OPEN

ORIGINAL ARTICLE
Pediatric/Craniofacial

Surgical Classification of the Mandibular Deformity in Craniofacial Microsomia Using 3-Dimensional Computed Tomography

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Background: Grading systems of the mandibular deformity in craniofacial microsomia (CFM) based on conventional radiographs have shown low interrater reproducibility among craniofacial surgeons. We sought to design and validate a classification based on 3-dimensional CT (3dCT) that correlates features of the deformity with surgical treatment.

Methods: CFM mandibular deformities were classified as normal (T0), mild (hypoplastic, likely treated with orthodontics or orthognathic surgery; T1), moderate (vertically deficient ramus, likely treated with distraction osteogenesis; T2), or severe (ramus rudimentary or absent, with either adequate or inadequate mandibular body bone stock; T3 and T4, likely treated with costochondral graft or free fibular flap, respectively). The 3dCT face scans of CFM patients were randomized and then classified by craniofacial surgeons. Pairwise agreement and Fleiss' κ were used to assess interrater reliability.

Results: The 3dCT images of 43 patients with CFM (aged 0.1–15.8 years) were reviewed by 15 craniofacial surgeons, representing an average 15.2 years of experience. Reviewers demonstrated fair interrater reliability with average pairwise agreement of 50.4±9.9% (Fleiss' κ = 0.34). This represents significant improvement over the Pruzansky–Kaban classification (pairwise agreement, 39.2%; $P = 0.0033$.) Reviewers demonstrated substantial interrater reliability with average pairwise agreement of 83.0±7.6% (κ = 0.64) distinguishing deformities requiring graft or flap reconstruction (T3 and T4) from others.

Conclusion: The proposed classification, designed for the era of 3dCT, shows improved consensus with respect to stratifying the severity of mandibular deformity and type of operative management. *(Plast Reconstr Surg Glob Open 2016;4:e598; doi: 10.1097/GOX.0000000000000582; Published online 27 January 2016.)*

Craniofacial microsomia (CFM) is the second most common congenital facial anomaly after cleft lip and palate and arises from malformation of the first and second branchial arches.[1–4] It is clinically manifest by variable hypoplasia of the *o*rbits, *m*andible, *e*ar, facial *n*erve, and adjacent *s*oft tissue (collectively known by the acronym "OMENS") and ranges in severity.[5,6] Ninety

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Received for publication June 18, 2015; accepted December 2, 2015.
Funding to support this investigation was received by the Center for Human Appearance at the University of Pennsylvania.
Presented at the Sixteenth Biannual Meeting of the International Society of Craniofacial Surgeons, September 14–18, 2015, Tokyo, Japan.

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DOI: 10.1097/GOX.0000000000000582

percent of patients have unilateral or hemifacial microsomia, and 10% present bilaterally.[2] Mandibular hypoplasia is usually the earliest and most obvious sign of CFM.[7]

Multiple classifications have been proposed since CFM was first described in 1881,[6,8–14] but collectively they are limited by the heterogeneity and multiple components of disease phenotypes, ever-evolving understanding of the disease, use of diagnostic modalities, and treatment paradigms. Yet, adequate classification of the deformity dictates the type and timing of intervention and is particularly important for the mandible. In 1969, Pruzansky[15] proposed classification of the dysplastic CFM mandible into 3 types based on clinical examination and plain radiographs.[16] Kaban et al[16] subsequently proposed further subdivision of type 2 deformities based on whether the temporomandibular joint was functional or not. The Pruzansky classification with subsequent Kaban modification has been the most widely used system over the past several decades.

However, the dissemination of computed tomography (CT) with 3-dimensional CT (3dCT) reformats over the past 15 years greatly improved the resolution of craniofacial bony assessment and has shifted the paradigm of CFM mandibular evaluation.[17–19] In parallel, new treatment modalities for CFM, such as mandibular distraction osteogenesis, have been introduced during this same period.[20] In this context of changing diagnostic and treatment patterns, recent publications have questioned the reliability of the Kaban modification of the Pruzansky classification.[21] When we assessed the reproducibility of classifying 38 CFM mandibular defects using the Kaban modification of the Pruzansky classification across 16 craniofacial surgeons, we found a lack of interrater agreement.[22] This constitutes a considerable and far-reaching problem: if surgeons are not cognizant of their disagreement in diagnosis, differing viewpoints about treatment and prognosis are likely to be obscured as well.

