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Unilateral Condylar Hyperplasia- Case Series with Review of Recent Imaging Modalities

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Abstract

Background & Aim: Condylar hyperplasia (CH) is a rarity with characteristic excessive bone growth mostly presenting unilaterally, resulting in facial asymmetry. It is an uncommon malformation of the mandible involving change in size and morphology of the condylar neck and head. Classification of the different types of CH can differ depending on the authors. Although, clinical examination and history provided by the patient are key inputs for the diagnosis of this entity, advanced imaging is an essential tool in deciding the correct time and type of treatment.

Case Report: Here we present two cases of Unilateral Condylar hyperplasia with their respective scans and a review on etiology, classification and imaging modalities for the same.

Conclusion: Understanding clinical and imaging characteristics is important for treatment plan of unilateral condylar hyperplasia and appropriate diagnostic aids.

Keywords: ***Positron emission tomography; Mandibular condyle; Condylar hyperplasia***

1. Introduction

Functional difficulties, aesthetic alterations, and pain are among the common symptoms in oral and maxillofacial pathology. Facial asymmetry can originate from causes like aging, genetics, dental problems, trauma, TMJ disorders, muscular disorders, cranial nerve damage, or can be developmental in origin. Condylar hyperplasia can be seen as overdevelopment that may result in facial asymmetry, mandibular deviation, malocclusion, and joint dysfunction. It can be defined as self-limiting, non-

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neoplastic growth of one or both condyles causing an increase in bone mass of varying degrees in instances. Unilateral condylar hyperplasia (UCH) presents as unilateral excessive growth of the condyle, characterized by slowly progressing and enlarging condyle along with elongation of the mandibular body, culminating in a contralateral shift of the midline [1].

Historically, this condition was first reported by Lohamann in 1918 and Gruca and Meisels in 1926. Rushton in 1946 carried out an extensive literature review, reporting a total of 32 cases that were diagnosed and published [2]. Norman & Painter [3] conducted a historical review of CH and in 1980 published a series of cases that were previously described by Robert Adams in 1836, George Humphry in 1856, and others describing features of the disease, emphasizing the facial deformity and chin displacement as the main characteristics.

Facial analysis and imaging are the mainstays in the diagnosis of condylar hyperplasia. However, advanced imaging modalities such as CT, cone beam CT and single-photon emission CT have been advised to suggest whether the growth is active or not, which further helps in planning the treatment. MRI should be carried out when clinical and CT findings are suggestive of other diseases [4].

2. Case 1

A 22-year-old male reported to the Department of Oral Medicine and Radiology with facial asymmetry that he noticed since the last 3 years. Previously, patient underwent acupuncture for the disfigurement, suspecting Bell's palsy. Patient also noticed mild changes in his biting since the changes in his appearance. Extraoral examination revealed elongation on the right side with the lower half of the face directed towards opposite side, giving the right half of the face a longer appearance. Further flattening of face was seen on the left side, fullness over the right half of face and deviation of mandible to the left side with protrusion of chin. Intraoral examination revealed edge to edge bite anteriorly and evident shift of the mid-line shift to the side. Left side showed cross bite in relation to mandibular canine, first and second premolars. Patient did not give any history of difficulty in opening mouth. The case was provisionally diagnosed as condylar hyperplasia based on the history and clinical examination. (FIG. 1).

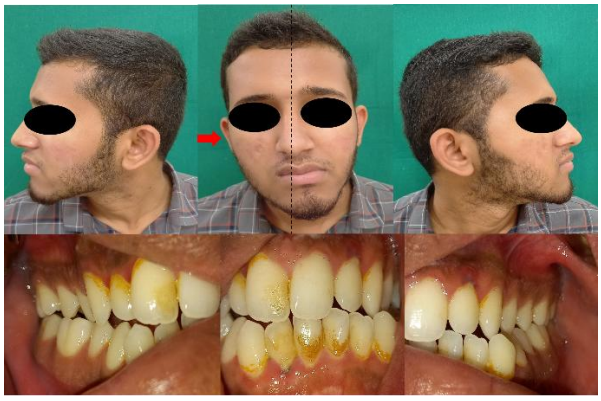


FIG.1. **Clinical features.**

Radiographic investigations (FIG. 2) were conducted to evaluate the osseous changes. OPG reveals elongated condylar process on the right side with a deep sigmoid notch. Height of mandibular body and length of ascending ramus appears same bilaterally.

