INTRODUCTION
Second to only cleft lip and palate, hemifacial microsomia (HFM), also referred to as craniofacial microsomia, is one of the most common congenital craniofacial anomalies. Now recognized as a spectrum, HFM mainly affects the derivatives of the first and second branchial arches in a unilateral manner, primarily manifesting through the orbit, mandible, auricle, facial nerve, and/or soft tissue. It has been posited that microtia is in itself an iso-type of HFM. Although many technical advancements have been made over the years, microtia reconstruction remains a challenge among this patient population.
Microtia is present in up to 66%–99% of all HFM cases6–7 and has been shown to significantly hinder patients’ psychosocial well-being.8,9 The reconstructive principles and techniques pertaining to microtia reconstruction are less reliable in this patient population due to the ipsilateral microsoma and subsequent asymmetry. Firstly, relying on contralateral measurements relative to the midline often results in the auricular construct being positioned too posteriorly.10 Also, the remnant earlobe and/or vestige skin is often located too anterior/inferior, and therefore cannot be used as an indication to the ear’s ideal location.11 This also makes reconstructing the earlobe more technically challenging.12 Furthermore, in patients with more severe facial asymmetry and hypoplasia, creating a construct identical in size to the contralateral side’s may result in an ear that appears disproportionately large.10 Classical coverage techniques are also unsuitable, mainly due to the taut and thin retroauricular skin,13,14 the underdeveloped temporoparietal fascia and underlying temporal muscle,12,15 the low hairline causing a lack of non-hair-bearing skin,13 and the unreliable arterial anatomy14,16. These factors, combined with a depressed temporal bone, may also hinder successful ear elevation with an appropriate projection and cranioauricular angle.12,17

In light of the many unique challenges plastic surgeons face regarding microtia reconstruction in HFM patients, the authors aimed to synthesize the literature to retrieve challenge-specific recommendations and highlight the strengths and limitations of each. Given the high degree of variability among observed techniques and the lack of a gold standard for treatment, a systematic review of the literature was deemed of utmost importance. The authors hope this review can provide an evidence-based guide for plastic surgeons and will ultimately improve surgical care for this patient population.

MATERIALS AND METHODS

A systematic search of the Pubmed, Medline, and Embase databases was carried out in line with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines,18 using the following search strategy: (“oculoauriculo-vertebral” OR “Goldenhar” OR “hemifacial microsoma” OR “craniofacial microsoma” OR “otomandibular dysplasia”) AND (“microtia” OR “ear”). The search was confined to the English and French languages, and articles from all years were considered. Following duplicate removal, the resultant articles were assessed by two independent reviewers according to strict inclusion and exclusion criteria (Fig. 1). Discrepancies were resolved by means of consensus. All studies that discussed their surgical microtia reconstruction technique in the context of HFM patients were included, regardless of study design. Those that were nonspecific to HFM, that discussed other types of craniofacial anomalies (eg, Treacher Collins), that did not touch on their microtia reconstruction technique, that described prosthetic-based reconstruction, or that did not have a retrievable full text were excluded. Qualitative data regarding study design, type and number of patients, surgical modalities used, follow-up times, surgical results, challenges addressed, specific recommendations, and their respective strengths/limitations were extracted. All included studies were assessed for level of evidence according to the Oxford Centre for Evidence Based Medicine criteria19 and all recommendations retrieved were given a grade according to the ASPS Scale for Grading Recommendations.20

