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CASE REPORT

BILATERAL CONDYLAR HYPOPLASIA WITH EAGLE'S SYNDROME: A RARE CASE REPORT

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ABSTRACT

Condylar hypoplasia is a multifactorial bone disease characterized by underdevelopment or incomplete development of the mandibular condyle. Eagle's syndrome refers to symptomatic elongation of the styloid process or calcified stylohyoid ligament with an unknown etiology. Hereby presenting a rare case of bilateral condylar hypoplasia with Eagle's syndrome in a 31 year old male patient who reported with trismus and tenderness in the temporomandibular joint. Clinical examination and radiological investigations revealed hypoplasia of the condyles with elongation of the styloid process seen bilaterally. Early diagnosis by thorough clinical and radiographic examination helps to achieve a pertinent management for the current scenario.

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INTRODUCTION

Condylar hypoplasia refers to underdevelopment or defective formation of the condyle which can be genetic (congenital) or acquired (Arun, 2002 and Ferguson, 1978). Genetic background includes association with syndromes like Treacher Collins syndrome, Crouzan syndrome, congenital hemifacial macrosomia, cleft lip and palate. Acquired condylar hypoplasia occurs as a result of local factors like trauma, mandibular infection, spread of middle ear infection, dietary and endocrine causes (Arun, 2002; Ferguson, 1978). The severity of deformity depends on degree of dysplasia or agenesis of the tissue involved. Orthopantogram is a widely used diagnostic aid for Eagle's syndrome. The elongated styloid process-pathognomonicof Eagle's syndrome, may lead to craniofacial, cervical and back pain but the cause and effect is rarely recognized (Mortellaro, 2002). The condition was first described in 1937 by Watt Weems Eagle as stylalgia, later to be known as Eagle's syndrome. The normal length of styloid process may vary from 20-30 mm long (Mortellaro, 2012 and Ilguy, 2005), however a 30mm or longer process is considered anomalous. It can be manifested unilaterally or bilaterally. It usually occurs as an accidental finding in routine radiographic investigations because it remains asymptomatic in most of the cases. The case becomes critical when the elongated styloid process comes in contact with the internal carotid artery on turning the head, causing compression of the artery or causing injury to the blood vessel, restricts the blood flow, potentially resulting in transient ischemic attack or stroke. Henceforth we present this uncommon case showing a combination of bilateral condylar hypoplasia with Eagle's syndrome.

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Case Report

A 31-year old male patient reported to the department of oral medicine and radiology with the chief complaint of trismus and tenderness on the left side of the face for the past 5years.Patient has difficulty in opening the mouth usually in the early morning. Patient did not undergo any treatment for the same and has reported for further management. Patient revealed non-contributory medical/ surgical history. Patient was unable to recall any previous history of trauma in the affected region. Intra oral examination revealed hypodontia and microdontia with evident malocclusion. Upon extra-oral examination, there was gross asymmetry on left side of the face (Figure 1) with deviation to the same side. Routine TMJ examination, was done to eliminate pain due to myofascial pain dysfunction syndrome and other temporomandibular joint disorders. There was evident clicking of both right and left temporomandibular joints with tenderness elicited on the left side while opening the mouth. Proceeding further with the investigations, panoramic radiograph revealed presence of underdeveloped condylar head showing decrease in the anteroposterior dimensions with a slender condylar neck and a decrease in the intra articular space on both right and left side. As an incidental finding, elongation of the styloid process was evident bilaterally (Figure 2). Furthermore, a cone beam computed tomography (CBCT) was made to affirm the condition and to proceed with the management. Coronal (Figure 3A, 3B,3C,3D)and sagittal (Figure 4) sections revealed a significant reduction in the dimensions of right and left condyles and thinning out of the condylar neck was also evident with decrease in the intra-articular space on the left side. Axial section (Figure 5) showed an altered shape of the condylar head bilaterally.

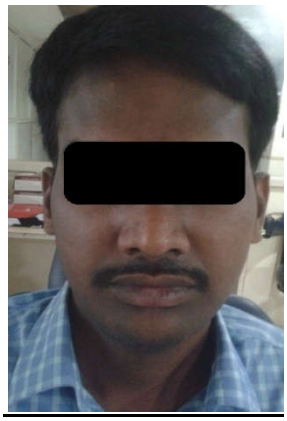

Figure 1. Shows gross asymmetry on left side of the face

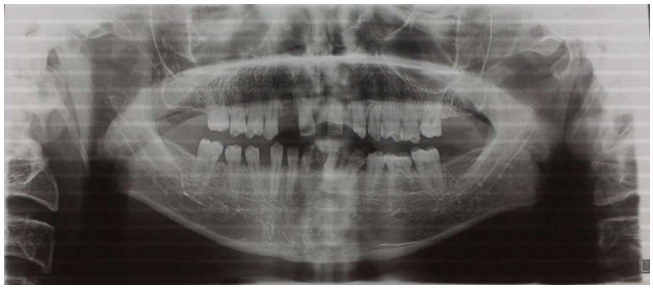

Figure 2. Panoramic radiograph revealing presence of underdeveloped condylar head showing decrease in the anteroposterior dimensions with a slender condylar neck and a decrease in the intra articular space on either side. As an incidental finding, elongation of the styloid process was evident bilaterally