Postero-anterior view revealed elongation of the right condylar head causing a shift in the midline. It further revealed a downward overgrowth/enlargement with the maxillary right alveolar ridge resulting in, shift in the maxillary occlusal plane. Lateral cephalogram revealed convex to straight soft tissue as well as hard tissue profile. Based on the radiographic findings, the case was finally diagnosed as unilateral CH of the right side.

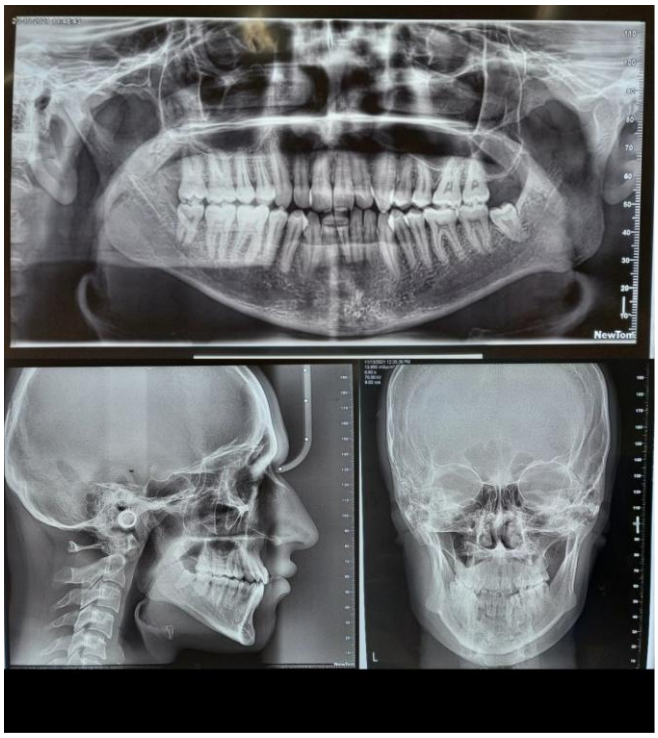


FIG.2. **OPG, Postero-anterior radiograph & lateral cephalogram.**

Since the patient was 22 years old, a PET scan was advised to assess the active growth component in the condyles and plan an appropriate multi-disciplinary intervention. Increased radiotracer uptake was noted in the right condylar head suggestive of active growth component. (FIG. 3) Patient was informed about the need to delay treatment and periodically assessed to determine the suitable time of treatment.