We designed a classification of the mandible in CFM based on 3dCT that correlates features of the deformity with surgical treatment and sought to validate it by assessing reproducibility of the classification among craniofacial surgeons.

Disclosure: *The authors have no financial interest to declare in relation to the content of this article. The Article Processing Charge was paid for by the authors.*

METHODS

Classification Based on 3dCT

A classification scheme for the mandibular deformity in CFM was designed and subsequently refined with emphasis on diagnostic specificity and clinical applicability as it relates to treatment, prognostic utility, and ease of use. The classification algorithm was developed, with intent to be similarly applied, using 3dCT reconstructions of the bilateral mandible and skull base in anteroposterior, lateral, and submental views. The input and critique of surgeons from craniofacial centers in several world regions was utilized to incorporate variations of treatment patterns.

The classification incorporates both diagnostic criteria based on 3dCT and corresponding treatment modality(s) based on the severity of deformity (Table 1 and Fig. 1). Each hemimandible is characterized as normal (T0), mild (T1), moderate (T2), or severe (T3 and T4). Type 1 is distinguished from type 0 if the mandible shows noticeable hypoplasia, in particular relative to the contralateral side in hemifacially affected patients. Type 2 is distinguished from type 1 if the ramus shows sufficient vertical deficiency to anticipate an ipsilateral cross bite or overjet of more than several millimeters in childhood but requires condyle that permits functional articulation. Type 3 is distinguished from type 2 by a ramus–condylar complex that is either absent or rudimentary to the degree that it would not likely be functional even if lengthened. Type 3 is distinguished from type 4 by a mandibular body that has sufficient bone stock to support a costochondral graft neocondyle construction, as opposed to requiring a free fibular flap.

Validation of Classification

Patients with a diagnosis of CFM were identified from our prospective craniofacial patient registry, following study review and approval by the institutional review board at the Children's Hospital of Philadelphia. Radiographic records were reviewed, and CT scan data retrieved. CT scans were included if they were performed before any mandibular surgical treatment and of sufficient fidelity to enable fine resolution 3-dimensional (3d) reconstruction. For patients with multiple CT scans meeting inclusion criteria, the scan at oldest age was utilized to enable the broadest distribution of patient ages in our sample. The 3dCT reconstructions of the bilateral mandible and skull base in anteroposterior, lateral, and submental views were assembled for each patient and deidentified. The image sets were then loaded onto an online survey platform (SurveyMonkey, Palo Alto, Calif.)

Table 1. Classification of the CFM Mandibular Deformity

	Type	Diagnostic Features	Anticipated Treatment
0	Normal	Normal	None
1	Mild deformity	Mandible mildly hypoplastic, condyle in normal position	Orthodontics or combined orthognathic surgery and orthodontics in adolescence
2	Moderate deformity	Mandibular ramus moderately deficient vertically, condyle functional	Childhood distraction osteogenesis
3	Severe deformity, adequate mandibular body	Condyle and ramus rudimentary/absent, adequate mandibular body bone stock	Nonvascularized (eg, costochondral) bone graft
4	Severe deformity, inadequate mandibular body bone stock	Condyle and ramus rudimentary/absent, inadequate mandibular body bone stock	Vascularized (eg, free fibular) bone flap

Evaluators were identified from membership in the International Society of Craniofacial Surgery and were selected to represent a range of experience and practice locations. Demographic information for each evaluator was collected including type of training, location, experience, number of patients with CFM seen per year, and CFM classification system used in clinical practice. An attempt was made to capture evaluators from our previous investigation on interrater reliability of the Kaban

