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Review

Long-term aesthetics, patient-reported outcomes, and auricular sensitivity after microtia reconstruction: A systematic review

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Summary *Background:* Auricular reconstruction for microtia is most frequently performed using autologous costal cartilage (ACC) or porous polyethylene (PPE) implants. Short-term results are generally promising, but long-term results remain unclear. Long-term outcomes were explored in this systematic review, and minimal reporting criteria were suggested for future original data studies.
Methods: A systematic literature search was conducted in MEDLINE, EMBASE, and the Cochrane Central Register of Controlled Trials from inception through October 14, 2020. Articles on auricular reconstruction in patients with microtia using ACC or PPE were included if postsurgical follow-up was at least 1 year. Outcome reporting was split into separate publications, and results on complications were reported previously. This publication focused on long-term aesthetic, patient-reported, and sensitivity outcomes.

Presentation at meetings: This paper has not been presented at any meetings, neither wholly nor in part.
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Results: Forty-one publications reported on these outcomes. Both materials led to aesthetically pleasing results and high rates of patient satisfaction. ACC frameworks grew similarly to contralateral ears, and the anterior surface of auricles regained sensitivity. Furthermore, postoperative health-related quality of life (HRQoL) outcomes were generally good. Data synthesis was limited due to considerable variability between studies and poor study quality. No conclusions could be drawn on the superiority of either method due to the lack of comparative analyses.

Conclusion: Future studies should minimally report (1) surgical efficacy measured using the tool provided in the UK Care Standards for the Management of Patients with Microtia and Atresia; (2) complications including framework extrusion or exposure, graft loss, framework resorption, wire exposure and scalp/auricular scar complications and (3) HRQoL before and after treatment using the EAR-Q patient-reported outcome measure (PROM).

Contents

Introduction

Microtia is a rare congenital malformation of the auricle with an incidence of around 2 in 10,000 births globally[1,2]. The malformations are commonly seen unilaterally, accompanied by atresia of the acoustic meatus as well as malformation of the auditory ossicles[3].

Auricular reconstruction using auricular costal cartilage is preferred by surgeons globally[4,5]. This technique has a relatively low complication rate[6] and provides durable results[7] due to the innate healing ability of autologous cartilage[8]. However, the steep learning curve[4,5,9] and possible donor-site morbidity[10] associated with autologous costal cartilage (ACC) has paved the way for alloplastic techniques, such as porous polyethylene (PPE) implants[11,12]. PPE implants, such as Medpor®, may provide excellent aesthetic results[5], but implant exposure is a potentially devastating complication as infection following exposure can necessitate explantation[13].

Surgical outcome assessment is highly variable and subjective, and the lack of standardized outcome assessments hampers comparability of results and the implementation of new techniques. Aesthetic results are often assessed by the surgeons themselves[14]. However, due to the aesthetic and functional aspects of microtia reconstruction, patient-reported outcomes are crucial to evaluate the benefits of surgery[15]. Furthermore, the stability and longevity of reconstructive results are vital since most patients undergo reconstruction as children.

To our knowledge, long-term outcomes of microtia reconstruction have not been reviewed. This study therefore aimed to systematically review the literature for long-term microtia reconstruction outcomes, with a post-surgical follow-up of at least 1 year. Our results on long-term complications were published previously[16]. Furthermore, we aimed to propose minimum reporting criteria to standardize outcome reporting in future studies.

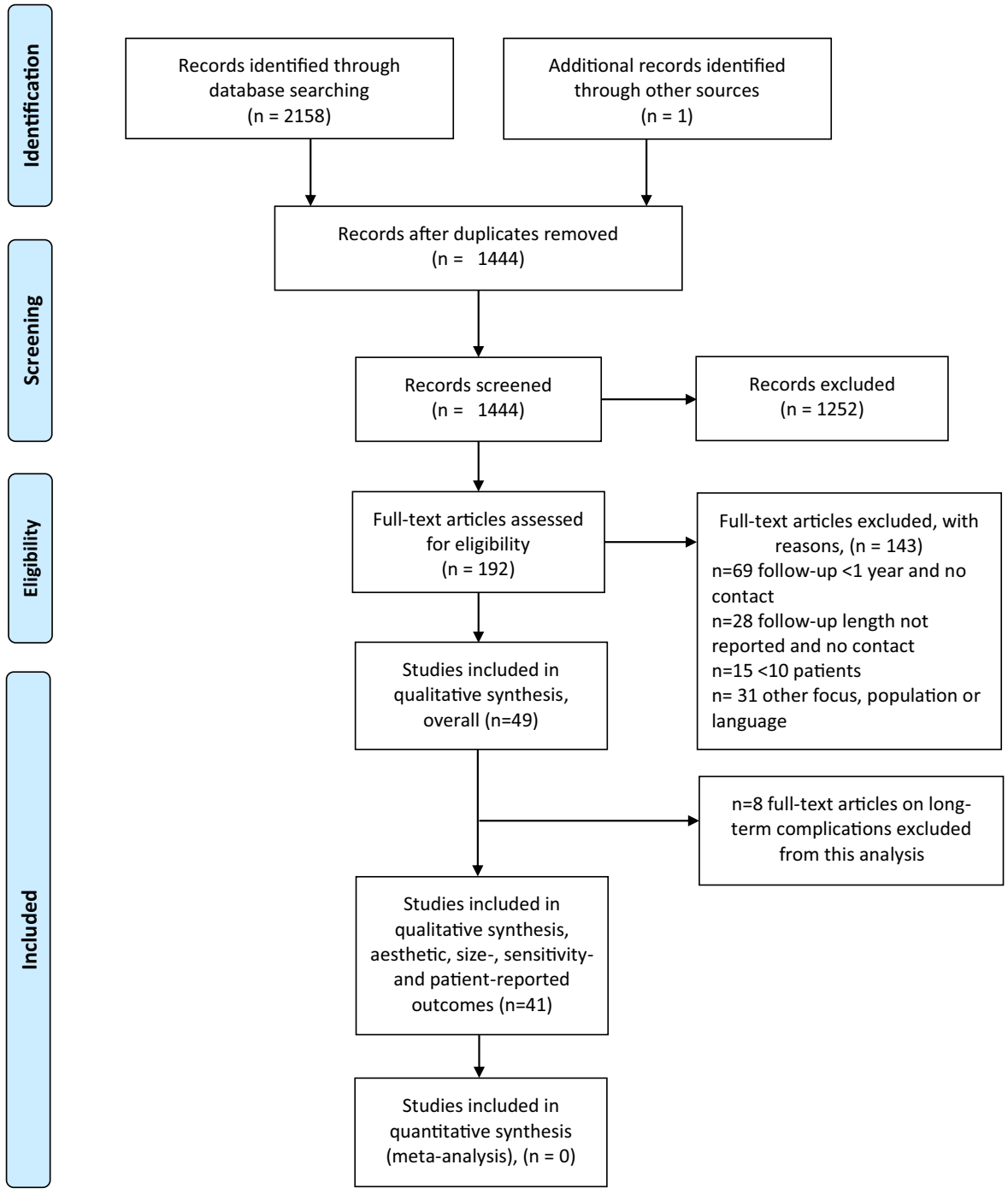


Fig. 1. PRISMA flow diagram of study inclusion process.

Methods

This systematic review was conducted in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA)[17] guidelines. A protocol was registered in Prospero (ID: CRD42020182099). We performed a systematic search of the available literature to identify publications on microtia reconstruction using ACC or PPE frameworks with a postoperative follow-up period of at least one year. The search strategy was created by a clinical librarian with experience in systematic literature searches at our center (FSEJ). The search was conducted in MEDLINE (PubMed interface), EMBASE (OVID interface) and the Cochrane Central Register of Controlled Trials (Wiley interface) from inception until the 22nd of April 2020 by a clinical librarian (FSEJ) and updated on the 14th of October 2020 by a reviewer (EMR). Conference abstracts, letters and editorials were excluded. The full search strategy is available in the Supplementary data file 1.

Table 1. Study characteristics.