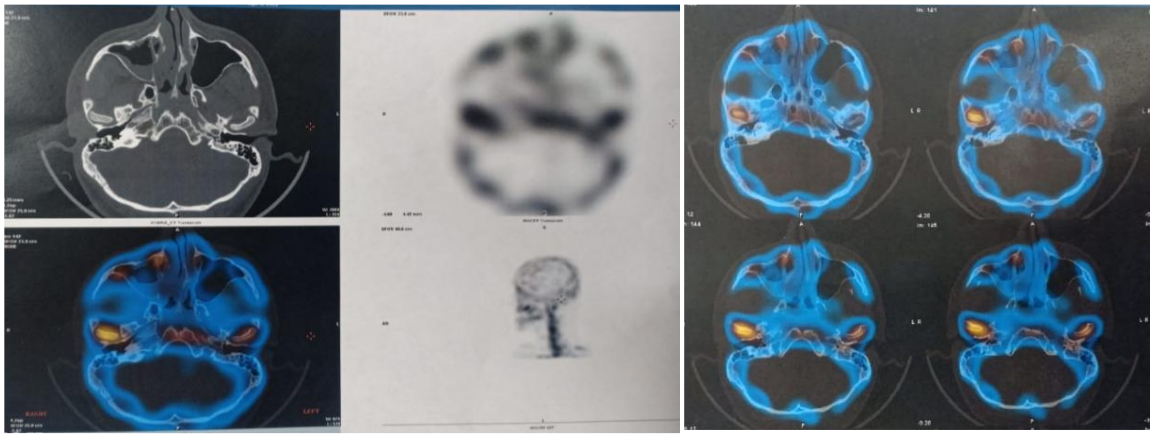


FIG. 3. **PET Scan.**

3. Case 2

A 25-year-old female reported to the Department of Oral Medicine and Radiology with facial asymmetry that she noticed since she was 13 years of age. Patient reports alleged fall from train in 2017 during which she fell on her chin. Patient was taken to a local hospital where primary management of contusion and laceration wound was done. Since there was no occlusal derangement and no complains in function, no radiographs were taken. Currently the patient reported to the hospital for management of facial asymmetry. No complains of pain and difficulty in function. On clinical examination, there was a gross facial asymmetry. Chin appeared evidently deviated to the left side. Flattening of the face was noted on the left side and fullness was noted in the appearance of right side of face. Middle third of face appeared depressed and mandible appears prognathic and deviated to the left side. On palpation, right condylar movements were not palpable. Intra-orally, open bite was noted in left side from lateral incisor to first molar region. Midline shift was noted and occlusal plane was tilted downwards on the left side (FIG. 4). Based on the clinical findings right TMJ ankylosis was considered as the provisional diagnosis.

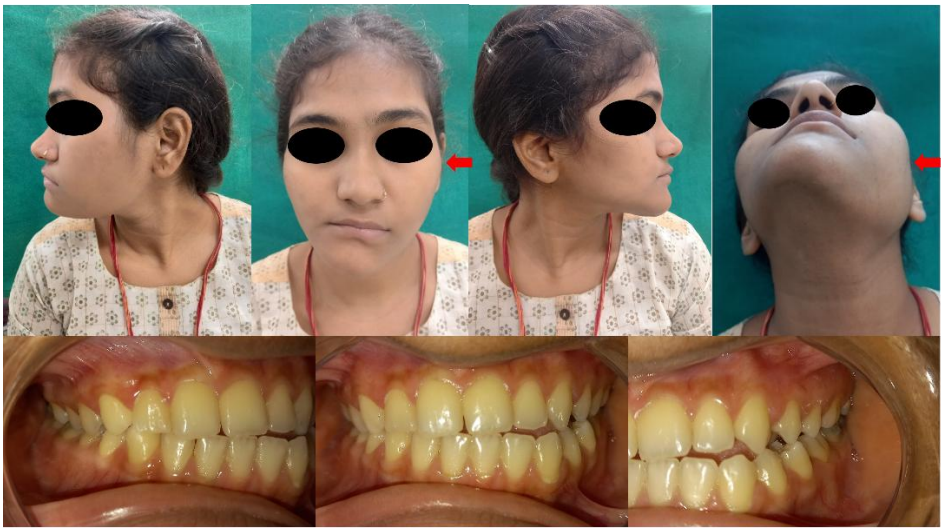


FIG. 4. **Clinical features.**

OPG was taken for initial radiographic evaluation, which shows an evident asymmetry between the right and left mandible (FIG. 5). Although the borders of the right condylar head were not clearly traceable, it appeared smaller in dimensions. The condylar neck region on the left side appears thinner. The height of right body of mandible appears less in comparison to left mandibular body with a prominent antegonial notch on the same side. The height of alveolar bone in right maxillary region appears less in comparison to the left side. The superior surface of the left condylar head was not clearly traceable, and the head appeared larger in dimensions in comparison to the normal. Deep and prominent sigmoid notch was noted on the left side. Intra-articular joint space was not clearly traceable on both the sides. Similarly, left condylar head was clearly enlarged as noted on CT scan, with increase in the bulk of cancellous bone and surrounded by thin layer of cortication (FIG. 6).

A PET scan was advised to assess the active growth component in the condyles before planning a multi-disciplinary intervention. Increased radiotracer uptake was noted in the left condylar head suggestive of active growth component. The need for delaying the surgical intervention was explained to the patient and periodic recall was suggested to direct the suitable time of treatment.

