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Unilateral Condylar Hyperplasia: Is Reactivation After a Long Latency Period of Inactivity Plausible?

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Abstract: Unilateral condylar hyperplasia (UCH) is characterized by an overgrowth of the mandibular condyle responsible for a facial and dental asymmetry associated with temporomandibular joint function and maxillary growth consequences. The diagnosis is based on a body of clinical, radiological and histological arguments. A 38 years old woman with a reactivation of UCH after a latency period of 16 years following an orthognathic surgery performed for facial asymmetry normalization is presented. She was addressed to our department for a facial progressive asymmetry relapse and dental prosthetic consequences.

The radiological images and the planar scintigraphy combined with single-photon emission computed tomography scans showed an active left unilateral condylar hyperplasia. A left proportional condylectomy was performed.

The case presented highlights the possibility for the UCH to be reactivated after a long period of latency, leading to a relapse of the occlusal and facial disorders and so advocates the need for first condylectomy or at least a long-term follow-up if condylectomy is not performed as a first-line treatment.

Key Words: Condylectomy, facial asymmetry, reactivation, unilateral condylar hyperplasia

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Unilateral condylar hyperplasia (UCH) is a benign pathology involving mandibular condyle. Nosologic, etiopathogenic, and therapeutic issues are still discussed. Epidemiological data indicate a female predominance suggesting estrogenic influence in UCH.¹ The condition is characterized by an asymmetry of the lower part of the face due to an abnormal growth activity in one of the mandibular condyles, which also affects dental occlusion, temporomandibular joint (TMJ) function and indirectly maxillary growth.

Obwegeser and Malek classified UCH into hemimandibular elongation, hemimandibular hyperplasia and a third group made up of combination of both.² Delaire suggested another classification based on dynamic overgrowth directions in which 3 types of UCH are also described: a vertical form (type 1), a horizontal form (type 2), and a combination of both (type 3).³ More recently, Wolford presented a classification system based on clinical, radiological and histological analysis, which described various types of condylar hyperplasia. In fact, this classification is more related to asymmetry than to real UCH.⁴

Diagnosis of UCH is based on clinical, radiological and histological features. Planar scintigraphy combined with single-photon emission computed tomography (SPECT) scans highlights the bone metabolic activity in the condyle characterizing 2 forms of UCH: active or inactive.⁵

Treatment of UCH is based on a multidisciplinary approach with maxillofacial surgeons and orthodontists. If the evidence of scintigraphic hyperactivity typically requires a precocious condylectomy, which is supposed to avoid the progression of the occlusal plane tilting and dentofacial adaptive deformities, the management of non-active forms is still debated. The way, as its timing, to perform condylectomy is also still discussed, either a high condylectomy with sole resection of the growth cartilage,⁶ or a low proportional condylectomy which allows the adjustment of the posterior vertical excess at the same time.⁷ Condylectomy can be performed as a single method or associated with secondary orthognathic surgery.⁸

The presented case highlighted the possibility of reactivation of UCH after a long latency period of inactivity and the necessity of a long-term follow up when condylectomy has not been realized as the first-line treatment.

CLINICAL PRESENTATION

This case concerns a 38 years-old woman addressed to our department for an asymmetrical long face syndrome. She had an orthognathic surgery 16 years ago for facial asymmetry performed outside our department; planar scintigraphy realized at this point revealed no condylar hypermetabolism and it was decided to perform a mandibular sagittal split osteotomy with a reduction genioplasty to normalize the lower third of the face (Fig. 1A and B).

During the examination, the patient had revealed a facial asymmetry relapse with a very progressive aggravation for 2 years. She also expressed a concurrent left mandibular dental prosthetic instability. Clinically, she was characterized by the deviation of her chin on right side and a slight increase of the vertical dimension of the face on the left affected side (Fig. 2A and B). She presented a tilt of the occlusal plane and a slight right side displacement of the midline of mandibular dental arch without right side cross bite (Fig. 2C). Clicking was self-reported in the right temporomandibular joint.

