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Case Report

Orthognathic Treatment for a Case of Unilateral Condylar Hyperactivity

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Abstract

Unilateral condylar hyperplasia can cause facial asymmetry, occlusal disturbance, functional and aesthetic problems. In order to better correct these problems, orthognathic treatment is the only option. Mostly, the surgery of excessive growth of one condyle includes 2-jaw orthognathic surgery, condyle osteotomy and genioplasty. In this case, a 20-year-old male patient affected by right condylar hyperplasia underwent a surgical treatment with high unilateral condylectomy associated to 2-jaw orthognathic surgery and genioplasty, as well as orthodontic treatment before and after surgery. Because of serious skeletal problems, the partial malocclusion were adjustmented after surgery, it is better to maintain periodontal health and more effective. Detailed digital diagnosis and careful clinical management throughout the treatment course can help to provide satisfactory aesthetic and functional outcomes after follow-up 24 months.

Keywords: *Unilateral Condylar Hyperplasia; Orthognathic Treatment; Asymmetry; Stability*

Introduction

Condylar hyperplasia (CH) is defined by excessive growth of one condyle (rarely bilateral) [1]. Unilateral condylar hyperplasia (UCH) is a rare condition of unknown etiology causing the progressive unilateral enlargement of the mandible, and deviation of the chin towards the unaffected sides [2,3]. Although UCH is selflimiting, it may be active after completion of normal growth [1], the excessive unilateral growth of the mandibular condyle can indeed lead to facial asymmetry, occlusal disturbance, functional and aesthetic problems [1,2]. UCH has rarely joint dysfunction, but a study showed that the progressive excessive growth of one condyle may cause symptoms of pain and articular dysfunction [1]. UCH is one of the main postnatal causes of facial asymmetry [4], the precise etiology of this growth anomaly, diagnosis and efficacy stability of UCH are still not entirely clear. Here, we will review the contents related to UCH and a characteristic case of UCH.

Epidemiology and etiology

UCH described for the first time in 1836 by Adams [5]. UCH also known as condylar hyperplasia, is characterised by growth resembling increased activity in one of the mandibular condyles [6]. In some studies, UCH usually occurs in the second or third decade of age, near or shortly after completion of the growth period. Normally, the active formation of cartilage gradually stops around the age of 20 years, the UCH active form is found most frequently in 11-year-old to 25-year-old patients [1,7,8]. UCH seems to have a similar incidence in both sexes as well as in different ethnic groups [2] however, in the study by Agarwal., *et al*. [1] expressed that UCH may be present in passive form as clinical sequelae, a higher prevalence has been found in women than in men. UCH can be due to hypo-development, hyper-development or can be recognised a "mixed form", growth of the condylar cartilage contributes to the increase of the mandibular ramus in height and to the increase of the overall length of the mandible, any interference which will affect the growth centre will result ill mandibular deformity [3,9].

The hypothesis that the cause in at least some patients with UCH is a somatic mutation in a gene that controls cell growth could not be confirmed [10]. The etiology of UCH has been related to genetic factors, congenital facial hemihypertrophy, trauma, infections, neoplasms, tumors, or hormonal conditions and circulatory problems [4,11,12]. However, the etiology of UCH excludes vascular and hormonal dysfunctions such as lymphomatosis, angiomatosis, acromegalia and Sturge-Weber syndrome. Nolte [10] found that condylar tissue and analysed eight overgrowth genes (AKT1, AKT3, MTOR, PIK3CA, PIK3R2, PTEN, TSC1, TSC2), but no mutation was detected in any of the overgrowth genes, and untargeted exome sequencing failed to detect any definitively causative variant in any other gene. UCH present in only one of a pair of monozygotic twins has been reported, implicating an environmental factor although the *denovo*-occurrence of a mutation after splitting of the fertilized egg cell into the twins could also explain this [10,13]. Obwegeser postulated the presence of two growth regulators in the condylar cartilaginous layer: the "M" (mass) stimulates the bone mass and the volumetric expansion of the condylar centres of growth, while the "L" (length) regulator stimulates the length. Under normal conditions or in a physiological growth phase, the two regulators are well balanced during their activities, and cease their activity at the end of the growth [14]. A variety of possible causes (inflammation, trauma, hormonal imbalance, hypervascularity) has been reported, but firm proof of causation is lacking [10]. UCH patients with asymmetric mandibular growth consisted of different subgroups, each with a different pathogenesis [7] and many literatures predominantly focused on clinical descriptions and not directed to detecting the cause of UCH [10] therefore, the aetiology is not so clear about UCH.