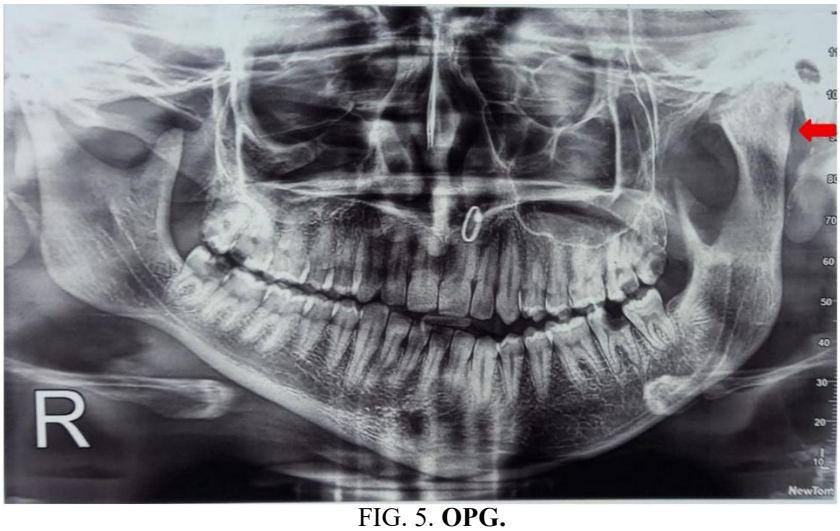


FIG. 5. **OPG.**

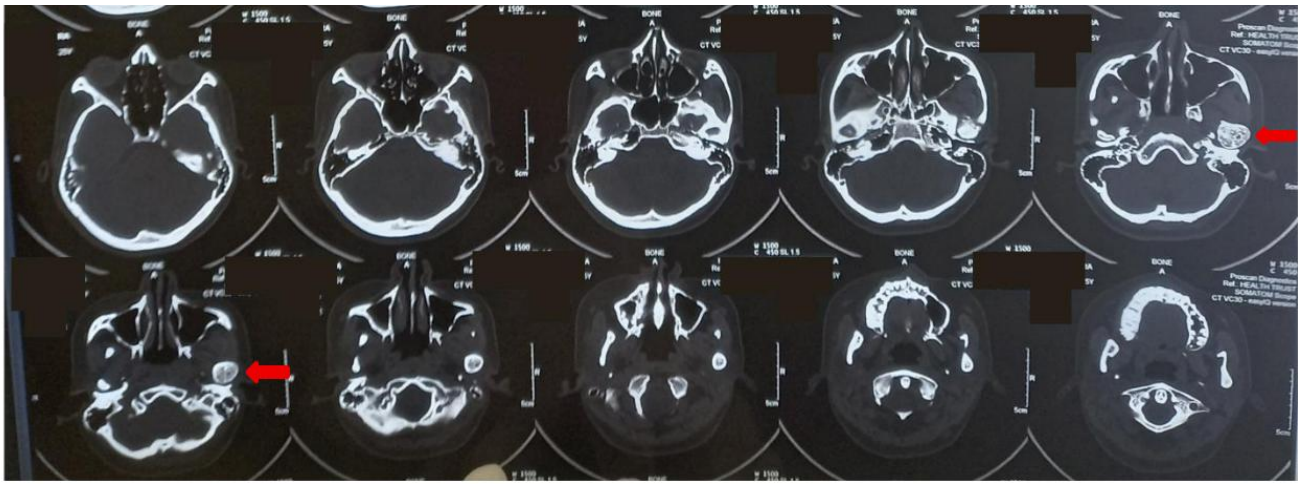


FIG. 6. **CT SCAN.**

4. Discussion

Unilateral condylar hyperplasia is a rare pathology, with an unknown etiology and is characterized by a progressive, independent growth, causing greater bone volume of one condyle over the other side [5]. Condylar hyperplasia can occur at any age but skewing in prevalence is noted towards adolescent years of age and the growth has been seen to continue beyond developing age. Sparse studies previously suggested an equal gender predilection, however a meta-analysis conducted in 2012 revealed that women develop CH significantly more frequently. Variations in the qualitative and quantitative effects of hormones are proposed to be responsible in the development of UCH, however there is no consensus in this regard [6]. Apart from this, research does not suggest any side predilection.

