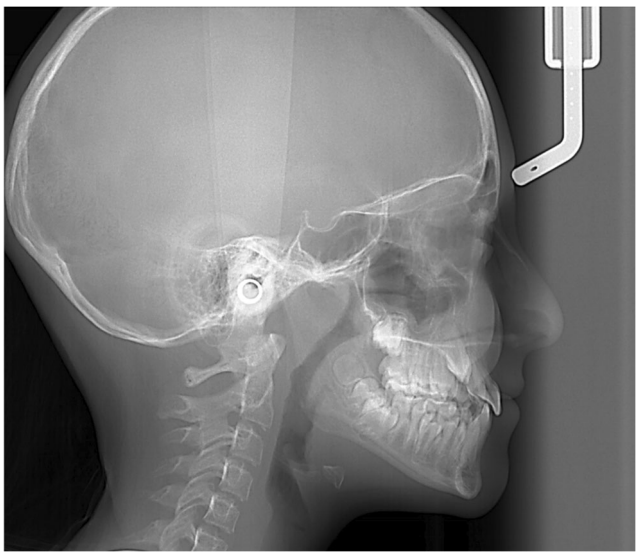

Fig. 6 Lateral cephalometric

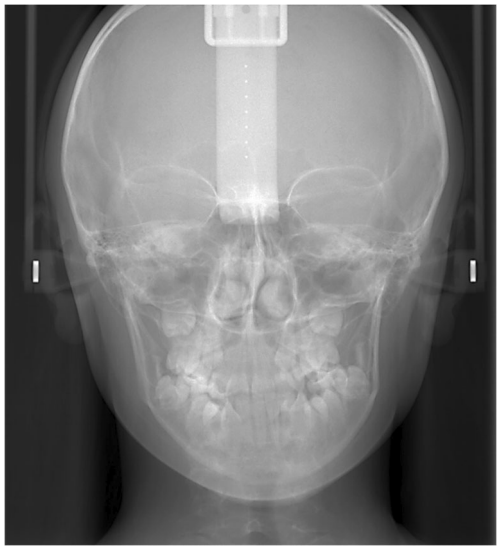

Fig. 7 Posteroanterior cephalometric

The foremost objective is to establish that the disease process has ceased. It will be accomplished with yearly follow-up clinical and imaging exams until the joints exhibit no further changes. The imaging exams will consist of small field-of-view (FOV) TMJ-focused CBCT scans. An MRI exam will also be acquired once no further osseous changes are observed in the CBCT exams. If there is an increase in the severity of the disease, intra-articular steroid injections will be considered. Once the disease process has halted, the patient will be re-evaluated for either the orthodontic or surgical-orthodontic treatment to correct the crossbite and facial asymmetry.

Conclusions

The etiology of BA is unclear. Both, developmental and degenerative, origins have been proposed. It has been postulated that it may represent a hyperactive form of degenerative joint disease (DJD) or may be caused by avascular necrosis (AVN) or hormonal abnormalities [1]. The disease generally occurs at the time of puberty during the second decade. Our patient was 9 years of age (pre-puberty) when diagnosed. She was in an excellent general health with no disease in other joints;

