Costochondral Graft in Young Children With Hemifacial Microsomia

Bin-Zhang Wu, DDS, Lian Ma, MD, PhD, Yang Li, MD, Shuo Chen, MD, and Biao Yi, MD, PhD

Abstract: Patients with severely hypoplastic mandibles usually require condylar reconstruction. This study aimed to describe costochondral graft (CCG) for condylar reconstruction and report subsequent outcomes of these grafts in young children with Pruzansky/Kaban type IIB and type III mandibular hypoplasia. This study included 4 young children with type IIB and type III hemifacial microsomia treated with CCG to reconstruct the condyle at the Department of Oral and Maxillofacial Surgery in our hospital from March 2008 to March 2014. Radiographic measurements and clinical examinations were conducted. The mean age of patients at operation was 3.8 years, ranging from 2.8 to 5.3 years. The mean follow-up period was 43.5 months, ranging from 23 to 63 months. Functional improvement was observed in all patients. The ribs had grown in all patients to date. Three patients had clinically improved face appearance with no significant chin point deviation and canting of the occlusal plane. Although the other patient had partly improved face appearance compared with preoperative condition, he still showed clinically significant facial asymmetry and chin deviation. Our results showed that condylar reconstruction with CCG is a feasible method in the treatment of type IIB and type III hemifacial microsomia in young children. These results will provide early preliminary suggestions of growth and stability of CCG in patients <5 years.

Key Words: Condylar reconstruction, costochondral graft, hemifacial microsomia, young children

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Branchial arch disorders involve a variety of developmental anomalies. Hemifacial microsomia (HFM) is among the more common conditions that affect primary aural, oral, and mandibular development. A pronounced variant of HFM called Goldenhar syndrome is diagnosed when epibulbar dermoids and vertebral anomalies are observed in addition to HFM. The condition is generally acknowledged to be extremely complex and heterogeneous. Gorlin et al[1] used the term oculo–auriculo–vertebral spectrum. Mandible deformity is the most common manifestation in HFM. Clinical classification is based on the structure and function of the affected mandible and temporomandibular joint, which can be classified into 3 types, which determine the surgical plan. Type I: mild glenoid, ramus, and condylar hypoplasia. Type IIA: moderate glenoid, ramus, and condylar hypoplasia, and good function of temporomandibular joint exists. Type IIB: mild-to-moderate glenoid, ramus, and condylar hypoplasia, and markedly abnormal location, being medial and anterior. Type III: mandibular ramus, condyle, and temporomandibular joint are totally absent.[2]

The treatment of type IIB and type III HFM patients usually requires condylar reconstruction with the use of autogenous grafts, distraction osteogenesis, and prosthesis implantation.[3,4] However, young children with type IIB and type III deformities usually have insufficient bone for distraction osteogenesis, and thus prosthesis implantation is not appropriate for these growing patients for lack of growth potential. Autogenous grafts are more common in condylar reconstruction.[3,5] Various reconstruction methods have different indications, for example, costochondral graft (CCG) is considered the best choice for young children with HFM.[6] Condylar reconstruction with CCG in the early time not only improves the mandibular function and facial appearance, but also makes normal growth of the affected maxilla because CCG has growth potential.[7] However, only a few have used CCG for condylar reconstruction on young children. The present study aimed to describe CCG for condylar reconstruction and report subsequent outcomes of these grafts in young children with Pruzansky/Kaban type IIB and type III mandibular hypoplasia.

PATIENTS AND METHODS

Patients

This study included 4 young children with type IIB and type III HFM treated with a CCG to reconstruct the condyle at the Department of Oral and Maxillofacial Surgery in our hospital from March 2008 to March 2014. The mean age of the children during operation was 3.8 years, ranging from 2.8 to 5.3 years. The Institutional Review Board approved this retrospective study and informed consent was obtained from each patient. Documentation included photographs, dental models, lateral cephalograms, posteroanterior cephalograms, orthopantomography, and spiral computed tomographs.