4.1 Etiology

Facial asymmetry can have varied causes and can be either be progressive or self-limiting based on the etiology and tissue of origin. Post natal facial asymmetries are most commonly due to TMJ and especially condylar pathologies such as

osteochondromas, condylar resorptions, infection-related growth deficiencies, trauma or CH. Condylar growth abnormalities can be prenatal or post-natal. One way to recognize facial asymmetries caused by condylar growth alterations in prenatal or postnatal origin is that in the latter cranial asymmetries are not observed [7]. Cranial asymmetry deformity is caused by alterations before 5 years of age, when the cranial base takes to form completely [8], placing CH in the postnatal category.

Though the etiology of CH has always been a point of discussion, local circulatory problems, endocrine disturbances, traumatic lesions, and arthrosis are the suggested etiologic factors of this pathosis [9,10].

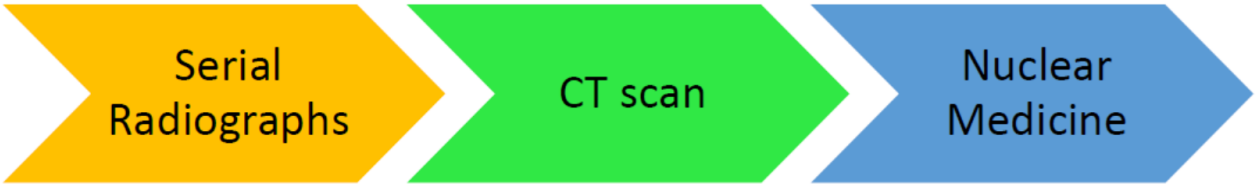
4.2 Classification

Condylar hyperplasia (CH) refers to a range of conditions characterized by abnormal and excessive growth of the mandibular condyle, resulting from various underlying causes. These abnormalities can significantly impact the mandible's size and shape, alter occlusion patterns, and indirectly affect the maxilla. This can lead to or exacerbate dentofacial deformities, such as mandibular prognathism, unilateral overgrowth of the condyle, neck, ramus, and body, as well as facial asymmetry, malocclusion, and pain. Proper identification of the specific type of CH is essential for understanding its progression in the absence of treatment, recognizing the clinical, radiographic, and histological features, and applying appropriate treatment strategies to address the condition, ensuring functional and esthetic improvements.

Originally, Obwegeser and Makek provided a clinical classification dividing the condition into two types namely, hemimandibular hyperplasia (3D enlargement of one side of mandible including condylar head and neck and ramus) and hemimandibular elongation (horizontal displacement towards unaffected side) [10]. Arora et al suggested an update in this classification, with the addition of a third category i.e. a blend of the two existing types with clinical features such as enlarged mandibular condyle, condylar neck and ramus of the mandible of the affected side, and in addition there is horizontal displacement of mandible and the chin to the contralateral side along with open bite on the affected side, which ultimately leads to flattening of face on the opposite side and fullness on the affected side [2].

Wolford et al introduced a classification differentiating horizontal and vertical growth vectors that are usually related to specific but different mandibular condylar pathologies. The classification proposed 4 basic categories of CH in relation to the clinical, imaging, growth, and histologic characteristics, effects on the jaws and facial structures, and treatment protocols that provide optimal treatment outcomes. Slootweg and Muller suggested a histological classification that differentiates condylar growth based on cellular characteristics of the tissues in the condylar complex [9].

4.3 Investigations



Although the diagnosis of condylar hyperplasia is mainly clinical, imaging modalities aid in recognizing the altered morphology and also determine bone activity.

Conventional Radiography: Before advanced and functional imageology, serial 2D radiographs were part of follow-up, that were compared at 6-month intervals to assess condylar growth activity. Computed tomography: Computed tomography (CT) has contributed to analyze the pathology, enabling recognition and classification of the degree of the disease providing a clear picture of the extent and involvement of osseous lesions.