The 3D reconstruction tomodensitometry highlighted the condylar asymmetry with an 11×22 mm left condyle and a 7×15 mm right condyle, each mandibular ramus was measured,

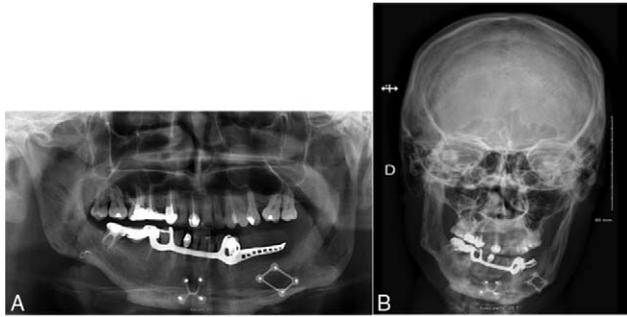


FIGURE 1. A. Orthopantomogram showing the osteosynthesis plates of the previous sagittal split osteotomy and genioplasty. Note the increased size of the left condylar unit and ramus. B. Facial radiograph showing the osteosynthesis plates of the previous sagittal split osteotomy and genioplasty. Note the facial asymmetry.

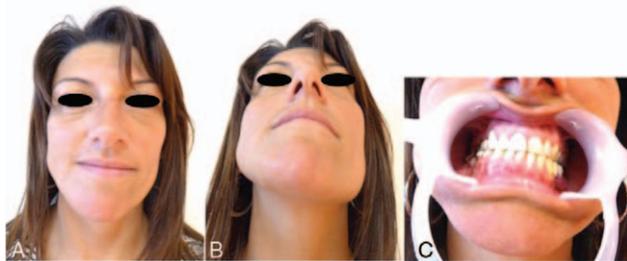


FIGURE 2. A. Facial photograph highlighting the facial asymmetry with chin deviation on the right side and the increased posterior facial height on the left side. B. Facial photograph showing the deformity of the inferior border of the mandible and the lower third facial contour asymmetry. C. Photograph of the dental occlusion. Note the occlusal plane tilt.

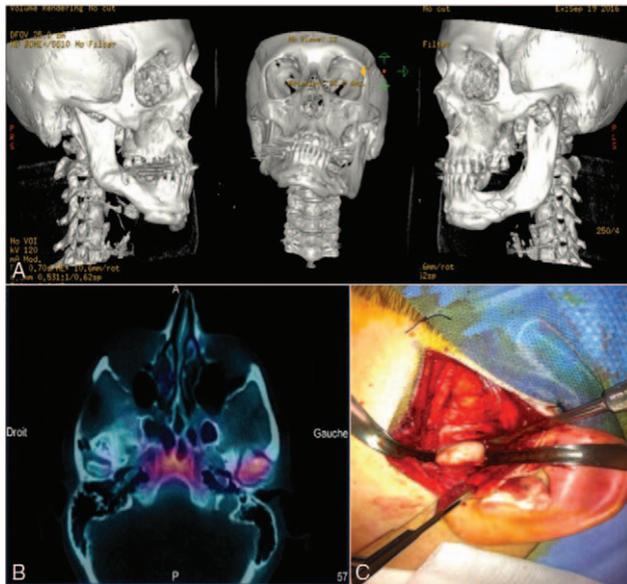


FIGURE 3. A. 3D reconstruction tomodensitometry images showing the condylar asymmetry and allowing us to measure precisely the height of the 2 condyles. B. Planar scintigraphy combined with SPECT image highlighting the left unilateral condylar hyperactivity. C. Intraoperative image of the left proportional condylectomy by preauricular approach. SPECT, single-photon emission computed tomography.



FIGURE 4. A. Postoperative facial photograph. 1 year follow up. B. Postoperative facial photograph. 1 year follow up. Note the lower third facial contour improvement. C. Postoperative photograph of the dental occlusion. 1 year follow up. D. Postoperative orthopantomogram. 1 year follow up. Note the condylar remodeling. E. Postoperative facial radiograph. 1 year follow up.

the height of the left side was evaluated at 79 mm and the right one at 64 mm (Fig. 3A). The planar scintigraphy combined with SPECT imaging showed left unilateral condylar hyperactivity compatible with an active left type 1 UCH (Fig. 3B). It was decided to perform a left proportional condylectomy (Fig. 3C). The histological exam performed with an hematoxylin and eosin staining confirmed the diagnosis of condylar hyperplasia, especially revealed substantially thicker proliferative and hypertrophic layer of the condylar cartilage with an increased prechondroblastic cells count.

She was treated during 3 months postoperatively with a maxilla-mandibular elastic therapy and then a new left mandibular prosthesis was realized. After a 1 year postoperative follow-up, the occlusal and morphoesthetic result is remarkably stable (Fig. 4A-E).

