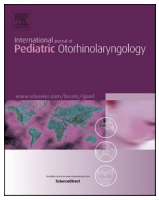
International Journal of Pediatric Otorhinolaryngology 186 (2024) 112095

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl



Comprehensive management and classification of first branchial cleft anomalies: An International Pediatric Otolaryngology Group (IPOG) consensus statement

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https://doi.org/10.1016/j.ijporl.2024.112095
Received 26 July 2024; Received in revised form 1 September 2024; Accepted 6 September 2024
Available online 7 September 2024

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ARTICLE INFO

Keywords:
First branchial cleft cyst
First branchial cleft anomalies
Parotid
Facial nerve
Surgical management first branchial cleft anomalies

ABSTRACT

Objective: First branchial cleft anomalies are rare congenital head and neck lesions. Literature pertaining to classification, work up and surgical treatment of these lesions is limited and, in some instances, contradictory. The goal of this work is to provide refinement of the classification system of these lesions and to provide guidance for clinicians to aid in the comprehensive management of children with first branchial cleft anomalies.
Materials and methods: Delphi method survey of expert opinion under the direction of the International Pediatric Otolaryngology Group (IPOG) was conducted to generate recommendations for the definition and management of first branchial cleft anomalies. The recommendations are the result of expert consensus and critical review of the literature.
Results: Consensus recommendations include evaluation and diagnostic considerations for children with first branchial cleft anomalies as well as recommendations for surgical management. The current Work classification system was reviewed, and modifications were made to it to provide a more cogent categorization of these lesions.
Conclusion: The mission of the International Pediatric Otolaryngology Group (IPOG) is to develop expertise-based recommendations based on review of the literature for the management of pediatric otolaryngologic disorders. These consensus recommendations are aimed at improving care of children presenting with first branchial cleft anomalies. Here we present a revised classification system based on parotid gland involvement, with a focus on avoiding stratification based on germ layer, in addition to guidelines for management.

1. Introduction

Branchial cleft anomalies of all kinds (types I, II, III and IV) are the second most common congenital abnormality found within head and neck, accounting for nearly 20 % of lesions [1]. First branchial cleft anomalies are much less common, comprising 1–18 % of all branchial cleft anomalies [2–4]. Because the overall incidence of first branchial cleft anomalies is low, they are more difficult to study [5]. These anomalies can form sinuses, fistulae or cysts [6]. First branchial cleft anomalies typically present during infancy and childhood, but diagnosis can be at any age [7]. They may present as cervical, parotid, preauricular or cheek swelling/masses and are often misdiagnosed as preauricular pits, benign parotid cysts or infected lymph nodes [5,6,8,9]. Many of these lesions have been noted to have openings to the external auditory canal and, as a result, can present with recurrent otorrhea [9]. Misdiagnosis can lead to incomplete resection with higher recurrence rates [5]. Recurrence of a pre- or post-auricular abscess with a history of incision and drainage should raise suspicion for a first branchial cleft anomaly [7].

The Work classification was introduced in 1972. The Work classification describes two variants of first branchial cleft anomalies based on location and germ layer; each variant is believed to result from duplication of the membranous external auditory canal [2,7,10]. Work type 1 lesions are ectodermal in origin and are typically found in the pre- or post-auricular area, lateral to the facial nerve [7,10,11]. Work type 2 lesions originate from mesoderm and ectoderm, contain both cartilage and epithelium, and form in the region of the angle of mandible [7,11]. Fistulae or sinus tracts, when they occur, are often intimately associated with the external auditory canal and can be associated with the parotid gland and facial nerve [7]. The Work system was proposed in an era before the advent of the subspecialty of pediatric otolaryngology. Despite it being the most commonly used classification system for first branchial cleft anomalies, many otolaryngologists question its clinical applicability and utility in the management of these lesions.