RESULTS

Following the removal of duplicates, the search yielded 762 studies, of which 11 met the inclusion criteria (Fig. 1). The majority were case series (n = 7/11), and follow-up times varied from 3 months to 10 years, with five studies not reporting any follow-up time. The majority of the studies utilized autologous rib graft as their surgical modality (n = 9/11). Both two-stage (n = 6) and three-stage techniques (n = 5) were described. In terms of level of evidence, seven studies were assigned a grade 4 level, while the remaining four were considered level 2b. These findings are summarized in Table 1. Regarding challenges addressed, seven studies (64%) presented techniques for the ideal placement of the construct,12,13,21–23,25,26 six (55%) addressed the low-set hairline,12,13,21–23,25,26 five (46%) provided recommendations to improve construct projection,12,13,21,22,23,25,26 two (18%) commented on how to utilize the remnant earlobe/vestige skin,11–14,23,26 five (46%) provided recommendations to improve construct projection,12,13,21,22,23,25,26 two (18%) commented on the anomalous course of the neighboring neurovascular systems,22,26 two expounded on their techniques for retroauricular coverage of the construct,21,22 and finally, two provided tips on how to achieve the most appropriate construct size.10,25 (See table, Supplemental Digital Content 1, which displays the challenge-specific recommendations and their respective advantages/disadvantages. http://links.lww.com/PRSGO/C145.) The individual recommendations from each study were consolidated, yielding a total of 22 that were of either grade C (n = 5) or D (n = 17) (Table 2).

DISCUSSION

To the best of our knowledge, this is the first systematic review addressing microtia reconstruction in HFM...
patients. This is an especially important endeavor given the relatively low quality of evidence regarding auricular reconstruction and the many persisting surgical challenges associated with this patient population. Eleven studies were included in this review, and these yielded 22 graded recommendations regarding the eight HFM-specific challenges identified (Fig. 2). Most recommendations were found to be of low level of evidence, although all authors report successful outcomes with their respective techniques.

Challenges Addressed

Regarding placement of the constructed auricle, most authors agree that the appropriate vertical location can be accurately determined while in the front-facing position, using the healthy earlobe as a guide to the inferior-most position. The main problem arises when attempting to determine the ear’s antero-posterior location. Mirroring distances from the lateral canthus to the helix and from the oral commissure to the lobule, as classically taught, will result in an ear positioned far too posteriorly. Using such principles as a guide, the surgeon must then subjectively determine the optimal placement to produce a more harmonious aesthetic result. Although no universal consensus exists to remedy this problem, multiple options have been proposed. Park and Park suggest using the posterior margin of the mandibular ascending ramus as a guide, while Qian et al. propose placing the crus of the helix 1.5 to 2 cm from the temple. Regarding the ear’s proper axis, the literature is no different from classical microtia reconstruction in that it recommends referencing the line along the nasal dorsum, and using the contralateral ear to determine the appropriate cranioauricular angle. Other studies propose novel methods of determining the ear’s ideal location, such as using stereophotogrammetry-based morphing of the unaffected ear onto the affected side through

Fig. 1. PRISMA flow chart for systematic review.
landmark-based facial proportions or by using a 3D printed model of the patient’s head and subjectively determining the best possible location. Some even advocate for skeletal correction before ear placement or waiting until at least 8 years of age to mitigate this challenge. Although auricle positioning was the most commonly addressed issue, some studies were nevertheless vague regarding their technique for placement of the construct, which is concerning given that this can be a major determinant of overall surgical success. Interestingly however, through their precise computer modeling, Coward et al. showed that freehand techniques yield fairly precise placement, potentially obviating the need for more sophisticated methods.

Regarding the abnormally low-set hairline, most authors are proponents of laser hair removal before undergoing two-stage reconstruction. Preoperative laser epilation facilitates construct inset and prevents potential thermal damage to the construct and overlying soft tissue. However, in three-stage reconstruction with an expander, Chen et al. demonstrate in their case series of 41 patients that treatment during the expansion period required fewer sessions than if started pre-operatively ($P < 0.001$). Qian et al. echo this, and explain that laser hair removal is more effective when the skin is thinner. Finally, Firmin also stresses the importance of sideburn reconstruction for optimal aesthetic appearance, through either a scalp transposition flap or a hair transplant.