First author and year of publication	Design	No. of cases, long term (total)	Patient age, years	Type of microtia, no	Implant	Surgery details: no. of stages, TE use, fascia flap use	Follow-up, years
Tanzer 1978	Cross-sectional	43 (44)[a]	6-20	-	ACC	3 ($n = 3$), 4 ($n = 22$), 6 ($n = 19$), no fascia	6-19
Thomson 1989	Prospective cohort	43	4 (1-15)	-	ACC	6, no fascia	≥2.5
Brent 1992	Cross-sectional	273 (500)[a]	11.4 (5-62)[b]	-	ACC	3-4, no fascia	5.3 (1-17)
Şengezer 1996	Case series	10	All 21	-	PPE	2, no fascia	1.1
Firmin 1998	Case series	300 (352)	-	24 atypical	ACC	Brent: 3-4 ($n = 184$), Nagata: 2 ($n = 144$), 1 stage; galeal flap in sulcus ($n = 24$)	≥1
Brent 1999 and 2002[c]	Cross-sectional	508 (1000)[a,d]	10.5 (5.5-62)[b]	-	ACC	3-4, no fascia or occipitalis fascia	7.7 (1-18)
Park 2002	Case series	13 (19)	19.1 (11-32)[b]	All dystopic	ACC	1-2, TPFF (around framework)	1-4
Cho 2006	Case series	37	7-51	17 lobule, 20 conchal	ACC	2, MFF	1-5
Dashan 2008	Prospective cohort	118 (366)[e]	5-10 (mode)[b]	329 lobule, 22 conchal, 13 small conchal, 2 anotia	ACC	3, TE, MFF	1-6
Jiang 2008	Case series	138 (3332)	5-11[b]	-	ACC	3, TE, RAF	1-8
Kizhner 2008	Case series	27	-	-	ACC	Nagata	0.8-7.3[f]
Öberg 2008	Cross-sectional	19	14.6 (10-20)	-	ACC	3, no fascia	3.6 (≥2)
Pan 2008	Case series	368	-	-	ACC	3, TE, subcutaneous fascial flap	3-5
Steffen 2008	Cross-sectional	60 (92)[a,g]	20.0 (12-58)[h]	-	ACC	Nagata	2.4 (1-6)
Wang 2008	Case series	438	7 (6-14)	2 anotia, 3 traumatic	ACC	2, TE, RAF	1-2
Jiang 2011	Case series	70	5-17	-	ACC	2, RAF	1-3
Kobayashi 2011	Case series	28	11.2 (10-12)	19 lobule, 9 conchal	ACC	2, TPFF	7.8 (6-10)
Öberg 2011	Cross-sectional	39	10.5 (6-19)[h]	-	ACC	3, no fascia	1.6 (0.5-5)[i]
Park 2012	Case series	19	13.5 (11-24)	All large remnant	ACC	1, MFF ($n = 8$), TPFF ($n = 7$), Grotting flap ($n = 1$), no fascia flap ($n = 3$)	≥1
Zhang 2012	Case series	27	9.2 (5-21)	14 lobule, 7 conchal, 5 small conchal, 1 anotia	PPE	3, TE, RAF ($n = 2$), no fascia ($n = 25$)	1.9 (1-3)
Braun 2013	Cross-sectional	15	18 (10-42)[h]	-	PPE	1, TPFF	4.9 (3-7)
Kim 2013	Case series	20	18.1 (12-49)	9 lobule, 6 conchal, 2 small conchal, 1 anotia, 2 atypical	ACC	Nagata with or without conchal bowl element	>1
Kristiansen 2013	Cross-sectional	59 (78)[a]	14 (9-23)[h]	-	ACC	3, no fascia	4 (0-10)[h,f]
Xu 2013	Retrospective cohort	126	14 (6-28)	80 lobule, 44 conchal, 2 anotia	ACC	2, RAF	2.5 (1.9-3.1)
Constantine 2014	Retrospective cohort	36	ACC 8.0; PPE 6.9 Both: 6.1	11 grade II, 25 grade III	ACC/ PPE	ACC: 3-4, TPFF PPE: 1, TPFF	ACC: 2-11 PPE: 2-6

(*continued on next page*)

Table 1. (*continued*)

First author and year of publication	Design	No. of cases, long term (total)	Patient age, years	Type of microtia, no	Implant	Surgery details: no. of stages, TE use, fascia flap use	Follow-up, years
Xu 2014	Randomized controlled study	216[j]	6-13	143 lobule, 71 conchal, 2 anotia	ACC	Framework modifications	2.1 (1.5-2.9)
Yotsuyanagi 2014a,b	Case series	137	12.2	All lobule type	ACC	2, TPFF	4.8 (3-7.5)
Chen 2015	Case series	18	9.7 (5.7-18.5)	All with CAA	PPE	1, superficial temporal flap	2.2 (1-3.5)
Johns 2015	Prospective cohort	23	6.1 (3-10)	6 grade II, 17 grade III	PPE	-	1[k]
Roos 2015	Retrospective cohort	22	7.5 (6.1-10.6)	-	ACC	3, no fascia	5.6 (2.1-10.6)
Sharma 2015	Case series	55	-	-	ACC	3, postauricular galeal flap	≥1
Akter 2017	Cross-sectional	50/27 (115/48)[a,l]	11 (9.8-14)[h] / 14 (11-19)[h]	-	ACC	2, similar to Nagata	2 (0.7-3)[f] / 1-9
Johns 2017	Prospective cohort	28 (40)	6 (3-10)	21 grade III, 3 grade II, 4 not described	PPE	-	1[k]
Kim 2017	Case series	51	16.5 (12-54)	-	ACC/ PPE	2, ACC framework, PPE elevation block, TPFF ($n = 49$), temporal fascia ($n = 2$)	1-4
Han 2018	Case series	22	-	-	ACC	2, superficial temporal fascia flap	2 (1.7-5)
Denadai 2019	Prospective cohort	38	16.4 (10-34)	22 lobule, 8 conchal, 6 small conchal, 2 anotia	ACC	2, Nagata	≥1 year
Ladani 2019	Case series	53	12.3 (9-34)	42 lobule, 9 conchal, 2 traumatic	ACC	2, MFF or TPFF	1[k]
Zhang 2019	Case series	17	22.3 (12-35)	-	PPE	1, TPFF ($n = 2$); 1, no fascia ($n = 2$); 2, RAF ($n = 13$	1-3
Li 2020	Case series	628	10 (8-31)	385 lobule, 122 conchal, 106 small conchal, 15 anotia	ACC	2, composite wedge elevation block[m], RAF	4[h]

ACC: autologous costal cartilage; PPE: porous polyethylene; MFF: mastoid fascia flap; TPFF: temporoparietal fascia flap; RAF: retroauricular fascia flap; TE: tissue expander; CAA: congenital aural atresia; CAS: congenital aural stenosis.
Values are presented as mean (range) or mean ± standard deviation, unless otherwise specified. Percentages are given if the sample size is larger than 50.

[a] Number of patients that responded (total number of patients who received a questionnaire).
[b] Patient characteristics are from large cohort, not described for follow-up cohort.
[c] Published twice in different years.
[d] Includes cohort from Brent 1992.
[e] Aesthetic assessment only done in 118 patients.
[f] <5% of patients with a follow-up of <1 year.
[g] Questionnaire sent to 100 patients and 68 patients responded including 8 patients with traumatic defects. These patients are excluded from our analysis.
[h] Median (range), in Akter et al. (2017) median (interquartile range).
[i] Population separated into short- (0.5-1.7) and long-term (1.7-6 years) follow-ups.
[j] Population randomized: group A with framework modifications (block of residual cartilage used between tragus and base frame of the inferior crus) $n = 50$; group A, original method, $n = 50$; group B with framework modifications (reinforcement with braided suture), $n = 58$; and group B, original method, $n = 58$.
[k] Follow-up exactly 1 year for all patients.
[l] Population from two centers: Great Ormond Street Hospital (first population) and the Royal Hospital for Sick Children (second population).
[m] Mixture of epoxide acrylate maleic and hydroxyapatite.

Table 2. NOS results for cohort studies and case series.

First author and year of publication	Study design	Selection			Comparability	Outcome			Summary
		Representativeness of exposed cohort[1]	Selection of non-exposed cohort[2]	Ascertainment of exposure[3]	Comparability[4]	Assessment of outcome[5]	Length of follow-up[6]	Adequacy of follow-up[7]	
Thomson 1989	Cohort	No clear description	☆	☆	☆☆	☆☆	☆	☆	**Fair**
Sengezer 1996	Case series	Selected group (male, 21 years)	No control	Written self-report	No control	Self-report	☆	No description	**Poor**
Firmin 1998	Case series	☆	No control	Written self-report	No control	Self-report	☆	☆	**Poor**
Park 2002	Case series	Selected group (dystopic microtia)	No control	Written self-report	No control	Self-report	☆	72%	**Poor**
Cho 2006	Case series	☆	No control	Written self-report	No control	Self-report	☆	☆	**Poor**
Dashan 2008	Cohort	☆	☆	Written self-report	☆	☆	☆	33%	**Fair**
Jiang 2008	Case series	No clear description	☆	Written self-report	No description	Self-report	☆	4%	**Poor**
Kizhner 2008	Case series	Selected group (successful surgeries)	☆	☆	☆	☆☆	☆	☆	**Fair**
Pan 2008	Case series	☆	No control	Written self-report	No control	Self-report	☆	No description	**Poor**
Wang 2008	Case series	☆	No control	Written self-report	No control	Self-report	☆	☆	**Poor**
Jiang 2011	Case series	☆	No control	Written self-report	No control	Self-report	☆	☆	**Poor**
Kobayashi 2011	Case series	☆	☆	Written self-report	☆	Self-report	☆	☆	**Fair**
Park 2012	Case series	Selected group (Large remnant)	☆	☆	☆	☆	☆	☆	**Fair**
Zhang 2012	Case series	☆	No control	Written self-report	No control	Self-report	☆	☆	**Poor**
Kim 2013	Case series	☆	No control	☆	No control	☆☆	☆	☆	**Poor**
Xu 2013	Cohort	☆	☆	☆	☆☆	☆	☆	No description	**Good**