Diagnostic approach for first branchial cleft anomalies varies within the literature. While ultrasound may be used as a screening modality in the work up of first branchial cleft anomalies, cross-sectional imaging with computed tomography (CT) and/or magnetic resonance imaging (MRI) is often necessary for diagnosis and proper management [2,7]. Definitive treatment is complete surgical excision to avoid complications, such as recurrent infections and poor cosmesis [2,3,7]. Revision surgeries can be problematic secondary to scar tissue and fibrosis adherent to branches of the facial nerve [3,12]. To date, literature pertaining to classification, work up and technical considerations for surgical resection of first branchial cleft anomalies remains limited.

The mission of the International Pediatric Otolaryngology Group (IPOG) is to develop expertise-based recommendations for the management of pediatric otolaryngologic disorders [14]. Here we present a revised classification system based on parotid gland involvement, with a focus on avoiding stratification based on germ layer, as well as provide guidance for the comprehensive management of children with first branchial cleft anomalies. These consensus recommendations are based on review of the literature and the cumulative expertise of pediatric otolaryngologists with a focus on treating head and neck pathologies.

2. Materials and Methods

Delphi method survey of expert opinion under the direction of IPOG was conducted to generate recommendations for the management and classification of first branchial cleft anomalies. Phase 1 included an extensive literature review with development of an online survey comprised of 25 questions pertaining to the classification, diagnosis and

surgical management of first branchial cleft anomalies. Emphasis was placed on capturing experts who regularly treat first branchial cleft anomalies at a range of institutions from around the world. Recommendations for this expert panel were solicited from presidents of the major international pediatric otolaryngology societies: American Society of Pediatric Otolaryngology (ASPO), The European Society of Pediatric Otorhinolaryngology (ESPO) and Asian Pediatric Otolaryngology Group (APOG) and Pediatric Ear, Nose & Throat Society in Africa (PentAfrica). A total of 47 participants were selected.

Phase 2 included sending the first round of the Delphi method online survey to the designated experts, analyzing the survey results and preparing the second online survey. Phase 3 included sending the modified second round of the Delphi method online survey to the designated experts, analyzing the survey results and composing management and classification recommendations based on the responses that reached consensus among the expert panel. See Fig. 1.

3. Results

The following consensus recommendations are the result of a Delphi method survey of expert opinion under the direction of IPOG. The recommendations are the result of both critical review of the literature and expert consensus among pediatric otolaryngologists with expertise in treating first branchial cleft anomalies. We achieved a 100 % completion rate for both first and second round surveys. Consensus was achieved when agreement was reached among at least 75 % of responders [13]. Consensus was reached for 68 % of questions administered to participants. The consensus recommendations for diagnosis and management of first branchial cleft anomalies are broken down into the following categories:

Classification (Table 1).

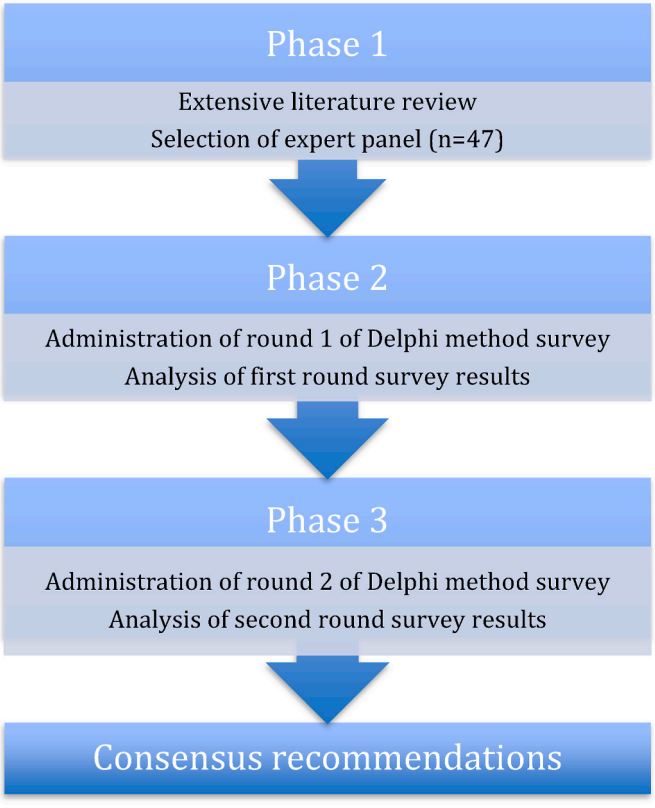

Fig. 1. Phases of Delphi method survey of expert opinion leading to consensus recommendations.