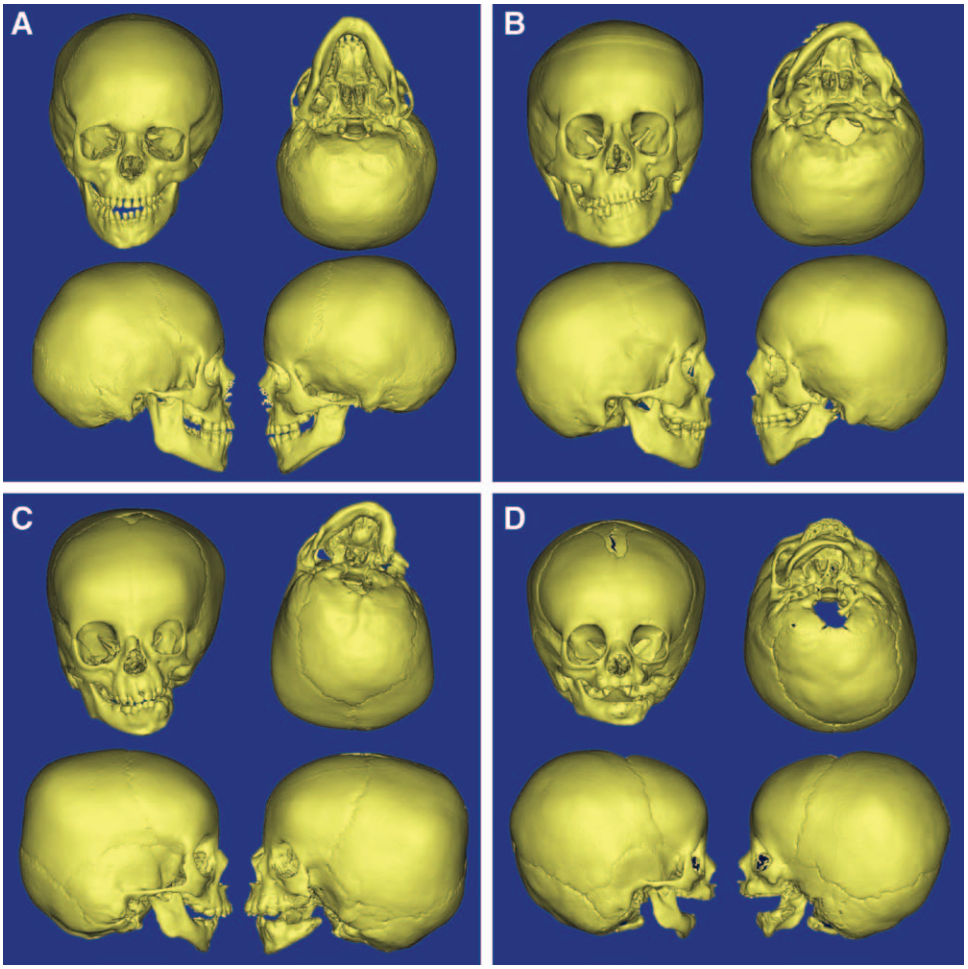

Fig. 1. The 3dCT reconstructions of left-sided CFM mandibular deformities, which are classified as (A) mild (hypoplastic, likely treated with orthodontics or orthognathic surgery; T1), (B) moderate (vertically deficient ramus, likely treated with distraction osteogenesis; T2), (C) severe—adequate mandibular body bone stock (ramus rudimentary or absent, adequate mandibular body bone stock to support costochondral graft), or (D) severe—inadequate mandibular body bone stock (ramus rudimentary or absent, inadequate mandibular body bone stock that necessitates free fibular flap).

modification of the Pruzansky classification[22] to control for intrarater variability and allow for potential comparison of these results with those of our previous study. Each evaluator then reviewed each CFM patient 3dCT radiographs in 4 views and was asked to classify 1 hemimandible (type 0 through type 4) based on the new classification. In hemifacial cases, the reviewer assessed the more severe side, except in 3 patients where the unaffected side was selected to include the type 0 (normal) classification in the evaluation. The order of 3dCT radiographs was independently randomized for each evaluator.

Several of the same 3dCT scans used in the previous assessment of the Pruzansky–Kaban classification[22] were used in this study, to enable comparison of how the 2 classification systems evaluated the same deformity.

Statistical Analysis

Statistical analysis was performed using STATA 12.0 (StataCorp, College Station, Tex.). Percentage agreement was calculated among evaluators. One-way analysis of variance was used to assess for differences in percentage agreement between individual evaluators and individual scoring levels. Fleiss' κ was used to assess interrater reliability using ReCal3 (available online at http://dfreelon.org/utils/recalfront/recal3/; accessed February–May, 2015). Comparison of evaluator percentage agreement between the Pruzansky–Kaban classification[22] and proposed classification was made using a two-tailed *t*-test with equal variances.

Institutional Review Board

This study was approved by the Institutional Review Board of the Children's Hospital of Philadelphia.

RESULTS

The 3dCT images of 43 patients who met inclusion criteria were assembled, which represented a patient age range of 0.1–15.8 years. CFM had been diagnosed as right-sided in 20 patients (47%), left-sided in 19 patients (44%), and bilateral in 4 patients (9%). The 3dCT images were evaluated by 15 craniofacial surgeons, representing a mean 15.2 years of experience (range, 1–40 years) and who each estimate seeing an average of 27 patients (range, 10–80) with CFM annually. Twelve surgeons (80%) were in academic practice; all 15 (100%) received fellowship training in craniofacial surgery, and they reported primarily using the Pruzansky (5, 33%), Kaban (3, 20%), OMENS (5, 33%), or a combination of the 3 (2, 14%) classifications (Table 2).