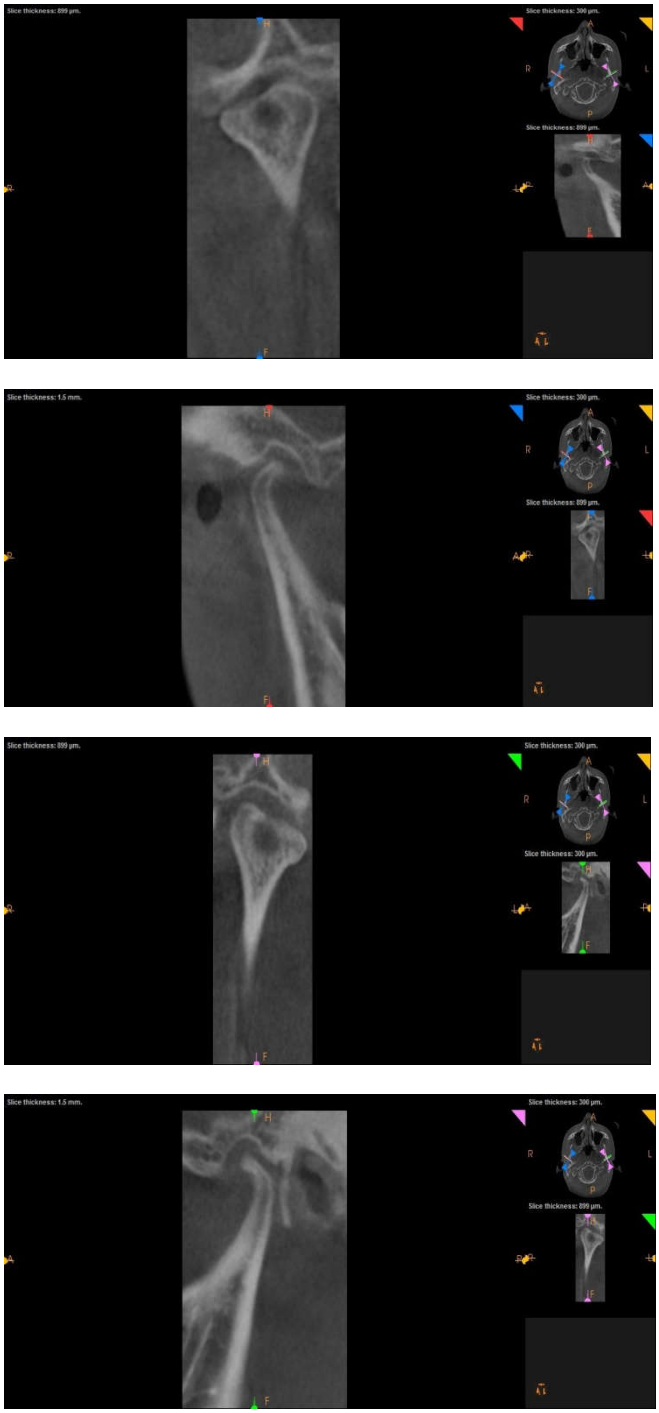

Figure 3A and 3B Coronal section of CBCT of right side and Figure 3C and 3D coronal section of CBCT of left side showingsignificant reduction in the dimensions of right and left condyles and thinning out of the condylar neck is also evident with decrease in the intra-articular space on the left side

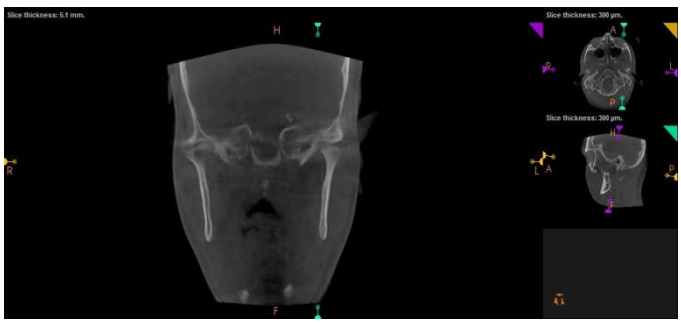

Figure 4. Sagittal section of CBCT showing reduction in the dimensions of the right and left condyles

