Course of the mandibular canal in hemifacial microsomia: a retrospective computed tomography study

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Objective. The aim of this study was to evaluate the anatomic course of the mandibular canal in patients with hemifacial microsomia (HM).

Study Design. In this retrospective study, 77 patients were included and stratified according to the Pruzansky-Kaban classification. The mandibular canal and the mandible were reconstructed on the basis of computed tomography data. The entrance, route, and exit of the mandibular canal (representing the entrance, route, and exit of the inferior alveolar nerve [IAN], respectively), and the antilingula were analyzed in different types of mandibular deformities in patients with HM.

Results. No significant difference in the course of the mandibular canal was detected between the affected and unaffected sides in patients with type I and type IIa HM. Abnormalities were observed in some patients with type IIb and type III HM. Significant differences were found between patients with type IIb and type III HM in the entrance ($P = .015$) and route ($P = .001$) of the canals. The antilingula was identified only in patients with type IIb and type III HM and was more common in patients with type III HM than in those with type IIb HM.

Conclusions. Variation of the anatomic course of mandibular canal exists in patients with Pruzansky-Kaban type IIb and type III HM. Evaluation of the course of the canal in patients with HM is recommended before surgical intervention to avoid IAN damage.
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Hemifacial microsomia (HM) is one of the most common congenital deformities in the craniofacial region, second only to cleft lip and palate, with a prevalence of approximately 1 in 3500 to 1 in 5600 live births.[1−3] The structures derived from the first and second branchial arches can be different in patients with HM compared with the normal population.[1,4] The OMENS (orbital asymmetry, mandibular hypoplasia, ear deformity, nerve development, and soft-tissue deficiency) and Pruzansky classifications are commonly used to describe the different phenotypes associated with HM.[4] Dysplasia of the mandible is one of the most common manifestations, which can lead to severe facial asymmetry, malocclusion, and temporomandibular joint dysfunction. The original classification of HM was proposed by Pruzansky and focused on the size and shape of the mandible and the glenoid fossa (Table I).[1,5] To better describe mandibular deficiencies, the Kaban modification of the Pruzansky classification (Pruzansky-Kaban classification) was introduced.[6] The Pruzansky-Kaban classification proposed further stratification of patients with type II HM on the basis of the relationship between the mandibular condyle and the glenoid fossa (see Table I). Patients with type I and type IIa HM exhibit mild hypoplasia of the ramus.[1,5,6] Type IIb (severely hypoplastic condyle) and type III (absence of ramus and glenoid fossa or no temporomandibular joint) are characterized by severe ramus and glenoid fossa deformities.[5,6]

Distraction osteogenesis (DO) and costochondral grafts have been widely used for the reconstruction of the mandibular ramus in children with type IIb and type III HM.[3,7] However, DO and costochondral grafts can be associated with a risk of inferior alveolar nerve (IAN) damage during the process of splitting of the ramus and fixation of the titanium plates. The abnormal course of the IAN in patients with HM has been reported to be associated with failure of the IAN block in previous studies.[8,9]

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Statement of Clinical Relevance

Hemifacial microsomia is the second most common craniofacial congenital deformities. We correlated the anatomic description of the course of the inferior alveolar nerve (IAN) with the severity of mandibular malformation. Better understanding of the anatomic course of the IAN in patients with hemifacial microsomia can help surgeons avoid acute IAN damage during surgery.

Table I. Pruzansky and Pruzansky-Kaban classifications

	Pruzansky classification	*Pruzansky-Kaban classification*
Type I	Small mandible and glenoid fossa with short ramus	
Type II (In Pruzansky-Kaban classification, type II is subclassified as type IIa and type IIb)	Abnormally shaped and short ramus	Type IIa: Glenoid fossa in acceptable position with reference to contralateral TMJ Type IIb: Glenoid fossa is inferiorly, medially, and anteriorly displaced with a severely hypoplastic condyle
Type III	Absence of ramus and glenoid fossa (TMJ)	

TMJ, temporomandibular joint.