(*continued on next page*)

Table 2. (*continued*)

First author and year of publication	Study design	Selection			Comparability	Outcome			Summary
		Representativeness of exposed cohort[1]	Selection of non-exposed cohort[2]	Ascertainment of exposure[3]	Comparability[4]	Assessment of outcome[5]	Length of follow-up[6]	Adequacy of follow-up[7]	
Constantine 2014	Cohort	☆	☆	☆	☆☆	☆☆	☆	☆	**Good**
Yotsuyanagi 2014ab	Case series	Selected group (lobule type	☆	Written self-report	No description	Self-report	☆	☆	**Poor**
Chen 2015	Case series	Selected group (CAA)	No control	Written self-report	No control	Self-report	☆	☆	**Poor**
Johns 2015	Cohort	☆	No control	☆	No control	☆	☆	No description	**Poor**
Roos 2015	Cohort	Selected group (strict inclusion criteria)	☆	☆	☆☆	☆☆	☆	☆	**Fair**
Sharma 2015	Case series	☆	No control	Written self-report	No control	☆	☆	☆	**Poor**
Johns 2017	Cohort	☆	No control	☆	No control	☆	☆	☆	**Poor**
Kim 2017	Case series	☆	No control	Written self-report	No control	Self-report	☆	No description	**Poor**
Han 2018	Case series	Selected group	☆	☆	☆	☆☆	☆	☆	**Fair**
Denadai 2019	Cohort	☆	☆	☆	☆☆	☆☆	☆	No description	**Good**
Ladani 2019	Case series	☆	☆	Written self-report	☆	☆	☆	☆	**Fair**
Zhang 2019	Case series	☆	☆	Written self-report	No description	Self-report	☆	☆	**Poor**
Li 2020	Case series	☆	☆	☆	☆☆	☆☆	☆	☆	**Good**

CAA: congenital aural atresia; CAS: congenital aural stenosis.

[1] All cohorts/case series that received a star where considered somewhat representative due to consecutive or non-random sampling.

[2] All studies that received a star used the contralateral ear as a control.

[3] Studies that received a star mentioned secure records such as lead plates or photographs or used a structured interview.

[4] One star was awarded if the contralateral (control) ear was described or if only unilateral microtia patients were included and the contralateral ear was used as a control. A second star was awarded for controlling for changes from baseline in the contralateral ear or assessing the preoperative situation (Thomson 1989, Xu 2013, Constantine 2014); or for controlling investigator- and test-related conditions (Roos 2015, Denadai 2019, Li 2020).

[5] Outcomes were assessed by a non-independent investigator using a clearly described or validated tool in studies that received one star. Studies that received two stars assessed outcomes through an independent or blinded investigator (Kim 2013, Constantine 2014, Li 2020), referenced secure records (Thomson 1989, Kizhner 2008, Han 2018) or both (Roos 2015, Denadai 2019).

[6] All studies received a star for a follow-up length of $\geq$1 year.

[7] Studies received a star if $>$80% of patients were followed for $\geq$1 year.

After removal of duplicates, two reviewers (EMR and ME) independently screened all records using Rayyan, a web-based software program.20. Titles and abstracts were screened for relevance, and full-texts of potentially eligible articles were subsequently assessed for inclusion. Studies that reported on the long-term outcomes (i.e. outcomes recorded during a follow-up period of at least one year) of auricular reconstruction using ACC and/or PPE frameworks in patients with congenital microtia were included. Studies that reported a range of follow-up durations and those that reported on acquired defects were included if data for long-term follow-up for microtia patients were reported separately. Authors were contacted if the duration of follow-up was not reported, data on microtia and acquired defects was not separated or when a proportion of patients had been followed up for more than one year, but this data was not reported separately. Studies were included if less than 5% of participants had acquired defects or were followed up for less than 1 year. Studies where more than 5% of the population had acquired defects or were followed up for less than one year were also included in case authors could provide long-term follow-up data on microtia patients. No restrictions were set on the study design, setting or patient characteristics. Disagreements were discussed, and a third reviewer (CCB) was consulted in case of disagreements. Reference lists of included studies were scanned for additional relevant titles.Included studies were divided for data extraction. Two reviewers (EMR and ME) independently extracted relevant data on the study characteristics (design, year of publication, number of patients, number lost to follow-up, number of procedures, surgical technique(s) and follow-up duration), patient characteristics (age, types of microtia and inclusion and exclusion criteria) and outcome data using a predefined form. A random check was performed, and any disagreements were discussed. Primary outcomes, as prespecified in our protocol, were aesthetic assessment, long-term postoperative complications and tactile sensitivity. Secondary outcomes were auricular height and width, patient satisfaction and health-related quality of life (HRQoL). Reporting was split into two publications to enable thorough discussion due to an overwhelming amount of data. The results on complications were published previously[16]. This publication reports on aesthetic outcomes (including size), patient-reported, and sensitivity outcomes. Authors were contacted in case of unclear data.

Risk of bias was assessed independently by two reviewers (EMR and ME). Risk of bias was assessed using the Cochrane Collaboration's tool for randomized studies[18] in Review Manager Version 5.4.1[19], the Newcastle-Ottawa Quality Assessment Scale (NOS) for cohort studies and case series[20] (Supplementary data file 2), as well as an adapted NOS-based tool[21] for cross-sectional studies (Supplementary data file 3). Any disagreements were discussed and resolved by a third reviewer (CCB), where necessary. Studies were rated good, fair, or poor as shown in the supplementary data files.

The Grading of Recommendations Assessment, Development, and Evaluation (GRADE) approach [22] was used to evaluate the overall quality of evidence for the most frequently reported outcomes, using the following domains: risk of bias, inconsistency (unexplained heterogeneity), indirectness, imprecision and publication bias. Our confidence in the evidence was categorized as 'high', 'moderate', 'low' or 'very low' based on the previous domains.

Results

After the removal of duplicates, 1444 manuscripts were screened for inclusion and 1252 were excluded based on title and abstract review. One-hundred and ninety-two full-text articles were assessed for eligibility, and subsequently, 49 publications were included overall.[7,23-62,70-76] The inclusion process is shown in Fig. 1. Forty-one studies reported on aesthetic outcomes, including auricular size, patient-reported outcomes, or sensitivity outcomes, and will be discussed in this publication[7,23-62]. Two publications by the same author were nearly identical and will be discussed as one[27,28]. Furthermore, two publications on the same cohort were also combined[49,50].

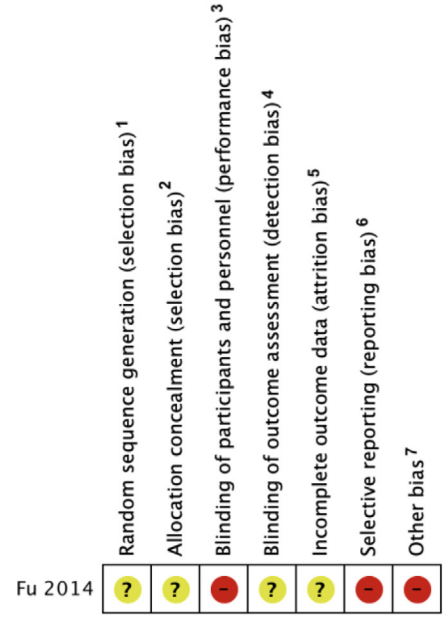
Study characteristics are summarized in Table 1. Twenty-one studies were case series [25,26,29,30,32,33,35,37-39,41,42,44,49,51,54,57,58,60-62], nine were cross-sectional studies[7,23,27,34,36,40,43,45,55], five were prospective cohort studies[24,31,52,56,59], and three were retrospective cohort studies[46,47,53]. One publication was a randomized and controlled study[48]. Thirty publications reported on ACC frameworks [7,23,24,26,27,29-41,44-46,48,49,53-55,58-60,62], seven on PPE frameworks[25,42,43,51,52,56,61], and two on both materials[47,57]. For reconstructions using ACC frameworks, a 'Tanzer', 'Brent', or similar method was used in 18% ($n = 1429/7919$). The 'Nagata' method, or similar two-staged procedure using a fascial flap in the elevation stage was used in 12% of cases ($n = 957/7919$). Tissue expansion was used in three-staged reconstruction in 51% of cases ($n = 4066/7919$), though one study ($n = 3332$) accounted for most of these cases[32]. Allogenic elevation blocks were used instead of banked ACC in two-staged reconstructions in 9% of reconstructions ($n = 679/7919$). Other methods were used in 7% of reconstructions ($n = 572/7919$), and the surgical methods were not described in 3% of ACC reconstructions ($n = 216/7919$).