- First branchial cleft anomalies represent reduplication of the ear canal.
- To date, the Work classification is the most widely accepted system to categorize first branchial cleft lesions.
- Many first branchial anomalies cannot be categorized using the Work system.
- Classifying first branchial cleft anomalies based on germ layer is often inaccurate and should be discarded.
- Type I first branchial cleft anomalies are those that do not involve parotid tissue and are lateral to the parotid fascia (Fig. 2).
- Type II first branchial cleft anomalies are deep to the parotid fascia, with either the lesion or tract within the parenchyma of the parotid gland or between the parotid gland and auricular cartilage (Fig. 3).

Work up (Table 2).

- Ultrasound may be used as a screening method depending on the case presentation.
- MRI with contrast or MRI with/without contrast are recommended to evaluate parotid gland involvement.

Surgical Management (Table 2).

- Include a portion of parotid gland or perform a superficial or total parotidectomy when the lesion involves parotid gland.
- Place a drain if a parotid dissection is involved with resection of an anomaly.
- A modified Blair incision is preferred if lesion involves the parotid gland.
- Criteria for resection of ear canal cartilage and/or skin are if the lesion: is definitively involving canal skin or cartilage, is adherent to cartilage or contains a fistulous tract opening to the skin or cartilage
- Where possible, attempt to follow a tract and resect it entirely versus just amputating or cauterizing the tract.
- If the lesion is postauricular, the tract should be dissected with minimal or no cartilage resection.
- Pack ear canal post op if canal wall cartilage or skin is included in resection to avoid external auditory canal stenosis.

Figs. 2 and 3: Schematic drawings representing the IPOG classification of type I and II first branchial cleft anomalies.

4. Discussion

The mission of the International Pediatric Otolaryngology Group (IPOG) is to develop expertise-based recommendations for the management of pediatric otolaryngologic disorders [14]. Comprehensive management guidelines for children with first branchial cleft anomalies have not yet been published in the otolaryngology literature. Survey of expert opinion under the direction of IPOG and allied pediatric otolaryngologists was conducted to generate consensus recommendations for the classification and management of first branchial cleft anomalies.

The Delphi method is commonly used in natural science and medical literature when seeking to develop a consensus among experts [15]. Designed in the 1950s, Delphi is a process for developing an informed consensus among a selected panel of participants regarding a complex problem via data collection through questionnaires [15]. Panels typically range from 10 to 100 members, and participants need to be recognized as domain experts [15]. In contrast to a focus group, the Delphi method is designed to avoid direct confrontation and biases that may result from the most dominant perspective, allowing for more expression of individual opinions [15]. Weakness of the Delphi method include its vulnerability to drop outs and its reliance on experts' judgments as opposed to data [15]. Here, we will discuss first branchial cleft anomalies and the consensus guidelines generated from our Delphi method survey.