The IAN is a branch of the mandibular nerve and is encased within the mandibular canal. The mandibular canal is surrounded by bony tissue, forming a forward, concave curve in the lingual half of the mandible. The IAN normally enters the mandible at the mandibular foramen on the lingual surface of the ramus. Branches of the IAN exit at the mental foramen at the buccal surface of the mandibular body. On radiographic images, the course of the mandibular canal is evaluated to trace the route of the IAN.

Preoperative evaluation of the course of the mandibular canal by using computed tomography (CT) could improve the safety of the surgical procedures.[10] The antilingula is the reference mark on the buccal surface of the ramus used by surgeons to identify the position of the mandibular foramen on the lingual surface.[11] Therefore, the purpose of this study was to qualitatively describe the entrance, route, and exit of the mandibular canal in patients with HM by using 3-dimensional (3-D) CT images. The prevalence and functionality of the antilingula were also evaluated.

MATERIALS AND METHODS

This retrospective study was conducted at the Shanghai Ninth People's Hospital during January 2014 to June 2018. All patients with HM who underwent CT for treatment purposes were included. Patients with bilateral HM were excluded from this study.

Image acquisition and processing

Each CT scan was acquired by using a spiral CT scanner (GE Healthcare, Fairfield, CT). The scan parameters were 120 kV and 178 mAs. The slice thickness of the CT image was 1.25 mm. The CT images were exported in Digital Imaging and Communications in Medicine format and imported to ProPlan software (Materialise, Leuven, Belgium) to reconstruct the 3-D model of the skull and the mandibular canal.

Image analysis and data collection

Two observers independently scored each patient on the basis of the Pruzansky-Kaban classification (type I, type IIa, type IIb, type III). Each observer independently evaluated the mandibular canal on axial, sagittal, and coronal sections and on 3-D reconstructed models. The entrance, route, and exit of the mandibular canals of each patient were recorded. The entrance was classified as "regular entry" (on the lingual side of the mandible) and "abnormal entry" with 2 subclassifications (entry from the top of the ramus or entry on the buccal surface) (Figure 1). The route was classified as "regular route" (the mandibular canal is in the cancellous bone of the lingual half) and "abnormal route," with 3 subclassifications (the mandibular canal remains on the buccal side of the mandible, bifid mandibular canal, or the canal descending from the anterior aspect of the ramus) (Figure 2). The exit point was classified as "regular exit" (on the buccal aspect of the mandibular corpus) or "abnormal exit" with 3 subclassifications (exit on the lingual surface, exit from the anterior aspect of the ramus, or exit point absent) (Figure 3). The antilingula was marked as present or absent

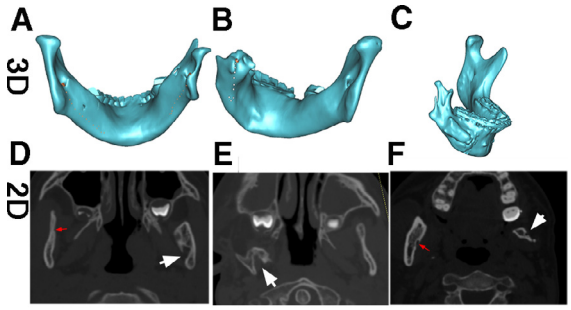

Fig. 1. Entrance of the mandibular canal as depicted in 3-dimensional (3-D) reconstructed models **(A−C)** and CT reconstructed images **(D−F)**. **A, D,** Regular (normal) entrance of the inferior alveolar canal (IAC) is on the medial side (lingual side). **B, E,** Entrance of the IAC is shifted to the top of the ramus. **C, F,** IAC entrance is located on the buccal side of the ramus. The white arrow indicates the entrance of the IAC on the affected side in axial section. The red arrow indicates the entrance of IAC on the unaffected side, which is normal.