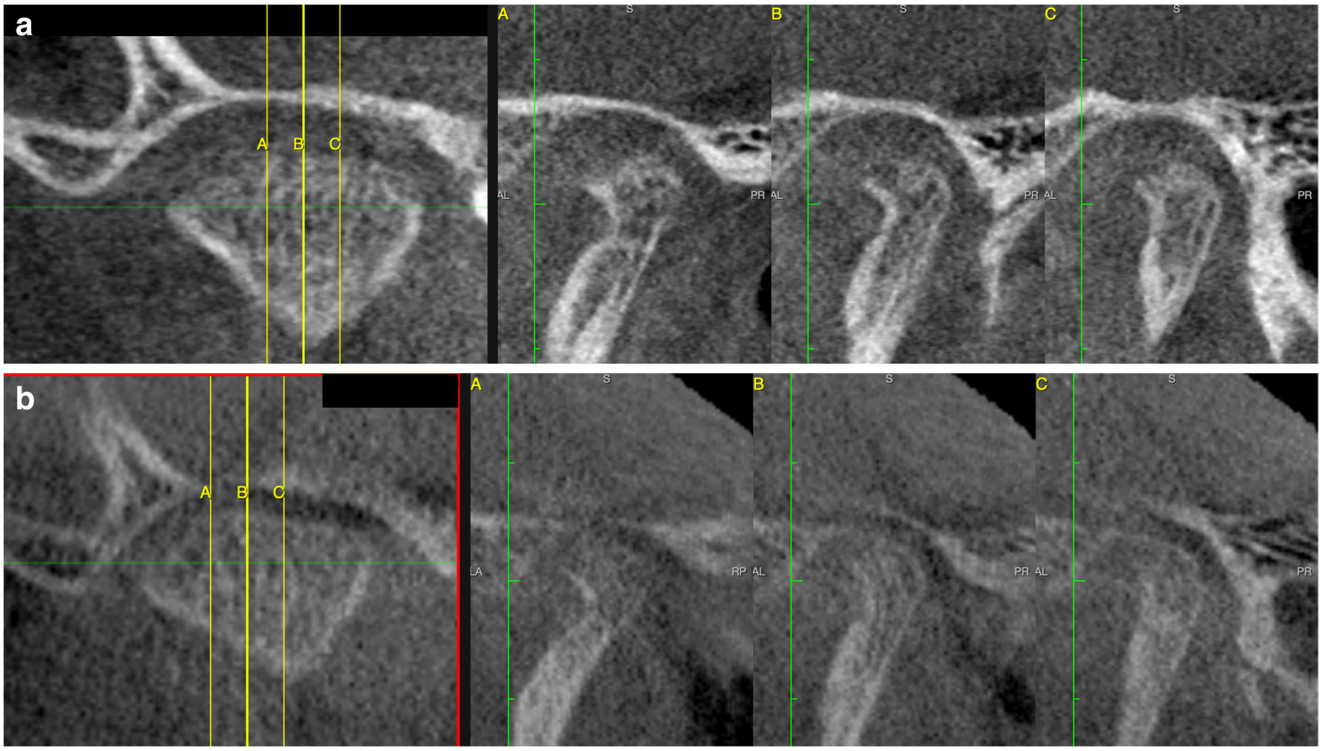

Fig. 8 **a** CBCT; axially corrected sagittal and coronal views of right TMJ (closed mouth): anterior osteophytosis, flattening and erosion of the anterosuperior surface and an Ely cyst in the subchondral area of the condyle. There is significant modification of the glenoid fossa; condyle is positioned posterior to the center of the fossa. **b** CBCT; follow-up axially corrected sagittal and coronal views of right TMJ (closed mouth): anterior osteophytosis and flattening and erosion of the anterosuperior surface of the condyle is present. Erosion and subchondral sclerosis are seen in the posterior slope of articular eminence

Rheumatic factor was normal. The disease was confined to the temporomandibular joints and in addition to the characteristics of BA, exhibited osseous, meniscus, and soft tissue changes commonly seen in DJD. In the right joint, the condyle exhibited classic features of DJD with significant modification of the fossa resulting in an increase in fossa size. The left condyle showed degenerative changes but significantly less pronounced compared with the contralateral condyle. As opposed to the right joint, left joint exhibited a reduction in joint space. Both joints had anterior displacement of the discs with pannus formation in the right joint causing the increased joint space.

AVN, also known as ischemic necrosis or aseptic necrosis, is necrosis of the epiphyseal or subarticular bone due to diminished or disrupted blood supply in the absence of infection [3]. AVN affects structural integrity of the bone which may cause the bone and eventually the joint to collapse resulting in the osseous changes observed in the affected joints. It may be associated with underlying hematological disorders, skeletal dysplasias, and chemotherapeutic agents, including exogenous steroid administration, trauma, decompression (barotrauma), and familial predisposition [4]. AVN lesions exhibit low signal in T1 images, high T2 signal in early (acute) phase and low signal in late phase of the disease [4]. Our patient did not exhibit the characteristics MRI features of AVN.

Estrogen receptors have been identified in the TMJs of female primates; in human TMJ tissues of patients affected by TMJ symptoms such as TMJ pain, headache, myofascial pain, clicking and popping, and crepitation; and in arthritic knee joints. Estrogen is known to mediate cartilage and bone metabolism in the female TMJ [2]. Wolford [2] hypothesize that an increase in estrogen receptors likely results in hyperplasia of synovial tissues followed by production of substrates that destroy supporting tissues of the disc causing anterior displacement. The hyperplastic synovial tissues then surround the condylar head and expose it further to substrates that cause generalized resorption. The destructive osseous and disc changes may exaggerate in response to joint loading from parafunctional activity, trauma, orthodontic treatment, or orthognathic surgery [2]. At 9 years of age (pre-puberty), the probability of estrogen levels high enough to cause joint destruction to an extent seen in this patient is low.