Classification

UCH can involve the upper jaw, the lower jaw or both, and develop on the horizontal plane, on the vertical plane or may affect both planes [9]. Different studies have different classifications, and clinically the overgrowth of mandibular condylar has been classified in various ways. The most commonly used subdivision is hemi-mandibular elongation and hemi-mandibular hyperplasia, or a combination of the two [15] others prefer to classify just the direction of asymmetry, such as vertical or transverse [16]. Different growth factors must exist, one for length and one for volume [14] examples of cranial base deformities are muscular torticollis, unilateral coronal craniosynostosis, and deformational plagiocephaly [4]. Condylar fracture, condylar hyperactivity, juvenile condylar arthritis, and hemifacial microsomia are examples of an asymmetry based on a condylar abnormality [17].

Obwegeser and Makek [15] classified the asymmetry associated with UCH into 3 categories. Type 1: hemimandibular elongation, with a horizontal growth vector (Figure 1). Type 2: hemimandibular hyperplasia, with a vertical growth vector (Figure 1); Type 3: a combination of type 1 and type 2. The condylar head and neck were lengthened in type 1, which is associated with a deviation of the chin toward the contralateral side and a mandibular midline deviation to the unaffected side. In contrast, in type 2, the condylar head and neck are longer and larger in diameter than normal. Type 2 is characterized by an ipsilateral open bite or compensatory vertical overdevelopment of the maxilla on the ipsilateral side with canting of the occlusal plane. Most commonly, there is no deviation of the mandibular midline with minimal deviation of the chin. The third type is a combination of types 1 and 2. Moreover, Wolford [18] established a classification based

on histology, clinical characteristics and images, effects on the maxilla and facial structures, and frequency, defining 4 types, with type 1A being the most frequent form and type 4 the least (Table 1) [15].

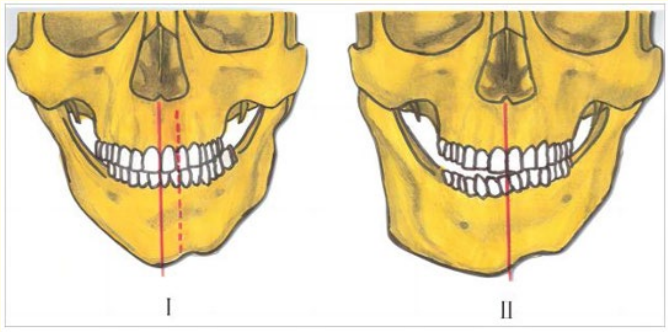

Figure 1: *Types of UCH attending Obwegeser and Makek's classification.*

Type I	Hemimandibular elongation, with a horizontal growth vector
Type II	Hemimandibular hyperplasia, with a vertical growth vector
Type III	Combination of both type I and II

Table 1: *Obwegeser and Makek classification.*

Diagnosis

An understanding of the normal growth of the mandible is the basis for early recognition and proper surgical treatment [3]. The differential diagnosis of an asymmetric mandible can be subdivided into 2 major categories: cranial base deformations and condylar abnormalities [4]. The diagnostic clinical impression of UCH is made from a combination of facial, intraoral, radiographic, or tomographic findings, but bone scintigraphy is the current gold standard to assess bone hyperactivity in condyles [19]. Single photon emission computed tomography (SPECT) is commonly used to evaluate the metabolic activity in several types of tissue, and it is considered to provide functional and morphological information about the mandibular condyle growth [20]. Moreover, there is a study to find that SPECT/CT is superior to planar BS and SPECT for the diagnosis of active CH, ^{99m}Tc-MDP hybrid SPECT/CT showed high diagnostic accuracy for the diagnosis of active CH [1]. In a normal population and in patients without progressive asymmetry there is only a limited difference between the left and right condylar bone activity. Most studies used a maximum difference in bone activity of 10% as the cut-off for differentiating between normal and abnormal condylar activity [21].