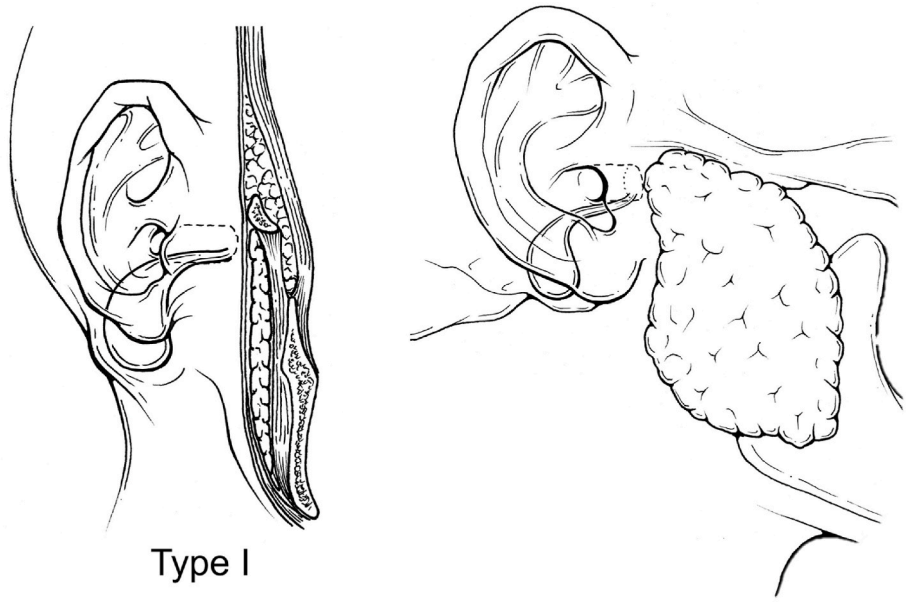

Fig. 2. Section and lateral views of type I first branchial cleft anomalies. The cyst and tract can be pre- or postauricular, are lateral to the parotid fascia and do not involve parotid parenchyma.

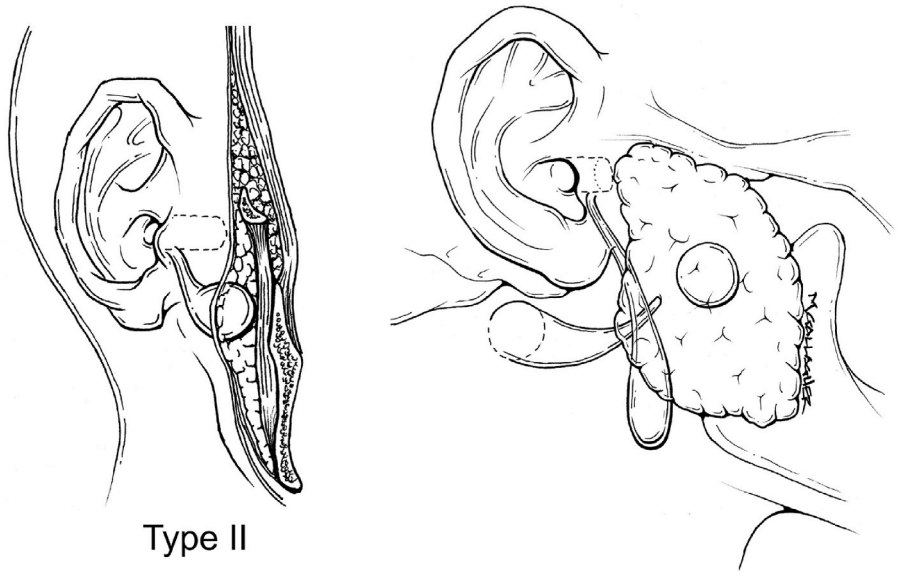

Fig. 3. Section and lateral views of type II first branchial cleft anomalies. The cyst and/or tract are deep to the parotid fascia or within the parenchyma of the parotid gland itself.

Table 1
Consensus recommendations for classification of first branchial cleft anomalies.

	Consensus Recommendation for Classification of First Branchial Cleft Anomalies
1	First branchial cleft anomalies represent reduplication of the ear canal
2	The Work classification is the most widely accepted system to categorize first branchial cleft lesions
3	Many first branchial anomalies cannot be categorized using the Work system
4	Classifying first branchial cleft anomalies based on germ layer is often inaccurate and should be discarded
5	Type I first branchial cleft anomalies are lateral to the parotid gland
6	Type II first branchial cleft anomalies are deep to the parotid fascia, either within the parenchyma of the parotid gland or between the parotid gland and auricular cartilage

Anomalies of the first branchial cleft occur secondary to incomplete fusion of the ventral part of the first and second branchial arches, typically around the seventh week of embryonic development [9]. This occurs around the same time as parotid and facial nerve development, thus there is an intimate relationship between these structures and first branchial cleft anomalies [9]. Aberrant embryologic development of the first branchial cleft results in malformed reduplication of the external auditory canal [2,10].