Surgical Management

Preoperatively, the surgeon worked with an orthodontic technician to design the optimal mandibular position using plaster dental models and radiographic and clinical results. Through the model surgery with plaster dental models, the orthodontic technician constructed a surgical occlusal splint to level the mandibular cant and align the chin point with the midsagittal plane. We used three-dimensional computer-aided surgical technology in the treatment of a child (patient 4) with Pruzansky/Kaban type IIB mandibular hypoplasia to improve the traditional surgical planning method. As such, surgeons can perform virtual plan and make a

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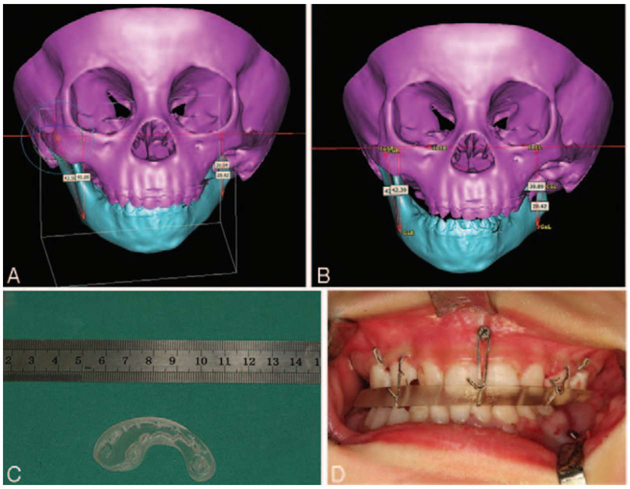

FIGURE 1. Three-dimensional computer-aided surgical technology was used in the treatment of patient 4 with Pruzansky/Kaban type IIB mandibular hypoplasia to improve the traditional surgical planning method. A, Precise digital dental models made by scanning the plaster dental models were incorporated into a three-dimensional computed tomographic scan of the patient's maxillofacial region. Then, the distance between the gonion and Frankfort plane was measured. B, The mandible was moved and rotated to the desired position to achieve symmetrical bilateral gonion. C, Surgical occlusal splint was prefabricated by computer-aided design/computer-aided manufacturing techniques. D, Accurate surgical occlusal splints enabled transfer of the surgical plan to the patient at surgery.

three-dimensional prediction of the patient's final mandibular position. First, we incorporated the precise digital dental models, which are made by scanning the plaster dental models into a three-dimensional computed tomographic scan of the patient's maxillofacial region. Second, we measured the distance between the gonion and Frankfort plane, and then moved and rotated the mandible to the desired position to achieve symmetrical bilateral gonion (Fig. 1A-B). Finally, the surgical occlusal splint was prefabricated by computer-aided design and manufacturing techniques, by which accurate surgical occlusal splints in the surgical plan could be transferred to the patient at surgery (Fig. 1C-D).[8] Intraoperatively, all patients underwent satisfactory nasotracheal anesthesia. A 4-cm submandibular incision was created on the surgical side approximately 1.5 cm below the inferior border of the mandible. Careful dissection was performed to expose the mandible; simultaneously, we preserved the marginal mandibular branch of the facial nerve. The periosteum was then incised along the inferior border of the mandible. A wide subperiosteal dissection was performed superiorly to expose the edge of the malformed mandible. The mandible was moved toward the unaffected side to align the maxillary and mandibular midlines with the midsagittal plane. The patient was placed under intermaxillary fixation using the surgical occlusal splint. A 4- to 5-cm-long rib was simultaneously harvested from the ipsilateral sixth rib along with a 1-cm cartilaginous cap (Fig. 2A). The cartilaginous head of the rib graft was shaped to resemble a condyle with approximately 0.5 cm in height. Then, the rib graft was placed in the suitable position as much as allowed by the contralateral temporomandibular joint and soft tissues. In 2 patients, the top structure of ramus on the affected side of the mandible was partially removed to place the rib graft in the optimal position. Pathologic examination of the specimens was also performed. The specimens were fixed in 10% neutral formalin and decalcified in 5% formic acid. Paraffin-embedded samples (6 mm) were cut and stained with hematoxylin and eosin. The rib graft was fixated using 4 microscrews with one 4-hole plate and 1 miniscrew (Fig. 2B). The donor and recipient areas were then closed. All patients remained in intermaxillary traction for 4 weeks and then functional traction for 8 weeks.