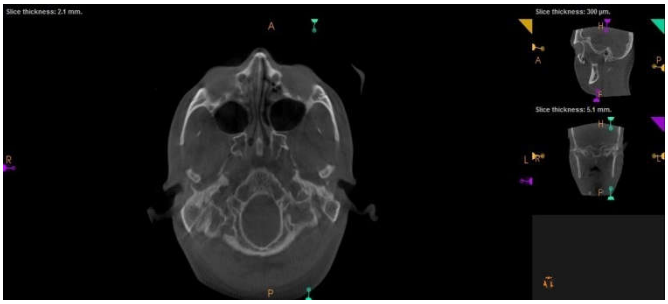

Figure 5. Axial section of CBCT shows an altered shape of the condylar head bilaterally

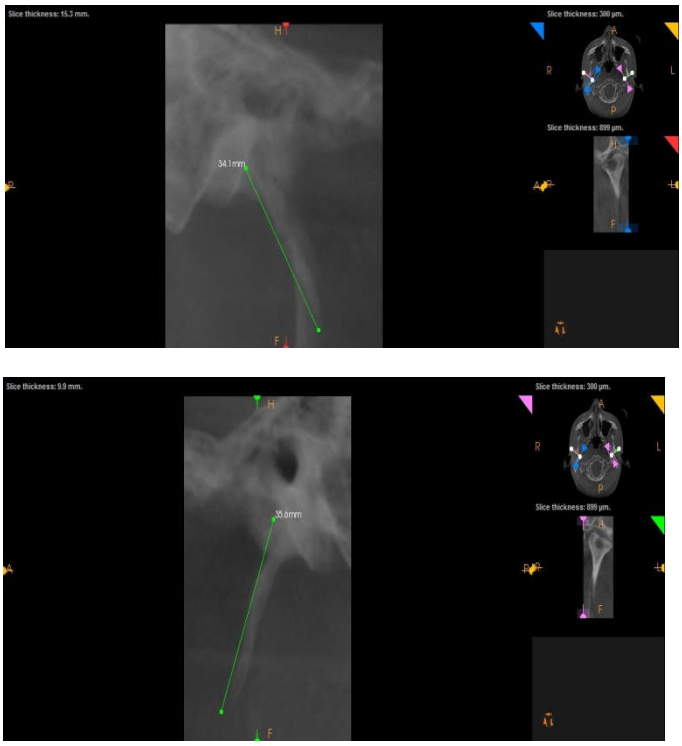

Figure 6A. The length of the styloid process when measured on the CBCT was found to be 34.1mm on the right side and FIGURE 6B styloid process measuring 35.6mm on the left side

DISCUSSION

The temporomandibular joint is a distinctive part of the human skeleton which is more vulnerable to malformation during and post development period and the zone most affected is the condylar cartilage- one of the growth centres of the mandible. Condylar hypoplasia is an uncommon malformation due to defective growth of the condyle. The etiologic heterogeneity of this condition is manifold.Since many cases of unilateral condylar hypoplasia have been reported, this case of bilateral condylar hypoplasia thus becomes something of a rarity when noticed along with the presence of elongated styloid process bilaterally.The length of the styloid process when measured on the CBCT was found to be 34.1mm on the right side and 35.6mm on the left side Figure 6. A thorough history followed by a proper clinical examination and a series of radiologic investigations helps us to formulate an appropriate treatment plan for the above condition. The main treatment goals include restoration of aesthetics and function.Surgery and orthodontics go hand in hand in the treatment for this case. Protocol should begin with alignment of the teeth with simultaneous surgical management of the condition.Surgical treatment modalities include distraction osteogenesis of the mandible or surgical reconstruction of the condyle with costochondral graft (Arun, 2002). The augmentation of the condylar unit preserves the intergrity and functionality of articulation. Literature reports that Ferri *et al*., have performed Caldwell and Letterman's vertical osteotomy (Ferri, 2006), whilst Rubio-Bueno *et al*., have performed horizontal ascending ramus osteotomy (Rubio-Bueno, 2000), to distract the mandible to the desired length.Thus, the patient is kept under observation during the course of the treatment. The most satisfactory management of elongated styloid process is surgical shortening through extraoral or intraorally.Usual treatment protocol for Eagle's syndrome involves Loeser and Caldwell approach, a classic transcervicalapproach which implies a long incision of approximately 10 cm at the superior two thirds of the anterior margin of the sternocleidomastoid muscle followed by a partial resection of the styloid process. Since the elongated styloid process bilaterally remains asymptomatic in this case, surgical intervention gains no significance.

Conclusion

Condylar hypoplasia is a temporomandibular joint disorder due to malformation of the condyles usually occurring unilaterally. Bilateral condylar hypoplasia is an uncommon entity and becomes something of a rarity when in occurrence with elongated styloid process (Eagle's syndrome) bilaterally. A keen observation of the diagnostic aids helps in spotting the incidental findings like Eagle's syndrome as in this case. Treatment protocol becomes necessary when the patient is symptomatic.

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