The Work classification is the most widely accepted schema for classifying first branchial cleft anomalies [7,10,11]. This classification system is based on location and germ layer comprising the cyst [2,7,10,11]. However, many otolaryngologists question the clinical applicability of the Work classification for first branchial cleft anomalies, particularly its emphasis on germ layer. The Work system does not consistently inform the technical approach required to completely resect these

Table 2
Consensus recommendations for work up and surgical management of first branchial cleft anomalies.

	Consensus Recommendation for Work Up and Surgical Management of First Branchial Cleft Anomalies
1	Ultrasound may be used as a screening method depending on the case presentation.
2	MRI with contrast or MRI with/without contrast are recommended to evaluate parotid involvement.
3	Include a portion of parotid gland or perform a superficial or total parotidectomy when the lesion involved parotid gland.
4	Place a drain if a parotid dissection is involved with resection of an anomaly.
5	A modified Blair incision is preferred if lesion involves the parotid gland.
6	Criteria for resection of ear canal cartilage and/or skin are if the lesion/tract: is definitively involving canal skin or cartilage or contains a fistulous tract opening to the skin or cartilage
7	Attempt to follow a tract and resect it entirely versus just amputating or cauterizing the tract.
8	If the lesion is postauricular, the tract should be dissected with minimal or no cartilage resection
9	Pack ear canal post op if canal wall cartilage or skin is included in resection

lesions. In 2018, Liu and colleagues found that Work type II lesions have a close relationship with the facial nerve [16]. Further, about half of Work type II lesions were located superficial to the facial nerve and half were deep to the nerve [16]. Facial palsy and external auditory canal stenosis are the most common complications of surgical resection of first branchial cleft anomalies [12,16].

In a 2023 study, Wilson and colleagues found that only 55 % of lesions were correctly categorized using the Work method [2]. Other studies also support the notion that classification of first branchial cleft anomalies based on germinal tissue type is of limited utility [1]. Wilson and colleagues suggested a new classification system based on involvement of the parotid gland, where type I cysts may be either postauricular or preauricular but superficial to the parotid gland, while type II cysts involve the parotid fascia, i.e., the superficial musculoaponeurotic system (SMAS), and/or the parotid gland to some degree [2]. They also found that type II cysts were more likely to have associated lymphadenopathy, involve inflamed or scarred tissue planes and have an increased rate of recurrence. Most notably, they identified that preoperative MRI was successfully able to predict parotid gland involvement, need for parotidectomy (partial or total) and facial nerve involvement [2].

A major impetus for the publication of these consensus recommendations was to update the classification guidelines for first branchial cleft anomalies, particularly to make them more clinically relevant for otolaryngologists. Building off the work previously shared by Wilson and colleagues, the findings of this IPOG survey of expert opinion support a revised classification system for first branchial cleft anomalies based on absence (type I) or presence (type II) of parotid gland involvement [2]. A recent publication by Kong and colleagues further supports the need for updated classification criteria [12]. The authors agree that the existing classification system presented by Work did not accurately determine the relationship between the lesions and the facial nerve. Kong et al. propose a system based on the relationship of lesions with the facial nerve and further differentiate lesions with significant scarring secondary to previous surgery [12]. Their approach involves localizing the facial nerve at the stylomastoid foramen on coronal T2-weighted MRI, further emphasizing the importance of MRI for diagnosis and surgical planning for resection of these lesions.

This IPOG survey of expert opinion has reached consensus that first branchial cleft anomalies should not be classified by germ layer but instead by the presence or absence of parotid gland involvement.

- Type I first branchial cleft anomalies are those that do not involve parotid tissue and are lateral to the parotid fascia.
- Type II first branchial cleft anomalies are deep to the parotid fascia, with the lesion and/or tract within the parenchyma of the parotid gland or between the parotid gland and auricular cartilage.