Table 2. Demographics and Characteristics of Evaluators

	Number (%)
Practice location	
United States	12 (80)
Canada	1 (7)
United Kingdom	2 (13)
Practice type	
Academic	12 (80)
Private practice	3 (20)
Specialty training	
Craniofacial surgery	15 (100)
Years of practice	
1–5	3 (20)
6–10	4 (27)
11–15	2 (13)
16–20	1 (7)
21–25	2 (13)
>25	3 (20)
Approximate no. of hemifacial microsomia patients seen per year	
10	2 (13)
11–20	5 (33)
21–30	6 (40)
31–40	0 (0)
>40	2 (13)

Table 3. Distribution of Classification Types

Type	Mean (%)	Range*	Standard Deviation* (%)
0 (normal)	3.0 (7.0)	0–8	2.2 (5.1)
1 (mild)	8.3 (19.2)	5–11	1.9 (4.3)
2 (moderate)	14.9 (34.9)	7–24	4.3 (10.0)
3 (severe)	11.4 (26.4)	7–18	3.9 (9.0)
4 (severe)	5.4 (12.4)	0–12	3.4 (7.9)

*By evaluator.

On average, evaluators classified 7.0% of patients as normal (T0), 19.2% mild (T1), 34.9% moderate (T2), 26.4% severe (T3), and 12.4% severe (T4) (Table 3). Evaluators demonstrated fair interrater reliability with average pairwise agreement of 50.4±9.9% (Fleiss' κ = 0.34). Average pairwise Cohen's κ was 34.0%. When distinguishing deformities requiring graft or flap reconstruction (T3 and T4) from others (T0–T2), reviewers demonstrated substantial interrater reliability with average pairwise agreement of 83.0±7.6% (κ = 0.64). Average pairwise Cohen's κ was 64.7%. Interrater agreement was slightly lower among evaluators with more experience (more than 13 years in practice, κ = 0.31) compared with those with less experience (fewer than 13 years in practice, κ = 0.37). Interrater agreement was similar between cohorts of surgeons who see a high volume of patients with CFM (25 or more patients per year, κ = 0.34) and those who see a smaller volume (fewer than 25 patients per year, κ = 0.33).

Evaluator agreement using this classification (50.4%; 95% confidence interval, 44.7–56.1) was

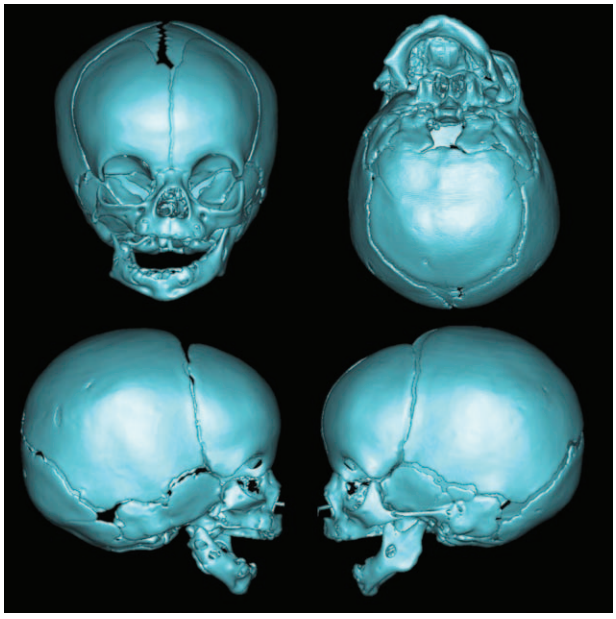

Fig. 2. Reviewers characterized the right hemimandible deformity in this patient as type 3 (66.7%) or type 4 (33.3%). This represents an example of considerably improved interrater agreement compared to when, in a previous study, evaluators were asked to categorize the same 3d reconstructions using the Pruzansky–Kaban classification system.

significantly greater than using the Pruzansky–Kaban classification[22] (39.2%, 95% confidence interval, 34.3–44.1; $P = 0.003$). To further compare the results from this study to those of the previous study, we reviewed the findings from several 3dCT scans that were used in both studies. These were individually illustrative of improved interrater concordance using a new classification system designed for 3dCT. For example, when the 16 evaluators in the previous study were asked to apply the Pruzansky–Kaban classification to the deformity in Figure 2, 6.3% rated it "0," 31.3% rated it "1," 18.8% rated it "2A," 12.5% rated it "2B," and 31.3% rated it "3." When asked to evaluate the same patient's deformity using the same 3d reconstructions in this study according to the new classification, no evaluators rated it "0," "1," or "2"; 66.7% rated it "3"; and 33.3% rated it "4."