PPE frameworks were implanted in single-stage reconstructions using a fascia flap for framework coverage in 31% of cases (52/167). Other methods, including tissue expansion and forgoing fascia coverage, were used in 31% of cases (52/167). Surgical methods for PPE implantation were not described in 38% of cases (63/167).

Long-term outcomes were available for 48% ($n = 3789/7919$) of ACC reconstructions and for 93% ($n = 155/167$) of PPE reconstructions. The duration of post-surgical follow-up ranged from 1 to 20 years. The number of patients included in total ranged from 10 to 3332, and number of patients with complete follow-up data ranged from 10 to 628.

Risk of bias assessment

The randomized controlled study was associated with an 'unclear' risk of bias in four of the domains and with a 'high' risk of bias in the remaining three[48]. Details on this assessment can be found in Fig. 2. Five observational studies were considered good quality[23,46,47,59,62], 11 were fair



1. Sequence generation not described
2. Concealment of allocation not described
3. Blinding of participants not described. High risk of bias awarded due to improbability of blinding of personnel due to the very different interventions (original/modified)
4. Blinding of outcome assessors not described.
5. Patients lost to follow-up (if any) not described
6. Patient satisfaction questionnaire outcomes selectively reported and incomplete.
7. High risk of bias pertaining to other bias due to follow-up measurements not performed at standardized times (range 1.5-2.9 years), the timing of the patient satisfaction survey not described and due to the data on the modified framework outcomes being described as non-significant in the text but significant in the tables (Asterix for p<0.05).

Fig. 2. Risk of bias assessment summary.

quality [24,31,33,36,39,41,45,53,55,58,60], and 23 were considered poor quality[7,25-27,29,30,32,34,35,37,38,40,42-44,49-52,54,56,57,61]. Details on the NOS quality assessment scores are presented in Tables 2 and 3.

Data synthesis

Due to significant heterogeneity in interventions and outcome assessments, as well as poor assessment and outcome reporting, quantitative data could not be synthesized using meta-analyses. Results were therefore summarized in a qualitative synthesis.

Aesthetic outcomes

Twenty-nine studies reported on aesthetic outcomes (Tables 4 and 5), excluding those that solely reported on size changes (Table 6)[7,25,26,29-32,35,37-39,41-49,51,54-58,60-62]. Aesthetic outcomes were most frequently assessed by the patient only (10 studies)[7,25,35,43,45,46,48,51,55,56]. Other assessors included the clinician, with[31,42] or without the patient[61], as well as one or several independent assessors[44,47,60,62]. The primary assessor was not described in 12 studies[26,29,30,32,37-39,41,50,54,57,58].

Sixteen studies assessed multiple aesthetic parameters[31,32,37-39,41,42,44,45,47,49,55,58,60-62], and 13 scored one or multiple parameters on a numerical scale[26,31,37,41-45,47,54,55,60,62]. The most frequently reported parameters overall were auricular size, auricular projection, structural definition or contour, and the location or position of the reconstructed auricle (Figure 3). Furthermore, 11 studies reported overall patient satisfaction[7,25,35,45,46,48,49,51,55,56,58].

For ACC reconstructions, five studies summarized the results from excellent or very good to poor[26,31,37,54,60] and five summarized results numerically[41,44,47,55,62] (Table 4). Results from four studies were completely reported and could be summarized ($n = 909$)[26,31,37,60]. Very good or excellent results were reported in 40% of cases, good in 26%, fair in 28%, and poor in 5% of cases. The highest proportion (64%) of excellent results was reported after two-stage reconstruction, including tissue expansion ($n = 279/438$)[37]. Furthermore, all numerically reported aesthetic results were above average (i.e. higher than the mid-point of the scale used; $n = 745$) [41,47,55,62], with the exception of patients who

Table 3. NOS results for cross-sectional studies.

First author and year of publication	Selection				Comparability	Outcome		Summary
	Representativeness of sample[1]	Sample size[2]	Non-respondents[3]	Ascertainment of exposure[4]	Comparability[5]	Assessment of outcome[6]	Adequacy of follow-up[7]	
Tanzer 1978	☆	☆	☆	☆	☆☆	☆	☆	**Good**
Brent 1992	☆	☆	Comparability not described	☆	No control	Self-report	☆	**Poor**
Brent 1999 and 2002	☆	☆	Comparability not described	☆	No control	Self-report	☆	**Poor**
Steffen 2008	☆	Not justified	☆	☆	☆☆	☆	☆	**Fair**
Öberg 2008	No description	Not justified	Not applicable	Written self-report	☆☆	☆	☆	**Poor**
Öberg 2011	☆	Not justified	Not applicable	Written self-report	☆☆	☆	☆	**Poor**
Braun 2013	☆	Not justified	☆	☆	No control	☆	☆	**Poor**
Kristiansen 2013	☆	Not justified	Comparability not described	☆	☆	☆	☆	**Fair**
Akter 2015	☆	☆	Comparability not described	☆	☆	☆	☆	**Fair**

[1] Studies received a star if all or nearly all eligible subjects were included (Tanzer 1978, Brent 1992, Brent 1999/2002, Akter 2015) or in case of consecutive or other non-random sampling.

[2] Four studies received a star as (almost) all eligible patients were included.

[3] A star was awarded if non-respondent characteristics were described (Steffen 2008) or if all subjects responded (Tanzer 1987, Braun 2013). This criterion was not applicable in two studies that conducted a cross-sectional assessment of patients (Öberg 2008 and Öberg 2011).

[4] Studies received a star for conducting a structured interview or sending out a questionnaire.

[5] Five studies received a star for using the contralateral ear as a control group. A second star was awarded for controlling for changes from baseline (Tanzer 1978), controlling for gender (Steffen 2008); for including a healthy control group (Öberg 2008); or controlling for accuracy of the sensitivity filaments used (Öberg 2011).

[6] Studies received a star for using clearly described or validated tools (non-independent assessment).

[7] All studies received stars for follow-up length.

Table 4. Aesthetic assessment outcomes, ACC frameworks.

First author, year of publication	Surgical details[a]	No. of cases	Assessor	Tool used/parameters reported	Results
Brent 1992	3-4, NF	273	Patient	Satisfaction	100% with severe defect, 83-98% with moderate defect, 98% with mild defect
Firmin 1998	Brent/Nagata	300	-	Summarized as very good, good, fair, or poor (no description of parameters)	Very good, $n = 60$ (20%); good, $n = 141$ (47%); fair, $n = 69$ (23%); poor, $n = 30$ (10%)
Park 2002	1-2, TPFF	13	-	Location	Downward migration (5 mm) $n = 3$
Cho 2006	2, MFF	37	-	Contour	Contour acceptable, $n = 33$
Dashan 2008	3, TE, MFF	118	Doctor/ patient	(1) Location, size; (2) symmetry of projection; (3) appearance of helix, antihelix, triangular fossa, earlobe, concha, and tragus; (4) convolution, thickness, and colour match; (5) stability and endurance. Each scored on 10-point scale. Summarized as excellent (no group <8), good (≥ 1 marked 7), fair (≥ 1 marked 6), or poor (≥ 1 marked ≤ 5).	Excellent, $n = 21$ (18%); good, $n = 75$ (64%); fair, $n = 16$ (14%); poor, $n = 6$ (5.1%)
Jiang 2008	3, TE RAF	138	-	colour, structures, and angle	colour redder, $n = 4$ (2.9%); structures vivid, $n = 138$ (100%); angle similar (nq), $n = 136$ (99%)
Pan 2008	3, TE, subcutaneous fascia flap	368	Patient	Satisfaction	Most patients satisfied with results
Wang 2008	2, TE, RAF	438	-	Location, size, projection, convolution, thickness, and colour match. Summarized as excellent, good, fair, or poor (no further description).	Excellent, $n = 279$ (64%); good, $n = 0$; fair, $n = 156$ (36%); poor, $n = 3$ (3%)
Jiang 2011	2, RAF	70	-	3D configuration and cranioauricular angle	Configuration/angle similar to contralateral, $n = 66$ (94%)
Kobayashi 2011	2, TPFF	28	-	Contour and cephaloauricular angle	Contour: all acceptable; angle 32.2° (reconstructed) vs. 35°
Park 2012	1, NF/MFF/ TPFF/ Grotting	19[b]	-	(1) skin colour, (2) coverage, (3) ear size, (4) bilaterally balanced projection.	colour: 3 (nf); 2.8 (1-3) (mff); 1.7 (0-3) (tpff); 3 (grotting) snr coverage: 3 (nf); 2.8 (1-3) (mff); 1.9 (0-3) (tpff); 3 (grotting) snr ear size: 2.7 (2-3) (nf); 3 (mff); 2.9 (2-3) (tpff); 3 (grotting) snr projection: 3 (nf); 2.9 (2-3) (mff); 3 (tpff); 3 (grotting) snr mean sum: 11.6 (11-12) (nf); 11.4 (7-12) (mff); 9.4 (6-12) (tpff); 12 (grotting) snr