The expert panel reviewed additional recommendations for diagnostic work up and treatment, and consensus recommendations are discussed here. While many experts endorse that ultrasound can be used as a screening method when working up these lesions, no consensus was reached to strongly recommend the use of ultrasound for diagnosis. Ultrasound can certainly be a valuable part of the diagnostic algorithm, particularly if there are limitations to obtaining cross-sectional imaging. MRI, either with or with/without contrast, is recommended to evaluate parotid gland involvement. The sensitivity and specificity of MRI is generally preferred as there is excellent discrimination of soft tissue structures compared to CT [2,3]. Additionally, many pediatric otolaryngologists are advocates of MRI in order to avoid excess radiation in the pediatric population. MRI has been shown to be more accurate than ultrasound or CT in the characterization of first branchial cleft anomalies [16].

Consistent with the literature, consensus was reached regarding the recommendation for aggressive surgical management aimed at complete excision of these lesions, as recurrence rates are relative high (14–22 %) [17]. Recurrence of these lesions tends to follow a course of repeated infections resulting in hypertrophic scarring, and revision surgery becomes challenging with vital structures at risk [2,3,7]. There was no group consensus for facial nerve identification and dissection when a portion of the parotid gland is resected; however, the majority of those surveyed report always finding the facial nerve, and the remainder of respondents indicate determining the need for facial nerve dissection based on the nature of the lesion. For those responding that use of facial nerve monitoring depends on the lesion, free text responses suggested that most ultimately use nerve monitoring and would only consider withholding monitoring in cases where there is a small lesion low in the parotid tail. The senior author advocates for facial nerve monitoring in any case where there is suspected parotid gland involvement. There was no group consensus regarding parotid dissection technique, aside from utilizing a Modified Blair incision when parotidectomy was indicated and placing a drain when either resecting a portion of parotid or performing a superficial or total parotidectomy.

Additional surgical recommendations include resecting ear canal cartilage or skin if either are definitively involved, or if the lesion is adherent to these structures. If either ear canal skin or cartilage is included in the resection, the ear canal should be packed to prevent external auditory canal stenosis. In cases where there is a fistulous tract opening to the skin of the face or neck, the fistula and surrounding skin should be resected. Where possible, an attempt should be made to follow a tract and resect it in its entirety. Minimal to no cartilage excision is expected when resecting a postauricular lesion. In general, more aggressive surgery involving cartilage resection, facial nerve dissection and resection of parotid tissue pertains to type II lesions.

The findings of this IPOG survey of expert opinion delineate classification, diagnostic and treatment recommendations for pediatric patients with first branchial cleft anomalies. These recommendations are based on extensive review of the existing literature as well as survey of expert practice patterns. As these guidelines are not based on formal data collection and analysis, recommendations are subject to change based on future research in this field.

5. Conclusion

First branchial cleft anomalies are rare congenital head and neck lesions. Literature pertaining to classification, work up and surgical treatment of these lesions is limited. The findings of this work support a revision of the classification system for first branchial cleft anomalies that is based on either the presence or absence of parotid gland involvement, without emphasis on germ layer.

- IPOG Type I first branchial cleft anomalies are those that do not involve parotid tissue and are lateral to the parotid fascia.
- IPOG Type II first branchial cleft anomalies are deep to the parotid fascia, with the lesion and/or tract within the parenchyma of the parotid gland or between the parotid gland and auricular cartilage.

Additional classification, diagnostic and treatment recommendations for the management of these lesions are discussed. Inherent in this discussion is that more aggressive surgery including cartilage resection, facial nerve dissection and resection of parotid tissue pertains to type II lesions. These consensus recommendations are aimed at improving care of children presenting with first branchial cleft anomalies.