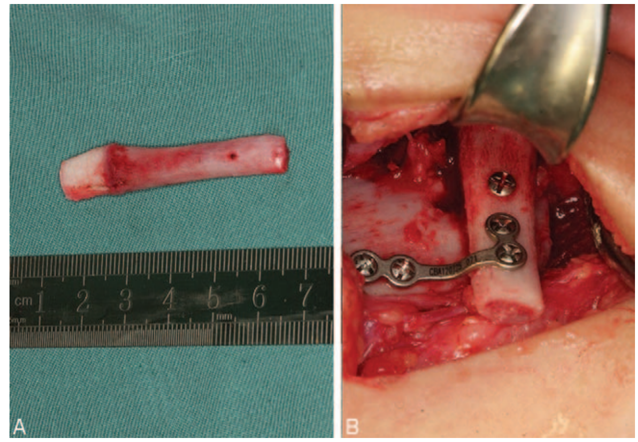

FIGURE 2. A, A 4- to 5-cm-long rib was simultaneously harvested from the ipsilateral sixth rib along with a 1-cm cartilaginous cap. The cartilaginous head of the rib graft was then shaped to resemble a condyle with approximately 0.5 cm in height. B, The rib graft was fixated in the suitable position using 4 microscrews with one 4-hole plate and 1 miniscrew.

Clinical Assessment Standard

Face appearance was classified as clinically improved, partly improved, and not improved. In the clinically improved classification, face appearance is rated very satisfactory by the children's parents and at least 2 professors, with no significant chin point deviation and canting of the occlusal plane. In the partly improved classification, face appearance is deemed satisfactory by the children's parents, but is deemed not satisfactory by at least 2 professors. A slight improvement is found compared with the preoperative condition, although facial asymmetry and chin deviation remain. In the not improved classification, face appearance is deemed not satisfactory by the children's parents and at least 2 professors; no improvement or a worsened condition is found compared with the preoperative condition, with significant facial asymmetry and chin deviation. Functional improvement was defined as improved chewing and better maximal mouth opening.

Radiographic Measurements

Lateral cephalograms were taken within 1 week preoperatively (T0), 1 week postoperatively (T1), and at the last follow-up (T2). Landmarks in the lateral cephalograms included Co (condylion at unoperated side, which is defined as the most posterior superior point of the condyle), Go (gonion at unoperated side, which is defined as the midpoint of the angle of the mandible found by bisecting the angle formed by the mandibular and ramus planes) (Fig. 3A); Co' (condylion at the operated side, defined as the most posterior superior point of the ramus/costochondral graft), and Go' (gonion at the operated side, defined as the midpoint of the angle of the mandible found by bisecting the angle formed by the mandibular and ramus/costochondral graft planes) (Fig. 3B).[9] Linear measurements in the lateral cephalograms were formed at Co–Go and Co'–Go'.

RESULTS

Clinical and Radiographic Analysis

We reported 4 children with HFM who received CCG for reconstruction of condyle; the classifications of mandible were type III and type IIB. The mean age at operation was 3.8 years,

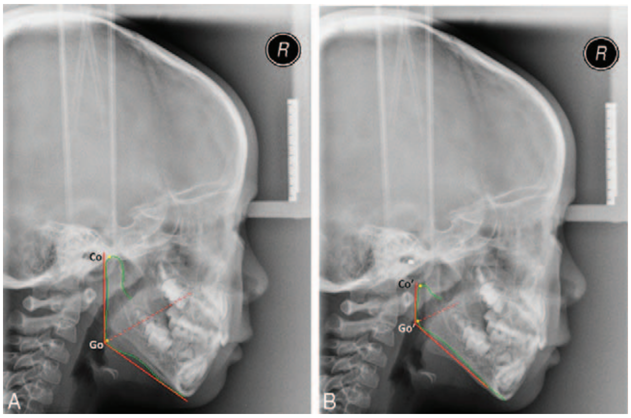

FIGURE 3. Landmarks in the lateral cephalograms: (A) Co and Go; (B) Co′ and Go′.

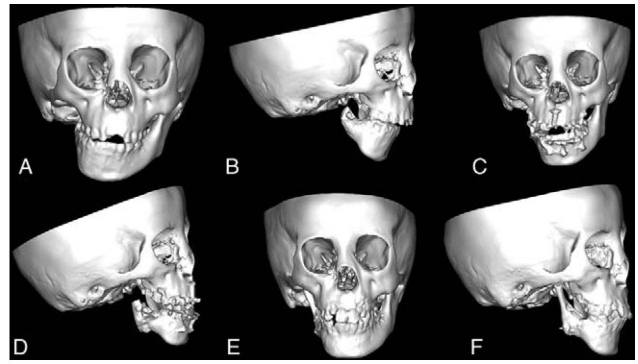

FIGURE 4. Preoperative three-dimensional computed tomographic scan at age of 3.8 years: (A) anteroposterior view and (B) right lateral view. Postoperative (1 wk) three-dimensional computed tomographic scan: (C) anteroposterior view and (D) right lateral view. Postoperative three-dimensional computed tomographic scan at age of 9 years: (E) anteroposterior view and (F) right lateral view.

ranging from 2.8 to 5.3 years. The mean follow-up period was 43.5 months, ranging from 23 to 63 months (Table 1). Functional improvement was found in all patients. The ribs had grown in all patients to date. Three patients showed clinically improved face appearance with no significant chin point deviation and canting of the occlusal plane, and the other patient (patient 2) had partly improved face appearance compared with preoperative condition, although he still showed clinically significant facial asymmetry and chin deviation.