4Literature search returned Boering's original publication and an abstract documenting 6 cases [5, 6]. Similarity of imaging features with conditions such as DJD and juvenile idiopathic arthritis (JIA) may result in misdiagnosis and underreporting. Our patient

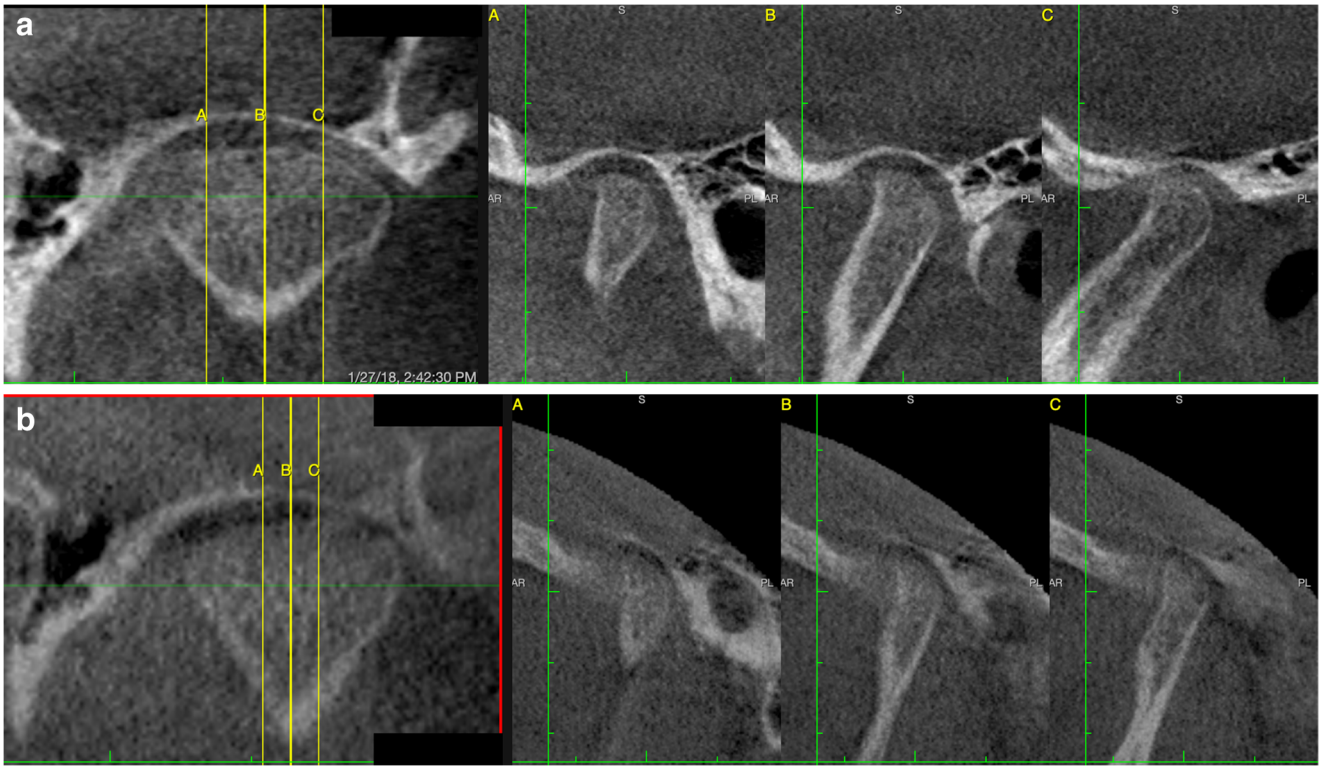

Fig. 9 **a** CBCT; axially corrected sagittal and coronal views of left TMJ (closed mouth): anterior osteophytosis, flattening of the posterosuperior surface of the condyle and reduced joint space are seen. There is flattening and subchondral sclerosis of the articular eminence. **b** CBCT; follow-up axially corrected sagittal and coronal views of left TMJ (closed mouth): There is an increase in flattening and erosion of the posterosuperior surface of the condyle. Flattening, erosion, and subchondral sclerosis are seen in the posterior slope of the articular eminence

predominantly exhibited imaging features generally associated with DJD. However, the osseous and soft tissue changes may have been secondary to disease processes such as AVN and hormonal changes. It may be helpful to measure estrogen levels in patients suspected of BA. This will not determine the cause but may point to the course the disease may take especially in younger, prepuberty patients whose estrogen levels would likely rise as the puberty approaches.