CRediT authorship contribution statement

Ashley L. Heilingoetter: Writing – review & editing, Writing – original draft, Resources, Methodology, Data curation, Conceptualization. **Goh Bee See:** Writing – review & editing, Methodology, Investigation. **James Brookes:** Writing – review & editing, Methodology, Investigation. **Paolo Campisi:** Writing – review & editing, Methodology, Investigation. **Sergio Santino Cervantes:** Writing – review & editing, Methodology, Investigation. **Neil K. Chadha:** Writing – review & editing, Methodology, Investigation. **Daniel Chelius:** Writing – review & editing, Methodology, Investigation. **Diane Chen:** Writing – review & editing, Methodology, Investigation. **Bob Chun:** Writing – review & editing, Methodology, Investigation. **Michael J. Cunningham:** Writing – review & editing, Methodology, Investigation. **Jill N. D'Souza:** Writing – review & editing, Methodology, Investigation. **Taseer Din:** Writing – review & editing, Methodology, Investigation. **Titus Dzongodza:** Writing – review & editing, Methodology, Investigation. **Christian Francom:** Writing – review & editing, Methodology, Investigation. **Thomas Q. Gallagher:** Writing – review & editing, Methodology, Investigation. **Mark E. Gerber:** Writing – review & editing, Methodology, Investigation. **Michael Gorelik:** Writing – review & editing, Methodology, Investigation. **Steven Goudy:** Writing – review & editing, Methodology, Investigation. **M. Elise Graham:** Writing – review & editing, Methodology, Investigation. **Benjamin Hartley:** Writing – review & editing, Methodology, Investigation. **Inbal Hazkani:** Writing – review & editing, Methodology, Investigation. **Paul Hong:** Writing – review & editing, Methodology, Investigation. **Wei-Chung Hsu:** Writing – review & editing, Methodology, Investigation. **Andre Isaac:** Writing – review & editing, Methodology, Investigation. **Kris R. Jatana:** Writing – review & editing, Methodology, Investigation. **Douglas R. Johnston:** Writing – review & editing, Methodology, Investigation, Conceptualization. **Fiona Kabagenyi:** Writing – review & editing, Methodology, Investigation. **Ken Kazahaya:** Writing – review & editing, Methodology, Investigation. **Jeff Koempel:** Writing – review & editing, Methodology, Investigation. **Nicolas Leboulanger:** Writing – review & editing, Methodology, Investigation. **Romain Luscan:** Writing – review & editing, Methodology, Investigation. **Sarah E. Maurrasse:** Writing – review & editing, Methodology, Investigation. **Erika Mercier:** Writing – review & editing, Methodology, Investigation. **Shazia Peer:** Writing – review & editing, Methodology, Investigation. **Diego Preciado:** Writing – review & editing, Methodology, Investigation. **Reza Rahbar:** Writing – review & editing, Writing – original draft, Resources, Methodology, Investigation, Data curation, Conceptualization. **Jeffrey Rastatter:** Writing – review & editing, Methodology, Investigation. **Gresham Richter:** Writing – review & editing, Methodology, Investigation. **Steven D. Rosenblatt:** Writing – review & editing, Methodology, Investigation. **Sophie G. Shay:** Writing – review & editing, Methodology, Investigation. **Anthony Sheyn:** Writing – review & editing, Methodology, Investigation. **Yilkal Tassew:** Writing – review & editing, Methodology, Investigation. **Patrick C. Walz:** Writing – review & editing, Methodology, Investigation. **Amy S. Whigham:** Writing – review & editing, Methodology, Investigation. **Joshua P. Wiedermann:** Writing – review & editing, Methodology, Investigation. **Jeffrey Yeung:** Writing – review & editing, Methodology, Investigation. **John Maddalozzo:** Writing – review & editing, Writing – original draft, Supervision, Resources, Methodology, Investigation, Formal analysis, Data curation, Conceptualization.

Declaration of competing interest

Below we have listed any relevant disclosures or conflicts of interest among the authors of our manuscript, "Comprehensive management and classification of first branchial cleft anomalies: An International Pediatric Otolaryngology Group (IPOG) consensus statement."

Daniel Chelius, MD: Leadership role and stipend as AAO-HNSF Annual Meeting Coordinator, 2021–2024.

Steven Goudy, MD, MBA: Founder and Chief Medical Officer of Dr. Noze Best.

Kris Jatana, MD, FACS, FAAP: Royalties (Marpac Inc.), Shareholder (Tivic Health Systems), Officer/Shareholder (Zotarix LLC, in collaboration with Grace Medical).

For all remaining authors of this work, there are no conflicts of interest or financial disclosures to report.

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