The diagnostic is confirmed only by histopathology, when the patient is subject to surgical intervention [19] but there are studies found that histopathologic analysis of a condyle does not confirm or exclude condylar hyperactivity, because currently, there is no consensus in the literature concerning UCH histopathology, Nevertheless, histopathologic examination should always be performed to rule out other diseases [7,22] moreover the radiological findings do not always correlate with the clinical symptoms [5]. A study found that blood flow measurements in the condylar region by using positron emission tomography scans seemed to rule out hypervascularization as a cause for condylar hyperactivity [4].

The early diagnosis of UCH is important because it may reduce further structural damage by early treatment, however, treatment protocols are different according to the patient's age, severity of the resulting asymmetry, and the active or passive status of the pathology

[1]. The diagnosis of UCH is made from a combination of clinical history, physical examination, and condylar bone scintigraphy [20]. So each individual patient with UCH needs to be checked carefully, for instance, by taking the objective elements (bone scan and clinical growth) and subjective elements (patient preferences) into consideration, and to choose the appropriate surgical technique, to obtain a more satisfactory effect.

Orthognathic treatment and stability

Unilateral condylar excessive activity results in asymmetrical development of the mandible, which leads to asymmetric facial deformities such as jaw deviation and malocclusion [6,19]. Orthognathic treatment is the only option of UCH patients. For the precocious condylectomy, some authors suggested patients are treated, either a high condylectomy with sole resection of the growth cartilage [12] In clinic, 4 different strategies for correction in UCH can be defined. Method 1 includes no joint surgery, but corrective orthognathic surgery is performed (end-stage). Method 2 includes a high partial condylectomy, followed by a wait- and- see for 6 months after which corrective surgery is performed (delayed stage). In method 3 a proportional condylectomy is carried out, with the aim of no further corrective orthognathic surgery (no stage), and method 4 encompasses combined condylectomy and corrective orthognathic surgery (one stage) [23]. Di Blasio., *et al.* [24] found the young patients up to 18 years, that after condylectomy, the healthy side continues to grow in a normal way, and the affected side first ceases growth after condylectomy, but then recovers to normal growth again. Xavier., *et al*. [25] reported a two-stage (delayed stage) treatment, and found the noticeable improvements in occlusion after the first stage (condylectomy). Moreover, Farina., *et al*. also found the condylectomy as a sole treatment (non-stage) reduced the need for secondary orthognathic procedures [26].

Although UCH is self-limiting, the progressive excessive growth of one condyle may compromise the neck, mandibular ramus, and body, and it causes symptoms of pain and articular dysfunction [19] so UCH will have a great psychological impact on the patients. Clearly, the choice of operation time is very important to relieve the psychological impact for UCH patients. However, the optimal surgery timing of UCH are largely dependent on the growth activity of the mandibular condyles [2]. A high mandibular condylectomy may arrest the progress of the condition for patients with clinically progressive mandibular asymmetry, removal of the superior portion of the condyle will stop the progression of the asymmetry and contribute to stable long-term results [6]. Although the active growth zone be removed can prevent relapse, UCH bone growth ceases spontaneously, surgical correction of the mandibular asymmetry yields stable results [2,22].

It is focused on the postoperative stability of the TMJ after surgical correcting the deformity of an active condylar growth. Postoperative stability of the TMJ is assessed clinically by using various parameters such as pain, free mouth opening, lateral and protrusive movements, presence or absence of clicking in the joints, bite force analysis etc [27]. The results that unilateral condylar fracture were underwent open reduction and internal fixation (ORIF), and clinically asymptomatic at six months after surgery showed that ORIF as a treatment modality, can help to obtain satisfactory occlusion earlier and minimizes restitutional remodelling of TMJ after condylar fractures [27]. The experienced pain is associated with the higher prevalence of osteoarthrosis, the TMJ pain may be possible not only that this is caused by the surgical procedure but that the underlying pathology (of UCH) can also be a contributing factor [22].