Regarding soft tissue coverage of the constructed auricle, most of the included articles advocate for a three-stage technique, whereby the first consists of expanding the retroauricular skin. Tissue expansion obviates the need for a fascial flap and skin grafts, which have been reported to result in uneven spotty skin, more visible scars, and alopecia. Despite slightly prolonging the overall treatment duration, it renders better aesthetic outcomes and preserves the temporal fascia for potential management of framework exposure in the third stage if necessary. However, in a large comparison of three coverage techniques, Park and Park conclude that in addition to being slow and cumbersome, the expansion technique yields the poorest results. They recommend the “embedded and elevation” technique in mild cases (mild to moderate degree of low hairline, usable remnant vestiges, mastoid skin available) and a grafted temporoparietal fascia flap in severe cases (mastoid depression, low hairline, small and/or remarkably low-set ear vestiges). It should be noted that some cases of fascial flap reconstruction resulted in poorly defined anteroauricular subunits from thick or contracted fascia and color mismatch from the scalp graft. This was, however, easily remedied by secondary debulking and regrafting with contralateral postauricular skin. Should surgeons still prefer the two-stage method, Yamada et al. suggest using a mastoid flap with a subcutaneous pedicle, combined with a temporoparietal fascia flap and skin graft, whereas Nuri et al. suggest using a pericranial flap instead. Surgeons should also remain suspicious of the quality of the vascular supply to the fascial flap, and may need to raise the superficial/deep temporal fascia in one piece.

Regarding earlobe reconstruction and usage of the remnant vestige skin, most authors propose carrying out a retrograde lobular transposition at either the first stage (in two-stage reconstruction) or the third stage (three-stage expander-based reconstruction). It provides a natural appearance and ideal location, and allows for earings. However, it can sometimes require future revision due to an “arch bridge” deformity. Retrograde lobular transposition is indicated when the vestige ear is located lower than usual (which is most often the case), but a mix of a V-Y advancement, Z-plasty, and/or lobular transposition may be used when the vestige is slightly higher than that. If one finds that the vestige skin cannot be used, it can safely be removed, in which case the earlobe is to be included in the construct design through a long helix and addition of remnant cartilage. Some even propose using the removed vestige skin as a skin graft for coverage of the fascial flap.

Regarding construct projection, surgeons need to take into account the degree of hypoplasia of the structures upon which the ear sits (mastoid, zygomatic process, temporal condyle). Lee and Oh also surmise that hypoplasia of the superficial temporal artery and its posterior branches might create an unfavorable environment for graft take, leading to partial graft loss, secondary wound contracture, and eventually insufficient projection. Regardless, the literature agrees that this should be compensated for, mainly by placing the conchal bowl more superiorly, to the upper, flat, and solid part of the temporal bone. In their embedded and elevation technique, Park and Park suggest inserting a preserved cartilage block underneath the framework during elevation. Similarly for porous polyethylene (PPE)-based reconstruction, Kimura et al. propose determining the correct projection with the help of a 3D printed model of the patient’s head, and then re-creating it intra-operatively by augmenting the construct with separate PPE blocks.

Regarding the anomalous superficial temporal artery and facial nerve, Firmin first suggests the use of a preoperative Doppler to map out the artery’s location, as it is often more posterior than expected. Surgeons should also be mindful as to the possible aberrant course of the facial nerve while elevating flaps. Firmin describes a case of iatrogenic injury to the frontal branch of the facial nerve coursing through the vestige skin resulting in ipsilateral paralysis of the frontalis. Yamada et al. also promote caution while raising the fascial flap, to avoid violation of the temporal branch of the facial nerve, and warn surgeons to be wary of the popular belief that an incision is “safe within the hairline.” The literature even reports a case of Frey’s syndrome following microtia reconstruction in a HFM patient, due to this aberrant nerve anatomy.