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Table 4. (*continued*)

First author, year of publication	Surgical details[a]	No. of cases	Assessor	Tool used/parameters reported	Results
Kim 2013	Nagata ± conchal bowl	20[c]	Ind.	(1) stability of crus helicis, (2) conchal definition, (3) smoothness of helical curve. Each scored on 3-point scale. Points summed (max score 9).	Mean sum: 8 (7–9) with conchal bowl; 3 (2–5) without conchal bowl SNR
Kristiansen 2013	3, NF	59	Patient	Satisfaction with appearance, shape, similarity, and size. Each scored on 4-point Likert scale.	73% satisfied with appearance, 77% satisfied with shape; 39% satisfied with similarity; 88% satisfied with size
Xu 2013	2, RAF	126	Patient	Satisfaction	Satisfaction dependant on smooth skin contour and good definition.
Constantine 2014	3–4, TPFF	17	2 ind/ blinded	(1) Protrusion, (2) definition, (3) shape, (4) size, (5) location, (6) colour match. Each scored on 5-point scale; summarized as means.	Protrusion: 3.9; definition: 3.1; shape: 3.2; size: 3.9; location: 4.3; colour: 4.2. All NS compared with PPE.
Yotsuyanagi 2014ab	2, TPFF	137[d]	-/ patient	Bulkiness, contour, position, size, sulcus, patient satisfaction.	Bulkiness: $n = 4$; unclear contour: $n = 3$; asymmetrical position: $n = 4/121$; asymmetrical size: $n = 2/121$; shallow sulcus, $n = 8/121$; asymmetry of earlobe: $n = 4/121$; almost all patients satisfied.
Xu 2014	-	216	Patient	Satisfaction	Great majority satisfied
Sharma 2015	3, postauricular galeal flap	55	-	Crus of helix, upper, middle, and lower third of helix, superior and inferior crus, middle part of anti-helix, anti-tragus, tragus, lobule, scaphoid fossa, triangular fossa, cymba concha, cavum concha. Each eligible for 1 point. Summarized as poor (1–5 points), average (6–8 points), good (9–11 points), or excellent (12–13 points).	First group ($n = 44$): poor to average, $n = 28$ Second group ($n = 11$): excellent, $n = 8$
Akter 2017	Similar to Nagata	69[e]	Patient	Questionnaire 1: Height, width, prominence, folds of ear, colour match, fitting with face, realness, helix, anti-helix, concha, tragus/anti-tragus, lobule, scar appearance, scar colour. Each scored on 5-point scale. Questionnaire 2: Helical rim, scaphoid fossa, conchal bowl, anti-tragus, triangular fossa, anti-helix, tragus, incisura, lobe, size, projection, position, rotation, skin coverage, scalp scars, similarity. Each scored on 5-point Likert scale. Satisfied: score of 4 or 5.	Questionnaire 1 ($n = 50$): median score colour match 5, all others 4. Questionnaire 2: median score for helix, scaphoid, concha, anti-tragus, triangular fossa, anti-helix, incisura all 4, tragus 4–5, lobe 5; size: 5, projection: 4, position: 5, rotation: 4–5, skin: 4–5, scars: 4 Satisfied: overall, $n = 58$ (84%); with similarity, $n = 53$ (77%); with size, $n = 57$ (83%); with shape, $n = 52$ (75%);

(*continued on next page*)

Table 4. (*continued*)

First author, year of publication	Surgical details[a]	No. of cases	Assessor	Tool used/parameters reported	Results
Kim 2017	2, TPFF, PPE block	51	-	Projection	Stable projection (NQ) $n = 51$ (100%)
Han 2018	2, superficial TPFF	22	-/ patient	Auricolocephalic angle, contour, satisfaction.	Angle 29.1° (reconstructed) vs. 30.8°; contour well preserved; all satisfied.
Ladani 2019	2, MFF/ TPFF	53	Doctor/ Patient/ Ind.	(1) Location, size; (2) symmetry of projection; (3) appearance of helix, antihelix, triangular fossa, earlobe, concha, and tragus; (4) convolution, thickness and colour match; (5) stability and endurance (5). Each scored on 10-point scale. Summarized as excellent (no group <8), good (≥ 1 marked 7), fair (≥ 1 marked 6), or poor (≥ 1 marked ≤ 5).	Excellent, $n = 8$ (15%); good, $n = 23$ (43%); fair, $n = 17$ (32%); poor, $n = 5$ (9%). Significant loss of projection (NQ) $n = 16$ (34%)
Li 2020	2, RAF, composite block	628[f]	3 ind/ blinded	(1) colour match, (2) flatness, (3) cranioauricular sulcus depth, (4) angle symmetry. each scored on 5-point scale. summed for global assessment (max: 20).	colour: 4.0 ± 0.7 (groin); 4.3 ± 0.5 (postauricular/groin); 4.2 ± 4.2 (scalp) ns. flatness: 4.9 ± 0.3 (groin); 4.9 ± 0.9 (postauricular/groin); 4.0 ± 1.0 (scalp)*** sulcus: 4.5 ± 0.8 (groin); 4.5 ± 0.8 (postauricular/groin); 3.7 ± 0.9 (scalp)*** angle: 4.6 ± 0.6 (groin); 4.6 ± 0.7 (postauricular/groin); 4.11 ± 0.9 (scalp)* global: 18.1 ± 1.6 (groin); 18.3 ± 2.1 (postauricular/groin); 16.0 ± 2.8 (scalp)***

ACC: autologous costal cartilage; NF: no fascia flap; TPFF: temporoparietal fascia flap; MFF: mastoid fascia flap; RAF: retroauricular fascia flap; NQ: not quantitatively reported; Ind: independent.

Values presented as mean (range), mean $\pm$ standard deviation, or no. of cases (proportions), unless otherwise specified. Proportions are given if the total number of cases is ≥ 50. Statistical significance: p*<0.05; **$p<0.01$; ***$p<0.001$; NS: not significant; SNR: significance not reported.

[a] Surgical details include number of stages, tissue expander use, fascia flap use.

[b] Reconstruction was performed without fascia flap in 3 cases, with a MFF in 8 cases, with a TPFF in 7 cases and a Grotting flap in 1 case.

[c] 12 patients were reconstructed with the conchal bowl element and 8 without.

[d] Sulcus and asymmetry scores reported per patient ($n = 121$), not per case ($n = 137$).

[e] Results for projection, skin coverage, and scalp scars were missing in 3 cases. Results for rotation were missing for 2 cases.

[f] Skin graft donor sites were the groin ($n = 202$), the postauricular area and groin ($n = 195$), or from the scalp ($n = 231$).

Table 5. Aesthetic assessment outcomes, PPE frameworks.

First author, year of publication	Surgical details[a]	No. of cases	Assessor	Tool used/parameters reported	Results
Şengezer 1996	2, NF	10	Patient	Patient satisfaction	High rate of patient satisfaction.
Zhang 2012	3, TE, RAF/NF	27[b]	Doctor/ Patient	Position, size, external shape, sulcus, and incisional scar. Summarized as excellent, good, fair, or poor (no further description).	Excellent, $n = 14$ (doctor), $n = 10$ (patient); good, $n = 9$ vs. $n = 12$; fair, $n = 3$ both; poor, $n = 1$ vs. $n = 2$
Braun 2013	1, TPFF	15	Patient	Scar aesthetics scored on 6-point scale (1 best, 6 worst).	Scar aesthetics: 3 (1-6)
Constantine 2014	1, TPFF	17	2 ind/ blinded	(1) Protrusion, (2) definition, (3) shape, (4) size, (5) location, (6) colour match. Each scored on 5-point scale.	Protrusion: 3.6; definition: 3.5; shape: 3.7; size 4.3; location: 4.2; colour: 3.9. All NS compared with ACC.
Chen 2015	1, superficial TPFF	18	Patient	Patient satisfaction	All satisfied
Johns 2017	-	28	Patient	Patient satisfaction	All patients had a high level of satisfaction.
Zhang 2019	1, TPFF/NF/ 2, RAF	17[c]	Doctor	Symmetry of position, size, texture, colour match, and structural details. Summarized as satisfying or not (no further description).	1, TPFF: 2/2 satisfying 1, NF: 2/2 satisfying; 2, RAF: 9/13 satisfying

PPE: porous polyethylene; NF: no fascia; TE: tissue expander; RAF: retroauricular fascia; TPFF: temporoparietal fascia; NS not significant; ACC: auricular costal cartilage; Ind: independent.
Values presented as mean (range) or no. of cases (proportions). Proportions are given if the total number of cases is ≥50.
[a] Surgical details include number of stages, tissue expander use, fascia flap use.
[b] RAF used in 2 cases and no fascial coverage in 25 cases.
[c] Single-stage and TPFF in 2 cases, single-stage and no fascia flap in 2 cases, and two-stage procedure with RAF in 13 cases.

underwent Nagata-style reconstruction without a conchal bowl element ($n = 8$)[44].