The condylar cartilage is a major growth site in the craniofacial complex and its involvement, consequently, has detrimental effects on the development of the facial skeleton in general and lower third of the face in particular. Since the condylar growth cartilage is present below the superficial articular cartilage layer, endochondral mandibular growth can be disturbed when there is erosion of the surface of the condyle. Mandibular growth disturbance can manifest unilaterally or

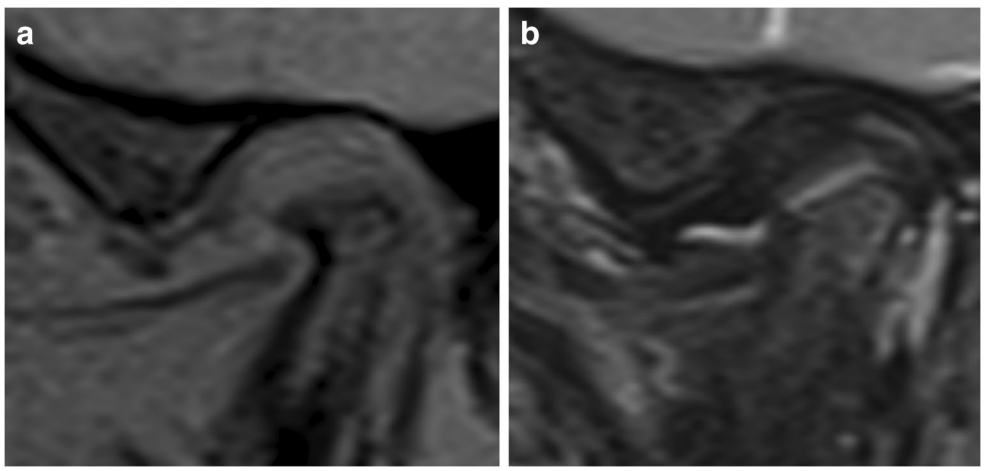

Fig. 10 **a** MRI; axially corrected T1 turbo spin echo fat saturated sagittal view of right TMJ (closed mouth): There is anterior displacement of the disc. Pannus is increasing the joint space. **b** MRI; axially corrected T2 fat saturated sagittal view of right TMJ (closed mouth): Bright signal from fluid accumulation in the inferior compartment of the joint

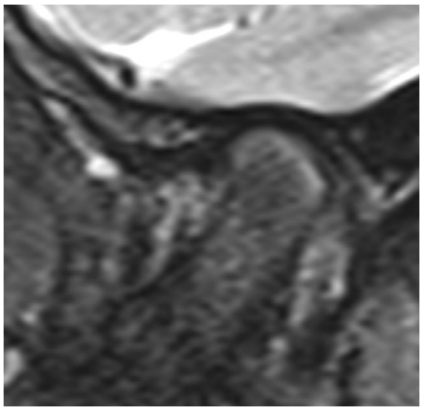

Fig. 11 MRI; axially corrected T2 fat saturated sagittal view of left TMJ (closed mouth): Anterior displacement of the disc

bilaterally leading to vertical and sagittal growth abnormalities. When children develop unilateral condylar erosion, asymmetry occurs with chin deviation to the involved side. From a functional point of view, overloading of the already diseased joint can lead to excessive condylar damage. As the condition progresses, muscle hypoactivity and malfunction may contribute to more craniofacial growth deviations. It is important that diagnostic images of the TMJs of young patients are thoroughly evaluated for any deviations from the normal. Early diagnosis and management may reduce the functional and esthetic sequela of BA.

Data Availability A signed copy of the "consent to publish" is available for review on request.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

Ethical Approval An ethical approval from the Institutional Review Board of the university was not required because all patients' data presented in this case report has been de-identified to maintain patient privacy.

Informed Consent A written "consent to publish" was obtained from the parent of the patient presented in this case report.

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