Regarding retroauricular coverage of the constructed auricle, skin grafting is most commonly used; however, the presence of arterial anomalies can negatively affect its healing process. Nuri et al. thus recommend using a free serratus fascial flap to cover the posterior aspect of the cartilage, whereas Park and Park describe a fan-shaped mastoid fascia flap.
| Author            | Year       | Study Design | Level of Evidence | Patients | Surgical Modality                  | Follow-up | Challenge(s) Addressed                                                                                       | Results |
|-------------------|------------|--------------|-------------------|----------|------------------------------------|-----------|----------------------------------------------------------------------------------------------------------------|---------|
| Chen et al.       | (2020)    | Case series  | 4                 | n = 41   | Low hairline Autologous rib graft 3-stage | 13.3 mo   | Earlobe reconstruction/usage of vestige skin • 90.2% were satisfied with the reconstructed ear, the rest were partially satisfied, none were unsatisfied • Expander leakage happened in five cases (12.2%) in late expansion period and were managed by expander replacement operation under local anesthesia • Similar rate of hospital stay (about 4 d to 1 wk for each stage) and recovery time (6 mo–1 yr) | Earlobe reconstruction/usage of vestige skin |
| Cheng et al.      | (2015)    | Case series  | 4                 | n = 42   | Moderate/severe HFM Lobule-type microtia Autologous rib graft 2 and 3-stage (mixed) | NS        | Earlobe reconstruction/usage of vestige skin • All lobular transpositions survived • Delayed healing due to local hematoma (n = 2) • Vaulted earlobes (n = 4) | Earlobe reconstruction/usage of vestige skin |
| Chowchuen et al.  | (2011)    | Case series  | 4                 | n = 23   | Autologous rib graft 2-stage | NS        | Location of the construct • Morphed ears were positioned slightly farther forward on the face than the natural ears in relation to nasion and subnasale (due to their smaller dimensions) • The vertical positions were generally very similar • Length, width, and insertion lengths of the natural ears generally were very similar to the morphed and artificial ears, and no statistically significant differences were found. | Location of the construct |
| Coward et al.     | (2014)    | Case series  | 4                 | n = 10   | Not specified | NS        | Size of the construct • Anomalous course of frontal branch of facial nerve • Anomalous course of superficial temporal artery • Projection of the construct • Location of the construct | Size of the construct |
| Firmin et al.     | (2001)    | Case series  | 4                 | n = 139  | Autologous rib graft 2-stage | NS        | Anomalous course of frontal branch of facial nerve • Anomalous course of superficial temporal artery • Projection of the construct • Location of the construct | Anomalous course of frontal branch of facial nerve • Anomalous course of superficial temporal artery • Projection of the construct • Location of the construct |
| Kimura et al.     | (2021)    | Prospective cohort   | 2b                | n = 6 grade III microtia Porous polyethylene1-stage | 3–4 months | Location of the construct | At 4 yr follow-up, in both cases and projections of the constructed ears were satisfactorily maintained. | Location of the construct |
| Nuri et al.       | (2017)    | Case series  | 4                 | n = 2    | Autologous rib graft 2-stage         | 4 yr      | Retroauricular coverage • Early postoperative infections or delayed healing of the coverage tissue did not occur in any of the reconstructed cases • 11 cases (21.2%) showed delayed resorption of the grafted cartilage | Retroauricular coverage |
| Park and Park     | (2018)    | Retrospective chart review | 2b                | n = 52   | Autologous rib graft 2, 3, and 4-stage (separate) | 33 mo (range: 6 mo–0 yr) | Coverage of the construct • Location of the construct • Low hairline • Projection of the construct • Retroauricular coverage • Size of the construct | Coverage of the construct |
| Author (Year) | Study Design  | Level of Evidence | Patients | Surgical Modality | Follow-up | Challenge(s) Addressed | Results |
|--------------|---------------|-------------------|----------|-------------------|-----------|------------------------|---------|
| Qian et al (2017) | Retrospective chart review | 2b | n = 111 | Autologous rib graft 3-stage | 8.3 mo (range: 5–20 mo) | Coverage of the construct | • 103 patients (92.8%) had satisfactory outcomes, seven patients (6.3%) had partially satisfactory outcomes, and one patient (0.9%) had an unsatisfactory outcome.
• First stage: hematoma in two cases (1.8%), infection of the tissue expander in one case (0.9%), necrosis of expanded skin in two cases (1.8%), and leakage of the tissue expander in one case (0.9%).
• Second stage: poor skin graft survival in one case (0.9%) and cartilage framework exposure in two cases (1.8%). |
| Xing et al (2020) | Retrospective chart review | 2b | n = 69 Goldenhar syndrome 53 lobule-type microtia, 16 conchal-type | Autologous rib graft 3-stage | (range: 6 mo–7 yr) | Coverage of the construct | • One case suffered expander leakage
• Hematoma occurred in four cases
• One case suffered skin necrosis and cartilage exposure
• The reconstructed ears showed correct position, clear outline, invisible scarring and contended bilateral symmetry. Sixty-three patients and their families (91.3%) responded satisfied or partially satisfied outcomes. |
| Yamada et al (2009) | Case series | 4 | n = 6 | Autologous rib graft 2-stage | NS | Anomalous course of facial nerve Coverage of the construct | • In types 1 and 2, the size of the reconstructed ear and the definition of the auricle seem to be well maintained, whereas in type 3 hemifacial microsomia, the auricle shrinks and the definition becomes poor over time. |
Finally, regarding sizing of the constructed auricle, Park and Park\textsuperscript{25} note that the vertical length should be made slightly smaller in severe HFM cases. This is due to the illusory effect that the reconstructed ear’s uppermost portion is higher than that of the contralateral ear when placing its lobule at the same height. Most authors do not
specify their technique for sizing, and likely replicate the normal ear’s size by freehand techniques. This seems to be a fairly reliable method, as Coward et al. demonstrated that as per their highly precise computer-based morphing technique, no appreciable differences were detected between their predicted construct size and those of the already reconstructed artificial ears in their HFM cohort.