With emerging trends in imaging, molecular or functional imaging techniques have been developed that enable to assess biological domain of pathologies. Therefore, it helps in determining the extent/severity of disease, selection of therapy, efficacy of a particular regimen, assessment of disease progression, and identification of recurrence. In cases of condylar hyperplasia, nuclear studies are considered for prompt diagnosis.

Bone scanning is a non-invasive technique to evaluate whether the hyperplastic growth is still active; commonly 99 technetium phosphate is used [11]. With the use of hybrid tests, nuclear medicine increases the diagnostic accuracy by 30% in osseous conditions including tumour detection and inflammatory processes, and providing a more accurate description of the disease location [12]. This inference is based on bone metabolic activity in condyles assessed by the comparative lateral uptake of 99mTc-MPD. Suh et al. suggested the need of a standardized value for the radiopharmaceutical uptake and the CT data to evaluate temporomandibular disorders [13].

SPECT aids in monitoring active and inactive growth patterns, and identifies the confines of the overgrowth, whether it is confined to the condyle region or extending to the whole mandible [14]. In comparison to bone scanning, the radioactive uptake in bone SPECT depends on the regional blood circulation and the absorption by the structure of hydroxyapatite crystals. The areas with tracer high are correlated with hyperemia and more metabolic bone activity. Since nuclear medicine helps identify activity at molecular level, it is highly sensitive for early detection of lesions, very much earlier than X-ray or tomographic images. Bone SPECT has been validated for early diagnosis of UCH, though occurring during active growth and development, it may be self-limited and expressed only by sequelae of the pathology [15,16]. Although the diagnosis is strictly clinical, the evaluation of bone metabolism by SPECT aids in differentiating the active/inactive stages [17].

Furthermore, Lopez suggested that fusion of functional SPECT scans and anatomic CT images improves the precision and specificity of the diagnostic tests and, consequently, allows better therapeutic decisions [18]. Image fusion for diagnostic purposes, as in the case of SPECT/CT, is known as a co-register or hybrid technique and it is used to improve the diagnostic precision and, therefore, to aid in the development of a better treatment plan positively determined by the prognosis [19].

Fokoue et al. suggested that the diagnostic accuracy of image fusion [SPECT/CT] is superior to SPECT alone in detection the hyperplasic area [20]. In a study, Agarwal et al. evaluated the diagnostic improvement obtained by the SPECT/TCT fusion compared with SPECT alone, which is more sensitive (80%), but SPECT/CT is more specific (100%) and accurate (85.5%), while planar scintigraphy had the lowest diagnostic performance [21]. On the contrary, Theerakulpisut et al., in a study of 61 scintigraphies, concluded that the diagnostic specificity is not improved by fused tests and, since the radiation is increased, did

not recommend its use [22]. Additionally, Verhelst et al. reported that the anatomic changes detected by CT in the hybrid test are evident only in 50% of the patients, adding a minimum benefit, and Liu et al. concluded that ROI delimitation in the drawing of condylar outline was not superior when SPECT/CT was used [23,24].

4.4 Treatment

Management of unilateral condylar hyperplasia is directed at stopping abnormal growth and to correct the already established facial asymmetry. The plan of action depends of the growth phase of the condyle. In active hyperplasia, many clinicians support the "wait and watch" conservative method, with the explanation of avoiding multiple surgeries in case of relapse. However, in active growth with severe deformity that affects esthetic and functions, high condylectomy can be performed in order to eliminate the growth center, which is followed by appropriate orthognathic surgery for skeletal correction.

In cases of adults, where the growth of the condylar head is established, orthognathic surgery is the ultimate option along with dento-alveolar alignment to restore esthetics and function. Orthodontic camouflage is an alternative approach that can be considered in minor skeletal deformities [25].

5. Conclusion

Clinical examination, radiography, planar scintigraphy, SPECT, and PET are diagnostic aids that can be utilized by the clinicians when planning surgery. Continued research, oriented to know the causes of CH and to establish a more standardized approach for diagnosing the activity of CH, is the key. Longer follow-up studies can aid in determining the prognosis of various treatment protocols.

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