Case Report

At present, UCH is rare in clinic. This case of a male patient aged 20 was transferred by orthognathic surgery of Peking University to our hospital for preoperative treatment. He started at the age of 12 when the right mandible was noticed to have increased in size, leading to marked facial deformity and typical malocclusion of the teeth. At the peak of growth, the parents agreed to follow up for observation. At 18-year-old, examination revealed a considerably enlarged right mandible and chin shifted to left side about 4 mm (Figure 2). Intraoral photos showed that the left and right molars are the mesial relationship, an open bite on the affected side (the right side, 17 and 47 crossbite, II° deepbite,I° crowning, mandibular midlining shifted to left side about 2 mm (Figure 2). X-ray examination revealed a considerably enlarged right mandible condyle and chin, bilateral joint asymmetry and skeletal class III malocclusion (Figure 3). This

patient had click on the both sides, a pain on the right side, opening type was abnormal before treatment. In addition, he had a bad habit of mouth breathing. The patient with an open bite on the right or compensatory vertical overdevelopment of the maxilla on the ipsilateral side with canting of the occlusal plane, moreover, and with a deviation of the chin toward the contralateral side and a mandibular midline deviation to the unaffected side, so the patient was considered type 3. According to age, esthetic compromise, severity of the malocclusion, and level of articular dysfunction etc., the orthodontist and the maxillofacial surgeon together made a thorough diagnosis and treatment plan. He was treated with 2-jaw orthognathic surgery, condyle osteotomy and genioplasty. This condyle osteotomy was performed through an intraoral incision, and the patient had less surgical invasion. See figure 4-7 for operate detail. Because skeletal problems of the patient was serious, it is difficult to decompensate and orthodontic treatment before surgery took nearly two years, so we suggest that postoperative adjustment of partial malocclusion is more favorable for maintaining periodontal health and stable efficacy. The post-operative course was uneventful and he gained a reasonably good bite and good masticatory function in terms of debonding (Figure 8-11).