Other Considerations

It is worth mentioning that many of the categories discussed above may be impacted by the type of construct used (ie, PPE vs. autologous rib graft). The use of a PPE-based construct may require some form of adaptation to the challenges encountered. Namely, given the precarious nature of the superficial temporal system, classical coverage with a superficial temporal fascia flap may need to be revisited. Also, staged hair removal may need reconsideration, as this procedure is usually done in one stage when a polyethylene framework is used. Another option always available is an ear prosthesis, despite most surgeons agreeing that microtia is best addressed among HFM patients through surgical means. Many studies have shown that prostheses can be reliably used in the HFM population, and with fairly low complication rates. Finally, it is also important to be mindful that microtia often presents with concomitant hearing loss. Surgeons should consider treatment of the external ear and the hearing apparatus as a unified system, especially since treating hearing impairment has been suggested to have an even greater positive psychosocial impact than microtia reconstruction. Obtaining a CT-scan of the temporal bone between 5 and 6 years of age to assess the external ear canal and the middle/inner-ear structures should be standard of care.

Limitations and Future Directions

The main limitation of this review is the low level of evidence of the included studies, decreasing the strength of the conclusions drawn from this synthesis. This review serves to further emphasize the need for large, prospective studies to determine the optimal surgical modalities for treating microtia in hemifacial microsomia patients. While 3D printing is becoming more accessible, some authors have proposed creating personalized splints anchored on the maxilla to guide proper placement in microtia reconstruction. Although this exact model cannot be used in HFM patients, perhaps an adaptation of such could be devised. This is especially intriguing, considering that many authors hold that the auricle is in reality not located in the face mask, but rather on the temporal bone and thus its proportions with cranium should be unaffected. Other than construct placement, there is yet to be a consensus regarding the superiority of construct coverage techniques, which may steepen the learning curve for young surgeons. This question would benefit from future, multi-center studies, as the relative rarity of this condition likely explains the paucity of a consensus. It is also curious to note that the majority of the studies included in this review originate from centers located in Asia. This points at a potential publication bias and a lack of general interest among researchers from other parts of the world. Given that this condition is no less common in North America, the authors hope this review encourages surgeons to continue reporting their outcomes in the field. Finally, although not a classical indication, surgeons should expect that in the wake of the COVID-19 pandemic, more patients may seek microtia reconstruction in general to allow for adequate mask wear, further strengthening the timeliness of such a consolidation.

CONCLUSIONS

Although HFM is one of the most common craniofacial anomalies and most of its population has microtia, many challenges regarding its surgical treatment have persisted. This is why the authors sought to retrieve evidence-based recommendations regarding these challenges, which are graded and presented in this review. By providing surgeons with a consolidation of available options and their respective level of evidence, the authors hope a transition toward a more evidence-based care for microtia reconstruction in HFM patients will be facilitated.

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