Overall patient satisfaction with the aesthetic outcome, reported by seven studies on ACC reconstructions, was high[7,35,45,46,48,49,55,58]. Two studies reported satisfaction outcomes on multiple parameters[45,55] ($n = 128$). Summarizing the results: 79% of patients were satisfied with the appearance of the auricle; 76% were satisfied with the shape; 85% were satisfied with the size, and 60% with the similarity of the reconstructed auricle. Furthermore, median scores for projection, position, rotation, skin, scars, and auricular structures ranged from 4 to 5 (on a 5-point Likert scale) after Nagata-style reconstruction in the study by Akter et al.[55].

Furthermore, a study by Li et al. compared skin grafts for two-staged reconstruction using a composite block for auricular elevation ($n = 628$)[62], and a study by Park compared fascia flaps used during single-stage reconstruction[41]. Scores for flatness, sulcus depth, and the cranioauricular angle were significantly better for grafts harvested from the groin ($n = 202$) or postauricular area and groin ($n = 195$), compared with skin grafts harvested from the scalp ($n = 231$)[62]. In the study by Park, scores for colour match and skin coverage were highest for auricles reconstructed without additional fascia flap coverage ($n = 3$), with a mastoid fascia flap ($n = 8$) or with a Grotting flap ($n = 1$), compared with those reconstructed with a temporoparietal fascia flap ($n = 7$), though the statistical significance was not described[41].

Two studies on PPE implantation used numerical scales for the aesthetic assessment of the auricle[42,47]. One of these studies, on three-stage PPE implantation after tissue expansion ($n = 27$), summarized results from excellent to poor, and most cases were deemed excellent or good[42] (Table 5). This study also separated the doctor and patient's assessments: clinicians awarded an 'excellent' grade more frequently, while patients scored ears as 'good' and 'poor' more frequently. In the other study, where reconstruction was performed in a single stage using temporoparietal fascia flap coverage, aesthetic results were above average for all parameters assessed ($n = 17$)[47]. This study also compared ACC and PPE reconstruction scores, and found no statistically significant difference between the two. However, definition ($p = 0.05$), shape ($p = 0.08$), size ($p = 0.05$), and colour ($p = 0.05$) scores approached significance, the former three favoring the PPE group and the latter the ACC group. Overall patient satisfaction, reported by three studies, was high after PPE reconstruction[25,51,56].

Auricular size

Auricular size outcomes were reported by eight studies on ACC reconstructions (table 6)[7,23,24,27,33,46,48,53]. Four studies

Table 6. Size outcomes.

First author, year of publication	Surgical details[a]	No. of cases	Assessor	Follow-up range (years)	Measurement method	Results
Tanzer 1978	3/4/6, NF	37	Patient/ Doctor	6-19	Initial: direct measurement after reconstruction. Follow-up: from tracings (doctor) and direct measurement (patient-reported). Measurements averaged.	Increase in size: $n = 31$ (reconstructed) vs. $n = 33$. Height change: +3.6 vs. +4.4 (NS)
Thomson 1989	6, NF	29	Doctor	≥2.5	Initial: original lead plates Follow-up: measured from tracing	Increase in size: $n = 25$ (reconstructed) vs. 26. Decrease in size: $n = 4$ (reconstructed) vs. 3. Perimeter change: reconstructed greater in 86% of patients*** (compared with lead plates) and 90% (normal)***. SNR between groups.
Brent 1992/ 1999/2002[b]	3-4, NF	508	Patient	1-18	Unclear if measured.	Decrease in size: $n = 0$
Kizhner 2008	Nagata	27		0.8-7.3	Initial: original template Follow-up: direct measurement	Mean change in height: -1.8* Mean change in width: +1.3*
Xu 2013	2, RAF	126	-	1.9-3.1	All using standard calliper	Height change (reconstructed/normal): children: -0.5 NS/+0.8*; adolescents: -0.4 NS/+ 0.1 NS; adults: -0.2 NS/+0.4 NS. SNR between reconstructed and normal measurements. Width change(reconstructed/normal): children: +1.2*/+0.8*; adolescents: +1.4*/+0.5 NS; adults: +0.1 NS/+0.0 NS. SNR between reconstructed and normal measurements.
Xu 2014	-	216	-	1.5-2.9	All using standard calliper	Height change: group A, original method +1.6*; group A, modified method +0.1 NS; group B, original +2.5*; group B, modified +0.1 NS Width change: group A, original +1.5*; group A, modified +0.3; group B, original +0.6*; group B, modified +0.3
Roos 2015	3, NF	22	Blinded	2.1-10.6	All from photographs	Height change: 6.2 (reconstructed) vs. 5.3 NS

NF: no fascia; RAF: retroauricular fascia.
Results reported in millimeters (if size change reported) or in number of patients with a size change (proportion). Statistical significance: *$p<0.05$; ** $p<0.01$; **$p<0.001$; NS: not significant. SNR: significance not reported.
[a] Surgical details include number of stages, tissue expander use, fascia flap use.
[b] Results combined as they pertain to the same cohort.

reported increased height of both reconstructed and normal ears[23,24,48,53], two studies reported decreased height of the reconstructed auricle after follow-up[33,48], and two studies reported no change in size[7,27]. Differences in auricular height change between normal and reconstructed ears were largely not statistically significant[23,46,53]. However, significant differences were found in one study, in the subgroup consisting solely of children, where reconstructed ears shrunk by 0.5 mm and normal ears grew by 0.8 mm during follow-up[46]. Furthermore, in another study, ears reconstructed without the authors' framework modifications were significantly larger at follow-up compared with the normal ears[48].

Two studies reported change in auricular width, and in both, wider ears were seen after follow-up[46,48]. Reconstructed ears were significantly wider compared with

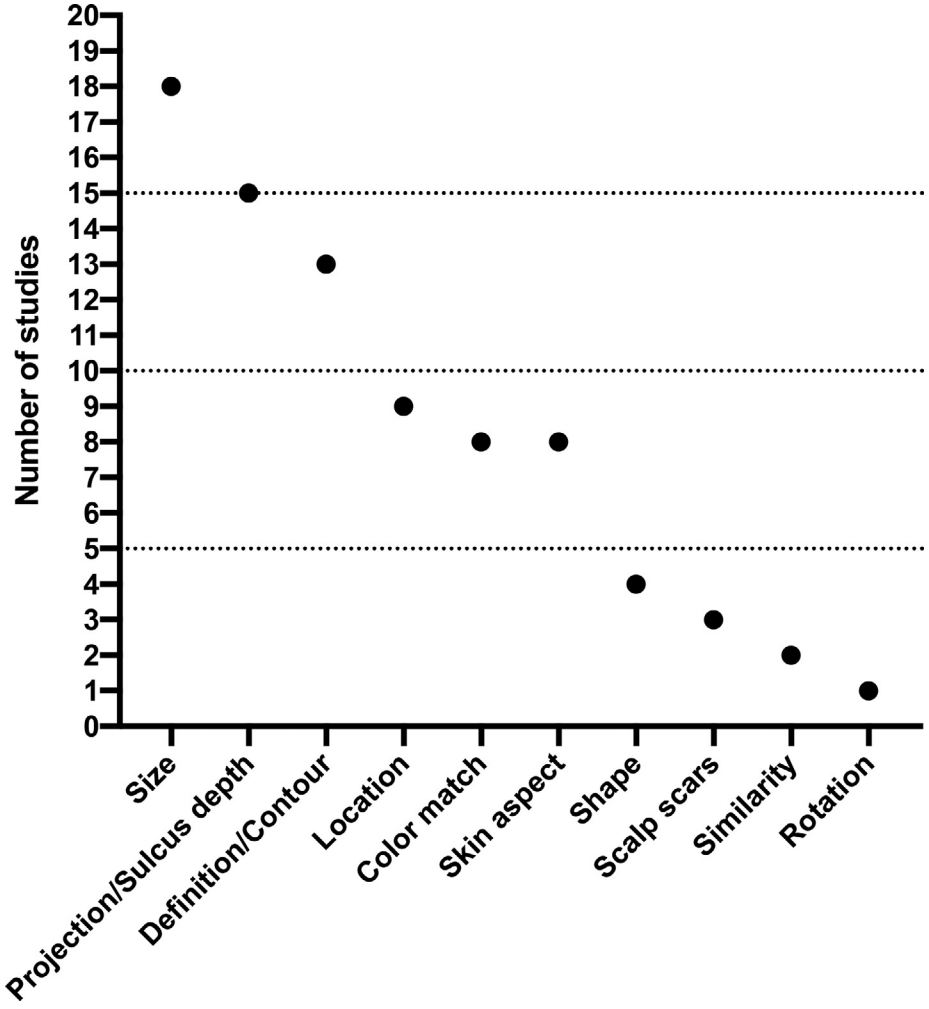

Fig. 3. The most frequently reported aesthetic assessment parameters.

normal ears in the adolescent subgroup in one study and in patients reconstructed without the authors' framework modifications in the other[48].