DISCUSSION

The widespread dissemination of 3dCT has revolutionized the evaluation of many craniofacial conditions for diagnosis, preoperative planning, and postoperative assessment. The variability and involvement of multiple anatomic components of CFM render 3dCT particularly valuable; the 3d dentoskeletal and soft tissue views provide fine resolution for analysis.[23] The 3dCT facilitates evaluation of deformity characteristics that enable decision making involving more recently popularized treatment techniques; for example, assessing mandibular body inadequacy in the setting of an absent ramus/condyle unit to support choice of a free fibular flap. Several studies attest to interest in a renewed classification of the CFM mandibular deformity in the era of 3dCT[22,24]; however, none have fully considered the evidence base or taken steps to validate a classification across a large series of patients.

An optimal diagnostic classification system should categorize a disease entity, facilitate communication among physicians and patients, and potentially guide treatment and prognosis. The different classes should be mutually exclusive and collectively exhaustive. Balancing a classification's specificity to distinguish nuances of the disease, ability to unify sufficient patients into groups to speak meaningful about them, and simplicity to be integrated into routine clinical use by a variety of surgeons, is all challenging. The challenge is compounded for a disease process that affects different parts of the head and neck heterogeneously and for which surgical treatment techniques vary among surgeons and generally lack an evidence base. The historical record of evolving classifications for CFM reflects this tension, and different classification systems prioritize one over the other.[6,8–17]

The proposed classification for the CFM mandibular deformity is based on 3dCT diagnosis and incorporates a common treatment modality for each type. We found that expert evaluators demonstrated fair interrater reproducibility with average pairwise agreement of 50.4% using this classification. This was a significantly higher degree of interrater agreement than found when similar evaluators used the Kaban modification of the Pruzansky classification (average pairwise agreement 39.2%.)[22]

We further found that this classification lended to substantial agreement ($\kappa = 0.64$) among evaluators distinguishing deformities requiring graft or flap reconstruction (T3 and T4) from others (T0–T2). This is a considerable clinical threshold, because it differentiates between deformities that mandate treatment with tissue transfer and those that can be suitably treated with distraction or orthognathics. When evaluators used the Pruzansky–Kaban classification for the mandible in Figure 2, for example, 56% labeled the deformity as class 0, 1, or 2a and 44% labeled it class 2b or 3—in other words, evenly split on whether tissue transfer would be typically indicated for reconstruction. Using the new classification, 100% of the evaluators considered it to be T3 or T4—either of which typically indicate tissue transfer for reconstruction. Furthermore, the breakpoint between T3 and T4 deformities may represent

the most subtle clinical distinction, given the subset of surgeons who do not use microvascular-free tissue transfer and would perform costochondral grafting for both deformity types.

Kaban et al[25] were critical of the previous study,[22] which found low interrater reliability among 16 craniofacial surgeons who used the Pruzansky–Kaban classification, because "their average of 15 years' of experience does not ensure that the Pruzansky and Kaban system was used correctly." We would disagree that an effective classification system should be predicated on "correct" usage—that it should be limited to those who have received specific training, been certified in its use, or some such arrangement. We believe that a classification is most effective when it is understandable, inclusive, and facilitates communication among professionals regardless of their depth of experience. Our findings that the interrater agreement was consistent between surgeons with higher- and lower-volumes of CFM patients, and slightly higher among surgeons with relatively less rather than more seniority, suggests that this classification is accessible and applicable across a range of levels of experience. That the interrater agreement of the Pruzansky–Kaban classification was limited—even among surgeons who all used the Pruzansky or full OMENS system to classify patients with CFM in their respective practices—speaks to its limited reproducibility in a period where 3dCT use has been widely adopted. But this may not discount its utility, say, within a single institution where tradition or formal training may better reinforce its correct use.