The growth pattern of patient 1 was optimal type. The ramus length (Co–Go) discrepancy between the affected and unaffected sides decreased from 14.8 mm immediately after operation to 5.8 mm at 5 years after surgery; the ratio of unoperated-to-operated sides also decreased from 1.56 to 1.14. The face was symmetrical, the maximal mouth opening was 30 mm at 5 years after surgery, and the mandible slightly deviated to the right side during mouth opening. The occlusion leveled well with the occlusal cant. Good result was achieved after the surgery (Fig. 4A-F).

The growth pattern of patient 2 was suboptimal type. The ramus length (Co–Go) discrepancy between the affected and unaffected sides had not changed from immediately after operation to 5 years after surgery. The face was asymmetric, the chin was obviously deviated to the left side, the left angulus oris was high, the maximal mouth opening was 25 mm at 5 years after surgery, and the mandible obviously deviated to the left side during mouth opening. Moreover, the occlusion was bad, and the occlusal cant was high in the left with overbite and overjet on the left.

The growth pattern of patient 3 was optimal type. The ramus length (Co–Go) discrepancy between the affected and unaffected sides decreased from preoperative 14.8 to 4.5 mm at 2 years after surgery. The ratio of unoperated-to-operated sides was maintained up to 2 years after surgery. The face was symmetric, the maximal mouth opening was 31 mm at 2 years after surgery, and the mandible slightly deviated to the left side during mouth opening.

The growth pattern of patient 4 was moderately suboptimal type. The ratio of unoperated-to-operated sides decreased from 1.71 immediately after operation to 1.56 at 23 months after surgery. The face was slightly asymmetric, the chin was slightly deviated to the left side, and the maximal mouth opening was 35 mm at 23 months after the surgery.

Histopathology

The surface of pseudocondyle in patient 2 showed irregular cellular rich zone and a thin calcified cartilage zone, and a small split could be seen at the surface (Fig. 5A). Superfacial bone showed irregular resorption and small foci of necrosis (Fig. 5B-C).

TABLE 1. Patient Demographics and Changes in Mandibular Length

					Radiographic Measurements, mm						
					T0		T1		T2		
Patient	Sex	Age at Operation, y	Follow-Up, mo	Condyle/Type	Length, mm	Normal Abn**	Length, mm	Normal Abn**	Length, mm	Normal Abn**	Change (T1–T2), mm
1	M	3.8	63	R*/III	19.3	2.11	26.5	1.56	41.9	1.14	+15.4
				L	40.8		41.3		47.7		+6.4
2	M	3.4	64	R	43.6	2.17	43.2	2.07	49.6	1.81	+6.4
				L*/IIB	20.1		20.9		27.4		+6.5
3	M	5.3	24	R	38.4	1.63	38.1	1.04	43.3	1.12	+5.2
				L*/IIB	23.6		36.7		38.8		+2.1
4	F	2.8	23	R	48.6	1.77	47.8	1.71	54.2	1.56	+6.4
				L*/IIB	27.5		27.9		34.7		+6.8

*Operated side.
**Ratio of unoperated-to-operated side. Abn, abnormal.

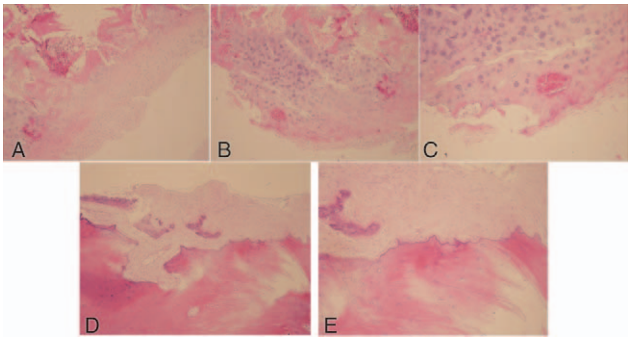

FIGURE 5. A, Surface of pseudocondyle in patient 2 showed irregular cellular rich zone and a thin calcified cartilage zone, and a small split could be seen at the surface (hematoxylin and eosin stain, ×10). B–C, Superfacial bone showed irregular resorption and small foci of necrosis (hematoxylin and eosin stain, ×20 and ×40). D–E, Histologic examination of patient 4 showed fibrous connective tissues lining the surface of the pseudocondyle and several calcified foci; no joint cartilage was found. Irregular bone resorption was observed (hematoxylin and eosin stain, ×10 and ×20).