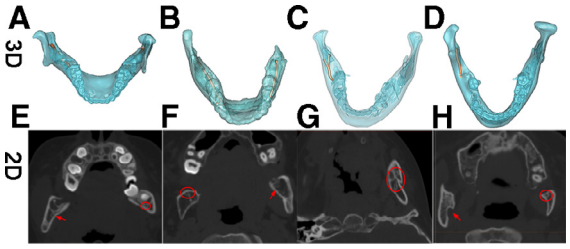

Fig. 2. Route of the mandibular canal as depicted in 3-dimensional (3-D) reconstructed models **(A−D)** and CT reconstructed images **(E−H)**. **A, E,** Regular (normal) route of the inferior alveolar canal (IAC) on a 3-D reconstruction model and axial image. **B, F,** The IAC runs along the buccal aspect of the mandible on a 3-D reconstruction model and axial image. **C, G,** Bifid mandibular canal on a 3-D reconstruction model and image. **D, H,** The IAC descending adjacent to the anterior boundary of the ramus. The red circle highlights the mandibular canal on the affected side. The red arrow indicates the normal mandibular canal on the unaffected side.

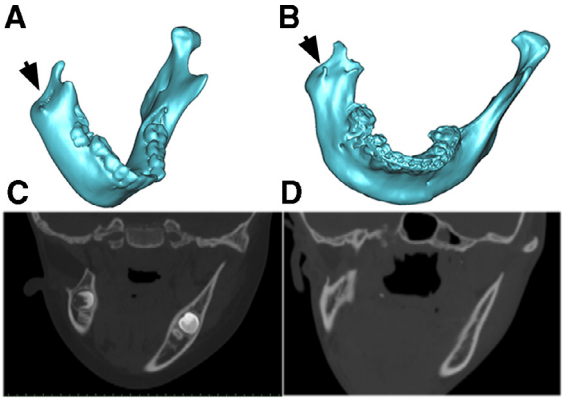

Fig. 4. The antilingula, shown in 3-dimensional (3-D) reconstruction models **(A, B)** and reformatted computed tomography (CT) images **(C, D)**. The antilingula has no direct connection with the inferior alveolar canal (IAC). The black arrowheads indicate the antilingula in 3-D reconstruction models.

(Figure 4). The evaluations completed by the 2 observers were compared, and all disagreements were resolved through discussion. If mutual agreement between the 2 observers could not be accomplished by means of discussion, a third observer, who was a senior staff member, took part in the discussion and made the final decision. All observers were oral and maxillofacial surgeons with a minimum 3 years of surgical experience. Observers 1 and 2 were trained 3 years and 6 years, respectively, in CT image interpretation.

Statistical analysis

Comparisons of the courses of the mandibular canals (normal and affected side) were performed by using the 2-sided Fisher's exact test at the 5% significance level. The association between the mandibular canal abnormality and the mandibular deformity was calculated with the Cochran Armitage trend test. Statistical analysis was done with SAS 9.4 (SAS Institute, Cary NC) and performed by a statistician.

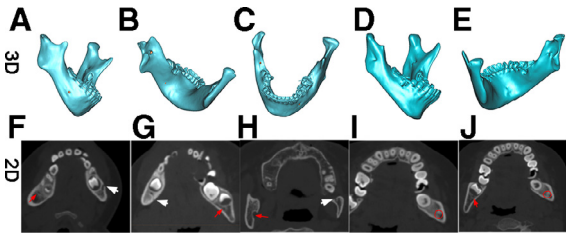

Fig. 3. Exit of the mandibular canal or inferior alveolar canal (IAC) as depicted in 3-dimensional (3-D) reconstructed models **(A−E)** and computed tomography (CT) reconstructed images **(F−J)**. **A, F,** Regular (normal) exit of the IAC on the buccal side of the mandible. **B, G,** The IAC leaving the mandible from the lingual side (medial side) of the ramus. **C, H,** The IAC leaving the mandible from the anterior aspect of the ramus. **D, E, I, J,** The IAC vanishes when it descends into the ramus. The white arrowhead indicates the exit of the IAC on the axial section. The red circle highlights the mandibular canal on the affected side. The red arrow indicates the normal mandibular canal on the unaffected side.

RESULTS

In total, 77 patients were included in this study (48 males and 29 females). Six patients were classified as having HM of type I (2 males and 4 females); 24 patients were classified as type IIa (13 males, 11 females); 32 patients were classified as type IIb (21 males, 11 females); and 15 patients were classified as type III (12 males, and 3 females). In the case of 7 patients who were originally diagnosed with type IIa or type IIb HM, further discussions were conducted with the third observer before the final classification was made.