Auricular sensitivity

Four studies, all on ACC reconstructions, reported on auricular sensitivity [34,37,40,59] (Table 7). Protective (tactile) sensitivity, assessed in three studies, was intact in most or all ears[37,40,59]. However, a study by Denadai et al. noted significant insufficiency in terms of protective and temperature sensitivity of the post-auricular sulcus ($p<0.01$) after Nagata-style reconstruction[59]. Furthermore, in the fourth study, the antihelix and helix of reconstructed ears were significantly less sensitive to heat, while lobules were significantly more sensitive to cold when compared with contralateral ears ($n = 19$)[34].

HRQoL

Six studies reported on quality of life outcomes after ACC reconstruction (Table 8)[7,23,27,36,45,55]. One used a generic HRQoL questionnaire in combination with a microtia-specific self-made questionnaire[36], and five used self-made questionnaires only[7,23,27,45,55]. None collected scores preoperatively. Postoperatively, 82% ($n = 105/128$) of patients reported being able to wear sunglasses[45,55] and 89% ($n = 106/119$) felt that the reconstructed ear was a part of their body[36,45]. The earlier studies by Tanzer and Brent also reported good rates of emotional or psychological benefit[7,23,27]. This effect was less pronounced in the later studies: no significant difference was found in patients' self-concept in the study by Steffen et al.[36], and Kristiansen et al. reported an unchanged mood in 75% of patients[45]. In the study by Akter et al., respectively, 13 and 14% of patients reported hiding their ears or feeling self-conscious postoperatively[55].

Three studies on PPE reconstructions reported on quality of life outcomes, two using generic and self-made questionnaires[43,52], and one using a self-made questionnaire only[56] (Table 9). Two studies collected preoperative scores in addition to postoperative scores[52,56]. All studies reported positive outcomes: both studies by Johns et al. found a significant decrease in negative emotions, as well as a decrease in social awareness or hiding ears[52,56]. Furthermore, Braun et al. reported low scores for scar-related limitations in daily life and shame[43].

Table 7. Auricular sensitivity results.

First author, year of publication	Surgical details[a]	No. of cases	Follow-up range (years)	Tool	Results
Wang 2008	2, TE, RAF	438	1-2	-	Sensation to light touch, pinprick, and temperature and static two-point discrimination present.
Öberg 2008	3, NF	19	≥2	Thermal sensitivity (SENSELab MSA Thermotest)	Heat: higher thresholds in antihelix and helix*, not lobe. Cold: lower threshold in lobe*. Antihelix and helix NS
Öberg 2011	3, NF	39	0.5-5	Protective sensitivity: SWMT of helix, antihelix, and lobule	Good results (normal—diminished protective): $n = 32$ (helix), $n = 38$ (antihelix), $n = 39$ (lobule). No difference short (0.6-1.7 years) and long (1.7-5 years) follow-up groups.
Denadai 2019	Nagata	38	1	SWMT (protective sensitivity), temperature discrimination	8/11 of auricular points no difference with contralateral NS: in upper, middle, and lower points of post-auricular sulcus significantly more insufficient protective and temperature sensitivity**.

TE: tissue expander; RAF: retroauricular fascia flap; NF: no fascia; SWMT: Semmes-Weinstein Monofilament Test.
Statistical significance: *$p<0.05$; ** $p<0.01$; **$p<0.001$; NS: not statistically significant.
[a] Surgical details include number of stages, tissue expander use, fascia flap use.

Overall quality of evidence

The overall quality of evidence was very low for all outcomes (Table 10). Publication bias could not formally be assessed due to the lack of quantitative synthesis.

Brief summary of complication results

The results on long-term complciations are described and discussed in detail our previous manuscript.[16] Briefly summarized, twenty-nine publications reported on complications during longterm follow-up[7,11,23,25-28,30-32, 35, 37-39,42,47,49,50,57,58,60,61,71-77] (editor: please not references[71-77] are the new references added by me, the corresponding number might change). Overall long-term complication rates were not reported. The incidence of individual complications during longterm follow-up was less than 10% after ACC reconstruction and less than 15% in PPE reconstruction. Framework resorption and wire exposure were reported even after an extended follow-up of more than five years after ACC reconstruction, while reports on the extended long-term results of PPE reconstruction were limited. Data synthesis was limited due to heterogeneity and poor study quality. Based on the most frequently reported complications, future studies should at least include framework extrusion or exposure, graft loss, framework resorption, wire exposure and scalp/auricular scar complications, largely in agreement with the current UK Care Standards for the Management of Patients with Microtia and Atresia.[63] Furthermore, complications should be stratified according to the subtype of microtia treated, and a surgical follow-up of at least five years is recommended.

Discussion

This systematic review aimed to summarize long-term outcomes after microtia reconstruction. Following a systematic and thorough search of the literature, 41 publications published between 1978 and 2020, were included in our analysis of aesthetic, sensitivity, and HRQoL outcomes. We found large variability in the reporting of these outcomes, as well as in postsurgical follow-up duration.

Aesthetic outcomes, assessed in 29 studies, were the most frequently reported outcome, next to complications, reported earlier[16]. The majority of both ACC and PPE reconstructions were graded good or excellent or received high marks on a numerical scale. Similarly, patient-reported satisfaction with the aesthetic outcome was generally high. However, the methods implemented to assess aesthetics varied considerably, auricles were graded by independent assessors only in just three studies, and requirements for point allocation on numerical scales were not clearly defined in any study. Furthermore, patient satisfaction was most frequently reported as a binary overall outcome, and just three studies reported satisfaction with multiple aesthetic parameters.

Neither standardized nor validated tool exists to assess the external ear. However, the International Society for Auricular Reconstruction (ISAR) agreed to the international use of the measurement tool developed following the Scientific Advisory Committee of Medical Outcomes Trust[55], previously published in the UK Care Standards for the Management of Microtia and Atresia[63], and used by Akter et al.[55], during the ISAR congress in Beijing in 2017. This questionnaire assesses patients' satisfaction with several aesthetic parameters including overall appearance, size, shape, projection, and the subunits of the ear on a 5-point visual analogue scale (VAS).

Table 8. HRQoL outcomes, ACC frameworks.

First author, year of publication	Surgical details[a]	No. of cases	Tool used	Results
Tanzer 1978	3,4, or 6, NF	42	Self-made questionnaire: effects on daily social contacts (post-operation)	None, $n = 23$; minor, $n = 15$; moderate, $n = 4$; severe, $n = 0$
Brent 1992	3-4, NF	273	Self-made questionnaire: psychological relief, emotional benefit	Relief: 100% (severe impact preoperatively); 9% unchanged (moderate impact preoperatively). Emotional benefit: 40-68%; no concern exposing ear in 86-95%.
Brent 1999/2002	3-4, NF	508[b]	Self-made questionnaire	The greater the emotional impact of the deformity, the greater the relief by its repair.
Steffen 2008	Nagata	60	Frankfurter Selbstkonzeptskalen (FSKN: self-concept scale) and self-made questionnaire (compatibility)	FSKN: +12 points overall NS. No change in self-esteem or self-rated performance abilities. Compatibility (with body): 87%
Kristiansen 2013	3, NF	59	Self-made questionnaire (functional, psychosocial)	Functional: 76% can wear sunglasses, 29% difficulty cleaning ear, Psychosocial: 86% would opt for ACC again, 91% feels compatibility, 24% happier, 75% unchanged mood, 2% sadder, 14% avoids certain activities, 10% afraid of ear falling off
Akter 2017	Similar to Nagata	69	Self-made (functional, psychosocial)	Functional: can wear glasses, $n = 60$ (87%) Psychosocial: hides ear with hat, $n = 9$ (13%); anxious about seeing hairdresser, $n = 11$ (16%); hides ear in photos, $n = 9$ (13%); avoids mirrors, $n = 5$ (7%); self-conscious, $n = 10$ (14%). Overall: would opt for ACC again, $n = 61$ (88%); would prefer prosthesis, $n = 2$ (3%); would prefer to do nothing, $n = 6$ (9%)

HRQoL: health-related quality of life; ACC: autologous costal cartilage; NF: no fascia.
Values presented as mean (range) or no. of cases (proportions). Proportions are given if the total number of cases is $\geq$50.
[a] Surgical details include number of stages, tissue expander use, fascia flap use.
[b] Includes cohort published by Brent in 1992.