DISCUSSION

Discreet facial asymmetry is normal; if markedly obvious, individuals will seek treatment for the problem. Different causes of facial asymmetry can be found such as craniosynostosis affecting skull base, craniofacial clefting, hemifacial microsomia or trauma to the mandibular condylar growth center. The UCH is a particular entity of facial asymmetry responsible for different specific clinical and radiological features.⁹ Vertical form, as the case presented above, is characterized by a unilateral increase in height of the affected side of the face. There is an asymmetry of the projection of the angle and the ramus. If the pathological condylar overgrowth is very rapid, an open bite on the affected side can appear. However, when the overgrowth is really progressive, the teeth on the affected side remain in occlusion at a lower level than the non-affected side, what is called tilted occlusion plane. In horizontal form, chin deviates on the non-affected side but angles remain at the same horizontal level. Occlusal analysis shows a displacement of the mandibular arch midline and a cross bite on the non-pathological side.² This phenomenon is particularly harmful to the temporomandibular joint.¹⁰

Diagnosis of condylar bone hypermetabolism is based on planar scintigraphy combined with SPECT scans. A difference in uptake values on the left and right condylar regions of 10% or more on scintigraphy is suggestive of active UCH. The SPECT scanning combined with planar scintigraphy identifies 19% more active UCH patients when compared with a standard planar bone scans.^{5,10} However, there is no uniform method for quantification of bone activity in planar or SPECT scans.^{5,10} As diagnosis of UCH is based on clinical and radiological features, SPECT imaging combined with planar scintigraphy only determines the bone metabolism activity in the overgrown condyle and is a guide for the therapeutic approach and its timing.⁸ Wolford goes further saying that bone SPECT scanning is unnecessary to show hyperactivity, that can be shown by lateral cephalograms and clinical diagnostic techniques with serial assessments (6- to 12-month intervals).⁴ We, therefore, think that SPECT scans combined with planar scintigraphy is interesting to highlight the intensity of the hypermetabolism in bone condyle and this to provide guidance on the timing of the surgical care. For some who still think that condylectomy is not legitimate, in non-active cases, or in cases when surgical treatment is delayed until the end of UCH growth,⁵ a more conventional orthognathic surgery is typically suggested to respect the TMJ. However, this approach can be problematic because the disorder has a dynamic component and the development of asymmetry is very unpredictable, sometimes very rapid and severe. Its repair may thus be managed more difficultly.¹¹ On the other hand, the case presented in this article, the first to our knowledge, may be compatible with a UCH reactivation after a long period of latency, leading to a relapse of occlusal disorder and facial asymmetry, more difficult to treat and to be accepted by the patient. Reactivation of UCH is an uncommon phenomenon but Wolford described a type of UCH caused by an accelerated growth of the normal growth mechanism, essentially with a vertical growth vector claimed to be present at any age and not to be self-limited.^{4,11} The authors agree to say that the case presented here lacks strong proof, especially concerning the past orthognathic treatment of the facial asymmetry, as these facts were reported by the patient herself and confirmed only by an orthopantomogram. But the patient's complaint of facial asymmetry relapse and dental prosthetic instability only since 2 years, associated with highly suggestive further examination and histological results provide sufficient arguments to raise the question of the possibility of reactivation of UCH after a long period of latency. Histologically, the hypertrophic layer of the condylar cartilage with an increased prechondroblastic cells count is clearly more compatible with the diagnosis of condylar hyperplasia than with condylar osteochondroma, which is the main differential diagnosis.¹² It remains difficult to answer the question of an acute reactivation after a period of total inactivity of the overgrowth process or a long active processing asymmetry with an exacerbation of the condylar hypermetabolism due to occlusal disorder and dental prosthetic instability. Both hypotheses are interesting to mention and suggest the necessity of very long term follow up of condylar hyperplasia untreated with first condylectomy but with orthognathic surgery.¹³

Finally, the choice between proportional or high condylectomy is controversial. For Wolford, high condylectomy with sole resection of the growth cartilage is a more functional surgery preserving the TMJ function.⁶ Proportional condylectomy as described by Delaire allows the adjustment of the posterior vertical excess at the same time, even if some remaining asymmetrical imperfections can be sometimes observed.^{7,14} We therefore think that high

condylectomy is legitimate in young patients, eventually associated with orthognathic surgery to correct later a possible remaining facial asymmetry. In our case, we chose to perform a proportional condylectomy because of the important vertical component asymmetry and also in order to avoid another corrective surgical procedure for asymmetry that the patient clearly did not want. In both cases, it is absolutely necessary to respect the articular disc, eventually to replace it if necessary and to enhance postoperative TMJ function by active rehabilitation.^{6,15}

CONCLUSION

The UCH that does not benefit from a condylectomy can be reactivated after a more or less lengthy period of latency. The UCH diagnosis is based on a body of clinical arguments in which SPECT-CT only provides information on the bone metabolism activity that guides the timing of the surgical management. Condylectomy should be performed as soon as UCH is diagnosed whatever the type of UCH is as the first-line treatment. If not, it appears essential to adopt very long term follow up of patients with past medical history of condylar hyperplasia not treated with condylectomy first but just by orthognathic surgery.