Figure 2: *Facial and intraoral photos.*

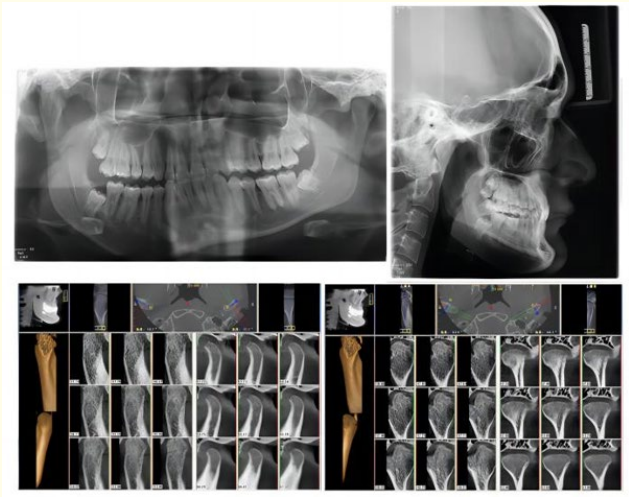

Figure 3: *Pretreatment radiographs.*

Figure 4: *Treatment details before operation.*

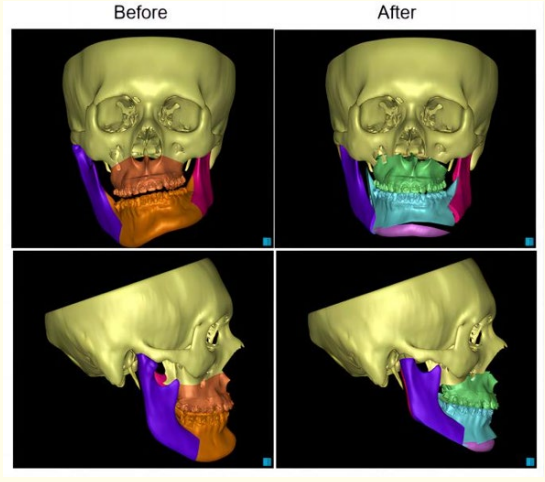

Figure 5: *Surgical digital simulation.*

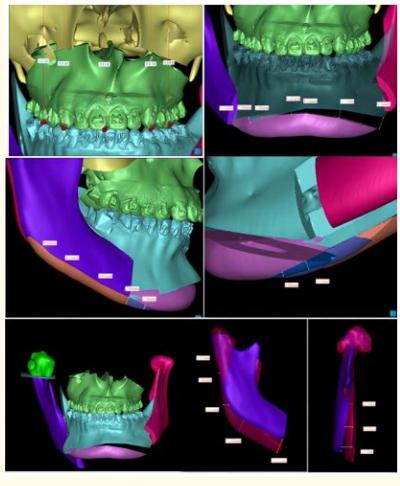

Figure 6: *Accurate simulation of 2-jaw orthognathic surgery and high unilateral condylectomy.*

Figure 7: *Facial and intraoral photographs immediately after orthognathic surgery.*

Figure 8: *Treatment details after operation.*

Figure 9: *Facial photos retention 2 years.*

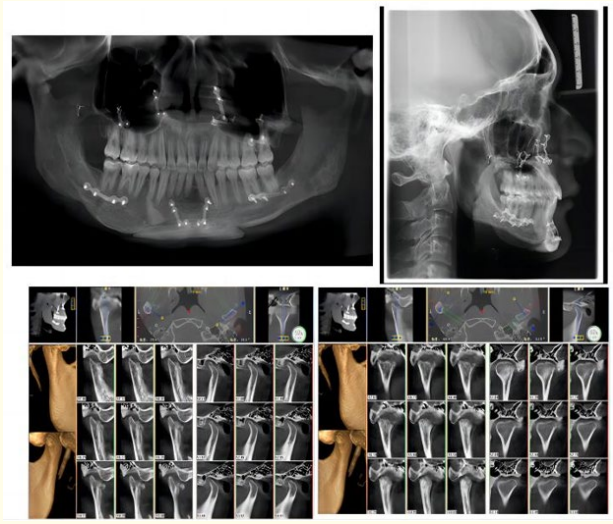

Figure 10: *X-ray examination after operation.*

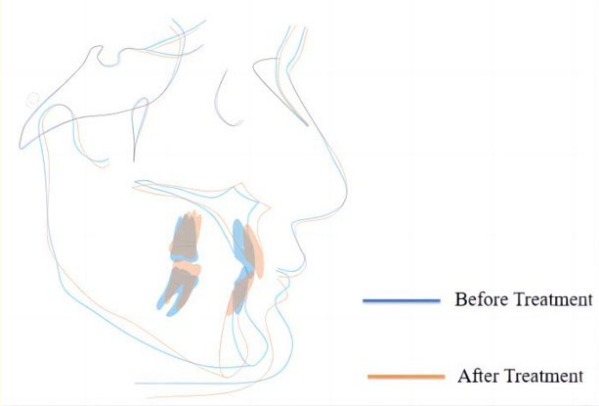

Figure 11: *Overlapping images of lateral cephalic films before and after treatment.*

The cosmetic result was satisfactory, the condyle of the affected side was not enlarged. A Study by Saridin found that the patients with UCH who underwent condylectomy had more joint-related temporomandibular problems, as well as higher postoperative pain [22] however, the pain of this patient undetected during the 2-year follow-up period. Although the study performed by Karssemakers., *et al*. [4] showed that a high condylectomy of the UCH patients may be indicated to arrest the progression of the condition, but the right condylar of this case of was not seen more changes for TMJ and masticatory function in follow-up 2 years, probably because of UCH self-limiting or the condyle stopped the mandible growth in the hyperactive side [4]. The development of condylar hyperactivity mainly involves the inactive cartilage remnants in the articular surface, which can be stimulated to become active once more, so the stable long-term results need continue to observe.

Conclusion

- Orthognathic treatment is the only option of UCH patients, and the combination of multi-disciplinary is also important during the treatment of UCH patients.
- Not need to full compensate of the teeth for serious skeletal problems before surgery. Postoperative adjustment of partial malocclusion maybe more favorable for maintaining Periodontal health of UCH patients.
- In clinic, only accurate diagnosis can have the functional, stable and a better prognosis results, either UCH patients or others.

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