Classification of a surgical disease is also most useful when it guides surgical decision making. We learned from discussing our work on this classification with other craniofacial surgeons that the anatomical variations of greatest interest were the ones that influenced branch points in their own surgical algorithms for CFM management. Two imperatives, thus, shaped our refinement of the classification: what are the most common features of algorithms for managing the mandible in CFM and what is the evidence base for these practices? For instance, the zygomatic arch in CFM demonstrates variability that may not correspond with the mandible other OMENS features,[26] yet if deformed may suggest to the surgeon the need to construct a neo-temporal fossa.[27] Given reports of high ankylosis rates associated with temporomandibular joint reconstruction as opposed to apposition of the rib/fibular construct with the skull base in type 3 or 4 deformities, we did not subclassify based on zygomatic arch deformity or degree of medialization of the condylar remnant. As another example, the degree of soft-tissue deficiency in a patient with CFM could influence the decision making for mandibular bony reconstruction. A free fibular flap with a large myofascial cuff would augment the soft tissue to a greater degree than a costochondral graft, given a patient with borderline mandibular body bone stock who could otherwise be a candidate for either procedure. Nonetheless, a patient with sufficient mandibular body bone stock (type 3) could just as easily undergo costochondral grafting, with structural lipoaspirated fat grafting to treat the soft-tissue deficiency. Furthermore, Lauritzen et al[13] point out that following adequate skeletal reconstruction in CFM, there may not be need for subsequent soft-tissue augmentation.

Thus, we aimed to streamline this classification to the fewest essential anatomic components to drive decision making, acknowledging that borderline cases could and should be decided by factors such as soft-tissue deficiency. We note that emphasizing the role of bony 3dCT does not dismiss the importance of soft-tissue evaluation and physical examination in CFM. The latter are important, but in our opinion are downstream modifiers of decision making after the 3dCT rather than the other way around.

The obvious advantage of a system that incorporates not only diagnostic classifications but also corresponding treatment modality is that it more directly guides surgical decision making. Lauritzen et al[13] used this approach to CFM in 1985 by proposing a treatment-guidance scheme dividing skeletal deformities based on management considerations. There are 2 potential disadvantages, however. First, as treatment modalities evolve, a classification that prescribes certain treatments' risks becoming obsolete. Second, in a situation where surgeons agree on a type of deformity but disagree on the appropriate treatment for it, a classification that prescribes certain treatments' risks objection from those who disagree with therapy only. While acknowledging these 2 limitations, what we discovered was that what many surgeons consider the most relevant anatomy from a diagnostic standpoint was that which distinguished among the appropriateness of different surgical interventions. In other words, what was diagnostically most relevant was intrinsically tied to the treatment options. As new treatment paradigms appear in the future, so likely will new aspects of the deformity become important from a diagnostic standpoint. Hence, even if the classification did not set out prescribed treatment for each type of deformity, new treatment innovations are still likely to render its diagnostic value obsolete. Given this, we prioritized designing a classification to be most fully useful to the current paradigm of treatment, rather than to try and limit its usefulness now to somehow extend its lifetime in the future.

Several limitations of the study warrant discussion. The distribution of our evaluating surgeons

is concentrated in North America and Europe, and future studies would benefit from representation of other regions. The classification scheme is tied to the 3dCT, and as the role of diagnostic modalities using ionizing radiation evolves, alternative diagnostic tests may supplant 3dCT, which could make this classification scheme less useful or even obsolete. Furthermore, because the spectrum of mandibular hypoplasia in CFM falls on a continuum, and intrinsic breakpoints do not exist to distinguish inherent deformity types to which responses could be compared, we used interrater agreement as the benchmark of validity. Next, the pool of raters overlapped with the previous study of the Pruzansky–Kaban classification, and their improved agreement could be due in part to repeat testing bias. Although this is conceivable, the 2-year interval between studies is likely to have eliminated much retention. Furthermore, given that this new classification was shared with evaluators only at the time they completed the web-based survey, their familiarity with it was limited; this should have biased our results disadvantageously, if anything. Finally, we attempted to reduce design bias by using 5 classification options to mirror the methodology of the previous study.

In conclusion, the proposed classification shows significantly improved agreement among surgeons in stratifying the CFM mandibular deformity compared with existing classifications. It shows substantial agreement among evaluators distinguishing deformities requiring tissue transfer-based reconstruction compared to those that do not. The improved, but only moderate, agreement across all deformity types likely reflects the challenges inherent to a disease with heterogeneity, a continuum of severity, for which many differing treatment modalities exist. Nonetheless, improved diagnostic agreement will hopefully prove to be an enabling step toward establishing a more firm evidence base in treatment and prognosis.

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ACKNOWLEDGMENT

We wish to thank Priya Reddy, MD, for her assistance with manuscript preparation.

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