Histologic examination of patient 4 showed fibrous connective tissues lining the surface of the pseudocondyle and some calcified foci. No joint cartilage was found, and irregular bone resorption was observed (Fig. 5D-E).

DISCUSSION

The first CCG for condylar reconstruction was reported by Gillies in 1920. Since then, a wide variety of autogenous grafts, such as clavicle and sternoclavicular joint,[10,11] fibula,[12] iliac bone,[13] and metatarsal bone,[14] have been reported for condylar reconstruction. However, autogenous CCG is considered the most common choice for condylar reconstruction in growing children with Pruzansky/Kaban type IIB and type III mandibular hypoplasia.[6] The cartilage/bone junction is considered a growth center in CCG with considerable growth potential. In 2016, certain authors have advocated nonvascularized (eg, costochondral) bone graft for severe deformity with adequate mandibular body and vascularized (eg, free fibular) bone flap for severe deformity with inadequate mandibular body bone stock.[15]

Costochondral graft in growing children is likely to be one of the reasons for positive outcome. Ross[6] reported a success rate of 80% when CCG was performed in children aged 3 to 9 years. The success rate decreases to 50% for patients aged $\geq$14 years. Goerke et al[16] also reported the functional improvement in 8 of 10 patients aged 3 to 11 years. Tahiri et al[17] recommended that the surgery should be performed in patients >5 years, and they indicated that the quality of the rib graft was suboptimal in patients <5 years. Kaban et al[2] recommended that the graft should be performed when the Pruzansky/Kaban type IIB and type III patients manifest shortening of the midface and tilting of the occlusal plane, which usually occurs at ages 2 to 5 years. Munro et al[18] reported 22 children aged <14 years who underwent temporomandibular joint construction using CCG; they recommended early surgery in the patients with HFM given that all the grafts had grown. The hypoplastic mandible influences normal maxillary growth, and serious mandibular deformity leads to secondary deformation of the midface owing to the asymmetric skeletal growth.[19] Early surgery in children with Pruzansky/Kaban type IIB and type III mandibular hypoplasia offers several advantages, including relatively easy for surgeon and patient, as well as psychologic benefits, which are very important to patients and their families.[2,6] In our study, we also expected the optimal rib growth to correct facial asymmetry and improve functional problems, such as malocclusion and occlusal cant before primary school; therefore, the selected children with mean age of 3.8 years, ranging from 2.8 to 5.3 years, at surgical correction.

The growth pattern of the CCG was classified as overgrowth, optimal, suboptimal, no growth, or resorption by serial radiologic measurements and clinical appearance. Guyuron and Lasa[20] concluded that the growth pattern is difficult to predict. Overgrowth pattern can be classified into linear and lateral contour overgrowth of the articulating surfaces, which sometimes occur simultaneously.[21,22] Linear overgrowth is characterized by asymmetric or symmetrical prognathism without hypomobility. Patients with lateral contour overgrowth of the articulating surface might develop contour abnormality with mandibular hypomobility or ankylosis.[20] The optimal pattern is proportional growth of the costochondral rib graft relative to the face. The suboptimal pattern is facial asymmetry and tilting occlusal plane after CCG; hence, a secondary distraction should be performed. Wan et al[23] documented 27 patients with CCG (30 rib grafts) and found undergrowth in 17 rib grafts (57%), which required subsequent distraction, and overgrowth in 3 patients, which needed further orthognathic correction. No growth and resorption of the CCG were detected by radiological comparison. Goerke et al[16] reported 10 patients aged 3 to 11 years treated with CCG, among which 5 patients did not need further surgeries to last follow-up, severe overgrowth was found in 1 patient, and partial or complete resorption was noted in 3 patients. In the present 4 patients, patient 1 and patient 3 were classified as optimal pattern, patient 2 was suboptimal pattern, and patient 4 was moderately suboptimal type. We would continue observing the 2 suboptimal cases until middle-to-late adolescence to determine whether the costochondral rib graft would not grow proportionally to the face at that time, in which a rib distraction is required. Simultaneously, we would still observe the evolution of grafts in patients 1 and 3.