Furthermore, this questionnaire covers the most frequently reported aesthetic parameters, identified through our systematic review of the literature. Due to the international consensus, supported by the results of this review, we encourage future studies to integrate this questionnaire in their outcome reporting.

Furthermore, in order to compare results and perspectives internationally, studies should provide descriptions and photographic examples of illustrative cases, ranging from very dissatisfactory results (1) to very satisfactory results (5). However, the comparability of results may nevertheless be limited by the subjective nature of assessments. Interestingly, a study previously found that a panel of 20 plastic surgeons were reliably able to rank auricles on a VAS, where the highest scoring auricle corresponded to conventionally used aesthetic proportions[64]. Large-scale implementation of the aesthetic outcome assessment scale, as well as clear illustrative cases may clarify whether these results can be extrapolated internationally.

No conclusions could be drawn on the difference in patient- and clinical-based assessments, as just one study published data on the surgeon and patient's assessments separately. Exploring possibly differing perceptions is vital in optimizing surgical counselling, and studies investigating these differences are warranted.

Sensitivity of the anterior surface of the auricle was satisfactory in all studies as little as 1 year after the last surgery. However, sensitivity of the retroauricular sulcus was insufficient compared with the contralateral side after Nagata-style reconstruction in the only study assessing the retroauricular sulcus[59]. These findings are in line with previous reporting on the reinnervation of skin flaps and grafts of the face, where sensation to light touch was present 2 years after Mohs surgery in 75% of patients who had undergone flap reconstruction, but only in 29% of patients who had undergone skin graft reconstruction[65]. None of the included studies evaluated sensitivity after PPE reconstruction. Evaluating the sensitivity of the reconstructed ear after alloplastic reconstruction is especially important as diminished sensitivity could expose patients to unnoticed wounds, which in turn could lead to framework exposure and subsequent infection necessitating implant removal[13].

Table 9. HRQoL outcomes, PPE frameworks.

First author, year of publication	Surgical details[a]	No. of cases	Tool used	Results
Braun 2013	1, TPFF	15	Glasgow (Children's) Benefit Inventory [GCBI and GBI], >0 points indicates benefit. Self-made questionnaire: 6-point scale for limitations in daily life, attraction of public attention, and feeling shame (of scalp scar)	Mean GBI score: 24 (range -6 to +56); mean GCBI: 31 (2-85) SNR Limitations in daily life: mean 1.3, attraction of public attention 13%; feeling shame mean 1.5
Johns 2015	-	23	behavioural assessment system for children (basc-2) and self-made questionnaire/ interview (negative emotions and social awareness)	BASC-2: decreased anxiety*, decreased depression**, increased social skills**. Self-made: decreased mean negative emotions ***, decrease social awareness***.
Johns 2017	-	28	Self-made questionnaire/interview (teasing, emotional, and psychosocial outcomes)	Increase in happiness***, decrease in teasing***, decrease in sadness**, decrease in worry**, decrease in feeling mad*, decrease in shyness**, decrease in hiding ears*. Greater change ages 6-10.

HRQoL: health-related quality of life; PPE: porous polyethylene; TPFF: temporoparietal fascia flap; Significance: * $p<0.05$; ** $p<0.01$; *** $p<0.001$; NS: not significant; SNR: significance not reported.
[a] Surgical details include number of stages, tissue expander use, fascia flap use.

Table 10. GRADE summary of findings.

Outcome	*No. of studies*	*No. of RCTs*	*Limitations*	*Inconsistency*	*Indirectness*	*Imprecision*	*Publication bias*	*Quality of Evidence*
Aesthetics ACC	23	0	Very serious [a]	Serious [c]	Very serious [d]	No serious imprecision	Unclear [e]	Very low
Aesthetics PPE	7	0	Very serious [a]	Serious [c]	Very serious [d]	Serious[f]	Unclear [e]	Very low
Aesthetics ACC vs. PPE	1	0	No serious limitations	NA	NA	Serious [f]	Unclear [e]	Very Low
Change in height ACC	5	1	Serious [b]	Serious [c]	Very serious [d]	No serious imprecision	Unclear [e]	Very low
Auricular sensitivity ACC	4	0	Very serious [a]	Serious [c]	Very serious [d]	No serious imprecision	Unclear [e]	Very low
Psychosocial outcomes ACC	6	0	Serious [b]	Serious [c]	Very serious [d]	No serious imprecision	Unclear [e]	Very low
Psychosocial outcomes PPE	3	0	Very serious [a]	No serious inconsistency	Very serious [d]	Serious [f]	Unclear [e]	Very low

ACC: autologous costal cartilage; PPE: porous polyethylene; RCT: randomized-controlled trial; NA: not applicable (due to just one study in the category).
[a] At least half of studies were graded poor quality or had a high risk of domain associated with ≥1 domain.
[b] Less than half of studies poor quality/high risk in at least 1 risk of bias domain.
[c] Considerable variation in results between studies.
[d] Considerable differences in population (or unclear differences due to missing patient characteristics) and considerable differences in interventions (or no description of interventions).
[e] Most or all studies had positive results, or very few studies reported on the outcome. No formal assessment performed.
[f] Small total population size (all <150).

Based on the included studies, reconstructed auricles generally grew at a similar rate to the contralateral ears. Decreased auricular height, reported by two studies, could perhaps have been attributed to a decrease in oedema as initial measurements were taken at the time of reconstruction in one of these studies[46]. In the other study, initial measurements were taken from original templates, not directly from the reconstructed auricles, introducing a greater margin of error[33]. The growth of auricles was most apparent in children[23,46] as well as in studies where most or all patients were followed for more than 5 years[23,53]. Several large anthropometric studies have shown auricles to reach approximately 85% of their mature lengths at 5 years of age[66,67], though others suggest that auricles grow throughout the entire lifetime[68]. Based on these studies, it seems reconstructed auricles follow the growth pattern of the normal auricle. However, studies assessing size in adults with auricles reconstructed as children are warranted to confirm this observation.

Finally, HRQoL outcomes illustrated benefit of both ACC and PPE reconstructions, though scores were moderate for ACC reconstructions and limited by the lack of preoperative measurements. Recently, the EAR-Q, an ear-specific HRQoL and satisfaction questionnaire developed by the authors of the CLEFT-Q and FACE-Q, has been released after field testing [69]. The ISAR congress in 2017 also agreed upon the international use of this patient-reported outcome measure (PROM). To fully assess benefit of reconstructions, surgeons should strive to collect pre- and postoperative scores.

The results of this systematic review are limited by a few factors. First, a proportion of patients potentially eligible for inclusion may have been missed, as we did not receive responses from the majority of contacted authors. These studies frequently reported a range for postsurgical follow-up duration, where an unknown proportion of patients was followed for less than 1 year. Furthermore, we were unable to perform meta-analyses on our data due to interstudy heterogeneity. Evidence levels of all outcomes were downgraded to very low, mainly due to the observational nature of most included studies, low study quality, and considerable variation in study characteristics.

Conclusion

To conclude, microtia reconstruction using ACC or PPE frameworks leads to pleasing results in the majority of cases reported, with high rates of patient satisfaction and good postoperative HRQoL outcomes. Furthermore, auricles reconstructed using ACC grow at a rate similar to the contralateral ear and exhibit largely normal sensitivity during long-term follow-up. Further research on auricular sensitivity after PPE reconstructions is warranted. No conclusions can be drawn on the superiority of either method due to the lack of comparative analyses. These conclusions are also limited by poor evidence quality and the lack of standardized reporting. To improve future reporting and the comparability of results, we propose the following minimum reporting criteria: (1) surgical efficacy measured using the tool provided in the UK Care Standards for the Management of Patients with Microtia and Atresia; (2) complications including framework extrusion or exposure, graft loss, framework resorption, wire exposure and scalp/auricular scar complications; and (3) HRQoL before and after treatment using the EAR-Q PROM. Standardizing international reporting is vital for evidence-based decision-making in this rare patient population, and centralizing patient-reported outcomes is crucial due to the aesthetic and functional nature of microtia reconstruction.

Declaration of Competing Interest

None.

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Ethical approval statement

Ethical approval was not required for this study.

Supplementary materials

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