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The Risk Factors for Facial Numbness After Microvascular Decompression in Patients With Trigeminal Neuralgia

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Object: Microvascular decompression (MVD) is the most popular surgical procedure for treating Trigeminal neuralgia (TN). In this article, the authors conducted a large case series in which patients underwent MVD for TN, and focus on surgical outcomes, intraoperative findings, complications and risk factors.

Methods: From January 2017 to June 2017, a total of 84 patients with TN were treated with MVD in our department. The authors retrospectively analyzed the surgical outcomes and postoperative complications of these patients. Risk factors were analyzed by binary logistic regression analysis.

Results: Of the 84 patients, 69 had complete postoperative symptom relief (BNI I-II). A total of 28 patients developed postoperative facial numbness (BNI III-IV) and 1 patient died intraoperatively. With binary logistic regression analysis, significant risk factors for postoperative Facial numbness (FN) were longer operation time (odds ratio [OR] 1.153, $P < 0.05$) and longer hospital stay (OR 1.371, $P < 0.05$). The patients' age, the length of the disease, the gender, and the side of the disease did not affect the occurrence of postoperative FN.

Conclusions: The study found that patients with TN treated with MVD had a good response rate after surgery. The incidence of FN

after surgery is not low, and longer duration of surgery and longer hospital stay are risk factors for FN. In the case of ensuring the success rate of surgery, reducing unnecessary operations, reducing the operation time, will help to reduce the occurrence of FN.

Key Words: Duration of surgery, Facial numbness, hospital stay, microvascular decompression, trigeminal neuralgia

Trigeminal neuralgia (TN) is a paroxysmal, knifecut-like or lightning-like pain. Typical trigeminal pain lasts for a few seconds and often occurs when brushing, washing, chewing, or even blowing, and atypical patients may experience persistent pain.⁴ The traditional concept thinks that TN is caused by vascular compression of the trigeminal root entry zone (REZ). It is a part of the nerve that is excessively peripheral to the periphery and is very fragile and prone to demyelinating changes.^{9,13,16} The TN has a variety of treatments. Treatment usually begins with carbamazepine and it often provides relief. The relief provided by carbamazepine or other drugs may decrease over time, about half of the patients become unwell to medication or the drugs produce debilitating side effects, so they seek other treatments, including percutaneous procedures, Stereotactic radiosurgery, glycerol injection. However, these treatments have a low long-term remission rate and increase the risk of facial numbness (FN) after surgery.^{5,6} In contrast, Microvascular decompression (MVD) is a better option with higher postoperative remission rates and lower postoperative FN.^{17,21} According to reports, there are many poor prognostic factors of TN surgery, including gender is female, venous compression, and atypical pain,^{1,4,15} but there are few reports on the risk factors of postoperative FN, which may be related to intraoperative nerve combing, sacrificial veins or intra-neural vessels.^{10,12,14} The FN after MVD is not uncommon, in this article, we aimed to investigate the risk factors for FN after MVD-treated TN patients.

METHODS

From January 2017 to June 2017, 84 TN patients who received MVD were enrolled in the Department of Neurosurgery, Xinhua Hospital, Shanghai Jiaotong University School of Medicine. The study excluded patients with secondary TN including tumors and vascular malformations, patients who underwent a second or more MVD treatment were also excluded. Through the electronic case system, outpatient review follow-up or telephone follow-up, we detailed the patients' age, gender, onset time, side of the disease, postoperative remission, and FN. At the same time, we accurately counted the duration of operation of each patient through the surgical video system. All surgeries are performed by the same surgeon to avoid differences in surgical outcomes due to different surgical techniques. Timing starts with the loose of the arachnoid under the microscope and ends at full decompression. We assessed the degree of postoperative facial pain and postoperative numbness according to the Barrow Neurological Institute (BNI) grade and treated patients with postoperative pain BNI grade I and II as remission, and III-V as non-remission. BNI grade I is considered to be completely numb-free after surgery, and grade II-IV is considered facial numb (Supplemental Digital Content, Table 1, <http://links.lww.com/SCS/A582>).

Operative Procedure: The operation took a standard retrosigmoid craniotomy, after exposing the sigmoid sinus and transverse sinus, the dura mater was opened, and the cerebrospinal fluid was released. The good visual field exposure was to fully release the arachnoid membrane rather than excessively pulling

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