Moss and Rankow[24] proposed functional matrix theory, in which the mandibular growth process is a secondary response to attached organs, tissues, and functioning spaces. Downward and forward mandibular growths affect secondary growth of the condylar cartilage. Peltomäki et al[22] histologically examined 7 removed overgrown CCGs from 6 patients, and found no particular histological finding regarding clinical linear or exuberant overgrowth. Furthermore, they evaluated that local factors, including mandibular movements and loading of the reconstructed temporomandibular joint, might influence the growth of the CCG. Ellis and Carlson[25] proposed that the cartilage of the costochondral junction is a growth center controlled by intrinsic factors. Both the intrinsic and extrinsic factors might affect growth of CCG.

Experimental[26–28] and clinical findings[21] proved that the size of cartilage at the costochondral junction likely influence the growth of CCG. Most articles on patients with CCG reported a cartilage cap of 0.5 to 2.5 cm.[20,29] Tideman and Doddridge[30] suggested preservation of a smaller cartilage cap to prevent fracture at the costochondral junction and decrease risk of overgrowth. Perrott et al[21] reported 26 patients with CCG in which only 2 to 4 mm of cartilage was used, and no patients showed clinically significant linear overgrowth. Goerke et al[16] described 10 children in which 2 mm of costochondral cartilage was used, and only 1 patient experienced linear overgrowth at 5.7 years after surgery when the patient was 17 years. They explained that hormonal changes at puberty led to overgrowth of graft. Tahiri et al[17] reported 22 patients in which 81.8% of the CCGs showed clinically adequate growth. They owed good outcomes to adequate cartilage size (approximately 7 mm), preservation of periosteal and perichondrial sleeve, blunt dissection and technique, brief immobilization, and early joint loading. We used approximately 5 mm of costochondral cartilage in the present study. The ribs had grown in all patients to date. Good face appearance with no significant chin point deviation and canting of the occlusal plane was observed in 3 children. The other child had partly improved function and face appearance compared with preoperative condition, although undergrowth was found.

Several articles have reported the use of CCG in patients to reconstruct condyle with a preauricular incision combined with a small submandibular incision.[16,19,21] A preauricular incision combined with intraoral incision was also documented,[31] which had no possibility of damaging the marginal mandibular branch of the facial nerve. Tahiri et al[17] used a single submandibular incision. In our cases, a single submandibular incision was enough for operation given that the patients were young children. No preauricular scar was found and relatively shorter operating time was achieved.

Importantly, we consider that the position and direction of costochondral rib graft might be factors that influence the growth of CCG. Glenoid fossa is difficult to identify in patients with Pruzansky/Kaban type IIB and type III mandibular hypoplasia, given that absent situation has been seen in several cases. In our experience, the rib graft was placed in a suitable position as much as allowed by the contralateral temporomandibular joint and soft tissues. Partial removal of the top structure of ramus on the affected side of the mandible was performed in 2 patients to place the rib graft in the optimal position. Long-term follow-up is warranted. Histopathologic examination revealed irregular cellular rich zone and a thin calcified cartilage zone in patient 2, and superfacial bone showed irregular resorption and small foci of necrosis. Histologic examination of patient 4 showed several calcified foci and no joint cartilage; however, irregular bone resorption could be seen. The histological appearance of pseudocondyle added new information to clinical type IIB HFM. In addition, histopathologic findings suggested that there were no true capsular elements in the pseudocondyle biopsied, thus this might offer further evidence to support joint reconstruction de novo with a CCG as opposed to joint (or pseudojoint) preservation by performing distraction.

Finally, we emphasize that further changes may occur upon growth and development of the children given that the growth of CCG is unpredictable. We will use combined orthodontic and surgical treatments according to the situation of the patients until completion of growth or when necessary to attain long-term effect. In this study, we only used CCG to reconstruct condyle; future investigation will focus on deformities of maxilla and soft tissues.

CONCLUSIONS

We presented 4 cases of CCG in patients aged 2 to 5 years with HFM, with 2 to 5 years of follow-up. We used lateral cephalograms to measure subsequent growth. We were able to observe moderate-to-adequate postoperative growth with face appearance and functional improvement in each patient. These results provide early preliminary suggestion of growth and stability of CCG in patients $<$5 years. Further studies in this age group with a larger sample and a longer follow-up period are warranted.

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