The entrance, route, and exit of the mandibular canal in all patients with type I and type IIa HM were normal. Some patients classified as having type IIb and type III HM exhibited abnormal findings (Tables II, III, and IV). It was not possible to trace the mandibular canal for 1 patient with type III HM, and this patient's data were not included among those of the 77 patients.

Fisher's exact test showed statistically significant differences in the entrance ($P = .015$) and the route ($P = .001$) of the mandibular canal between patients with type IIb and type III HM (see Tables II and III). The variation of the exit of the mandibular canal on the affected side showed no significant difference between type IIb and type III ($P = .091$), as shown in Table IV.

Table II. The entrance of the IAC in HM patients

Type	*Number of patients*	*Regular entrance*	*Abnormal entrances*		*Percentage of abnormal entrances (%)*	*P value*
			Entry from the top of the ramus	Buccal entry		
Type I	6	6	0	0	0	$P = .015$ between patients with type IIb and type III HM
Type IIa	24	24	0	0	0	
Type IIb	32	28	2	2	12.5	
Type III	15	10	3	2	33.3	

IAC, inferior alveolar canal; *HM,* hemifacial microsomia.

Table III. The route of the IAC in patients with HM

Type	*Number of patients*	*Regular routes*	*Abnormal routes*			*Percentage of abnormal routes (%)*	*P value*
			On the buccal side	*Bifid mandibular canal*	*Descending from the anterior aspect of the ramus*		
Type I	6	6	0	0	0	0	$P = .001$ between patients with type IIb and type III HM
Type IIa	24	24	0	0	0	0	
Type IIb	32	24	4	2	2	25	
Type III	15	6	5	1	3	60	

IAC, inferior alveolar canal; *HM,* hemifacial microsomia.

Table IV. The exit of the IAC in patients with HM

Type	*Number of patients*	Regular exit	*Abnormal exits*			*Percentage of abnormal exits (%)*	*P value*
			Lingual exit	*Exit from anterior aspect of the ramus*	*Absent*		
Type I	6	6	0	0	0	0	$P = .091$ between patients with type IIb and type III HM
Type IIa	24	24	0	0	0	0	
Type IIb	32	27	2	2	1	15.6	
Type III	15	12	1	1	1	20.0	

IAC, inferior alveolar canal; *HM,* hemifacial microsomia.

The Cochran Armitage trend test showed that type III HM may be associated with a higher prevalence of variations of mandibular canal ($\chi^2 = -2.244$; $P < .05$).

The antilingula was observed only in patients with type IIb and type III HM (Table V). Examination of the axial and coronal sections revealed that the antilingula was superior to the lingula in 11 cases, level with the lingula in 2 cases, and inferior to the lingula in 7 cases. There was no evidence that the lingula had a direct connection to the antilingula. This proved that the antilingula could not be marked as the entrance point of the mandibular canal (see Figure 4). The antilingula had a higher prevalence in the more severe mandibular deformities (type IIb and type III) compared with the less severely involved mandibles (type I and type IIa) ($P < .001$), but there was no significant difference in the frequency of antilingula between type IIb and type III.

DISCUSSION

This study evaluated the relationships between the mandibular canal and the Pruzansky-Kaban types of mandibular deformities in patients with HM. HM is a common craniofacial deformity, which manifests mainly as mandibular deformity that is associated with mandibular canal deviation. Pruzansky-Kaban is the most widely used system for the evaluation of the severity of mandibular deformity in patients with HM. Better understanding of the course of the mandibular canal can help avoid damaging the IAN during surgery.

On the basis of 3-D CT data and morphologic reconstruction, we were able to correlate the Pruzansky-Kaban classification with the course of the mandibular canal. To avoid nerve injury when performing the bone-splitting and fixation procedures, researchers have used fixed anatomic landmarks to quantitatively compare the affected

Table V. Antilingula in patients with HM

Type	*Number of patients*	*Antilingula*	*Percentage of Antilingula (%)*	*P value*
Type I	6	0	0	$P < .001$ between mild deformities (type I and type IIa HM) and severe deformities (type IIb and type III HM)
Type IIa	24	0	0	
Type IIb	32	13	40.6	
Type III	15	7	46.7	

HM, hemifacial microsomia.

side of the mandible in patients with HM.[12,13] Their results indicated that fixed anatomic landmarks appear to be in the same positions on both sides in patients with Pruzansky type I HM. In patients with type II deformity, there was a significant difference in the height of the ramus and the length of the mandibular canal between the 2 sides of the mandible.[12,13] However, those studies either used small sample sizes or focused only on low-grade mandibular deformation. Additionally, previous studies only used the Pruzansky classification.[12,13] The Kaban modification of type II can more precisely classify patients in terms of the morphology of the ramus and the condyle. This modification can influence the clinical decision.

The findings of the present study are consistent with those of a previous investigation because no obvious abnormalities of the mandibular canal were observed for patients with type I or type IIa HM in the Pruzansky-Kaban classification.[12] In patients with type IIb HM, however, the entry of the mandibular canal was detected at the superior aspect of the ramus (the sigmoid notch) or the buccal side of the ramus in some cases. In patients with type III HM, there was a higher frequency of entry deviation compared with those with type IIb HM. These findings can be important for mandibular molar removal, endodontic treatment, and the classic IAN block for pain control.[14]

In DO or sagittal ramus osteotomy, surgeons cut the cortex and penetrate the medullary bone on the buccal side of the ramus. As the route of the mandibular canal could shift to the buccal side or descend along the anterior aspect of the ramus close to the cortex in patients with types IIb and III HM, the IAN could be easily injured. For those patients, it is necessary to mark the mandibular canal with a surgical guide or at least take care during the surgical procedure to avoid IAN damage. However, the mandibular canal could not be traced in one of the patients with Pruzansky-Kaban type III HM; this problem has been reported in other studies as well.[13,14] We propose that the IAN may run within the muscles or connective tissues, rather than remain enclosed in the bony canal.[14]

In our study, bifurcation of the mandibular canal appeared only on the affected side in patients with type IIb or type III mandibular malformation. We propose that the bifid mandibular canal is caused by the mandibular deformation. From the embryologic standpoint, the IAN is closely related to mandibular osteogenesis.[15,16] Any disruption during mandibular development may also lead to IAN variation. This is supported by our finding that variations in the route of the mandibular canal are significantly more frequent in patients with severe mandibular deformity.

Deviation of the exit of the IAN in HM is seldom reported. In previous studies, the distances from the lingula to the mental foramen (exit point of the IAN) was found to be significantly shorter on the affected side in patients with type II HM.[13] The exit point variation may result in damage to the mental nerve (terminal branch of IAN) during mandibular flap surgery. It is necessary to trace the exit of IAN before flap surgeries in patients with HM to avoid this problem.

The antilingula was a common phenomenon in patients with type IIb and type III HM in our investigation. Some researchers propose that the antilingula can be used as a landmark to locate the mandibular foramen, whereas others suggest that the antilingula is a bony response to the muscles and tendons that attach to that area, rather than a structure with a relationship with the inferior alveolar canal.[11,17–19] In our study, we found that the antilingula had no connection with the entry of the mandibular canal. The antilingula may be the result of muscle or tendon insertion. When the condyle and ramus are severely damaged, the regular attachment of the soft tissues is disrupted, and new attachments of the muscles and tendons may lead to the generation of an antilingula.

CONCLUSIONS

This study focused primarily on the variations in the mandibular canal in patients with HM. Our findings showed the significant variation in the anatomic course of mandibular canals in patients with type IIb and type III HM. In patients with both type IIb and type III HM, the anatomic course of the mandibular canal was unpredictable. On the basis of our findings, we suggest that the mandibular canal be assessed with 3-D imaging before surgical intervention in patients with HM. Further study to analyze the relationship between the mandibular canal and the mandible should be conducted with larger patient samples.

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