Journal of Otology 13 (2018) 122–127

Contents lists available at ScienceDirect

Journal of Otology

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

Classification of facial nerve aberration in congenital malformation of middle ear: Implications for surgery of hearing restoration☆,

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ARTICLE INFO

Keywords:
Congenital middle ear anomaly
Facial nerve
Stapes
Oval window
Vibrant Soundbridge (VSB)

ABSTRACT

Objectives: Facial nerve aberration is the most troublesome situation in congenital malformations of middle ear. The aim of our study is to investigate its imaging and clinical features as well as relevant choice of surgical techniques for hearing improvement.

Methods: A retrospective study involving review of clinical data of 227 patients (256 ears) with congenital middle ear anomaly was undertaken, including preoperative computed tomography (CT) data, surgical records and videos.

Results: Aberration involving intratemporal facial nerve was found in 82/256 ears (32.03%) with congenital middle ear anomaly. The most common forms of aberration included overhanging over the oval window (50/82 ears, 60.98%), bifurcation (3/82 ears, 3.66%) and transverse over the promontory (3/82 ears, 3.66%), counting for 68.29% (56/82) of the cases with facial nerve aberration. Concomitant stapes malformation was found in 76/82 ears (92.68%) and atresia or stenosis of the oval window in 27/82 ears (32.93%). In 9/82 ears (10.98%) both stapes and oval window was absent. Elective surgeries for the purpose of hearing improvement included stapedotomy + piston implantation, labyrinthotomy, labyrinthotomy + total ossicular replacement prosthesis (TORP) implantation and Vibrant Soundbridge (VSB) implantation.

Conclusion: The majority of facial nerve aberration in congenital malformation of middle ear involves displacement of facial nerve, in addition to concomitant malformations of the stapes and/or oval window, which may influence the choice of surgery for hearing improvement. VSB implantation may be considered as a useful option.

1. Introduction

The aberrant intratemporal facial nerve is the most troublesome condition during middle ear surgery. Failure to recognize the mal-positioned facial nerve can be devastating. Before the early 1960s, it was believed that anomalies of the facial nerve do not occur until Fowler firstly reported a series of “hump anomaly” with facial nerve hump posterior and lateral to the prominence of the external semicircular canal (Fowler, 1961). The congenital malformation of facial nerve was gradually recognized, but still mainly in cases with microtia (Dickinson et al., 1968) or Treacher Collin's syndrome (Sando et al., 1968). For decades, accumulating data of aberrant facial nerve in congenital malformation of middle ear with no co-existing malformation of the outer ear were reported in a form of case report (Kieff et al., 1998; Inagaki et al., 2014) until very recently when Jahrsdoefer studied 54 patients with congenital middle ear malformations. He reported finding aberrant courses of facial nerve in up to almost 1/4 of ears (Jahrsdoerfer, 1981).

In congenital malformation of middle ear, the aberrant intratemporal facial nerve is extremely tricky for two reasons. Firstly, while the surgeon would be warned of possible facial nerve aberration when microtia and aural atresia are present, in isolated congenital malformation of middle ear, however, very limited information on the facial nerve may be available before surgical exploration. Secondly, due to the close anatomic relationship among the facial nerve, stapes and oval window (OW) (Som et al., 2016), intratemporal facial nerve displacement may likely influence the choice of surgery for hearing improvement (Jahrsdoerfer, 1981). In severe cases, aborted surgery is not rare (Liu et al., 2017). Accurate preoperative assessment on facial nerve is a critical part of surgical planning.

Computed tomography (CT) has been evolved and widely applied in preoperative evaluation of the fine structures of middle ear. Allowing

☆ This study was supported by Beijing Municipal Science & Technology Commission (No. Z171100001017079) for Prof. Shouqin Zhao.
Peer review under responsibility of PLA General Hospital Department of Otolaryngology Head and Neck Surgery.
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https://doi.org/10.1016/j.joto.2018.09.002
Received 30 April 2018; Received in revised form 9 September 2018; Accepted 19 September 2018

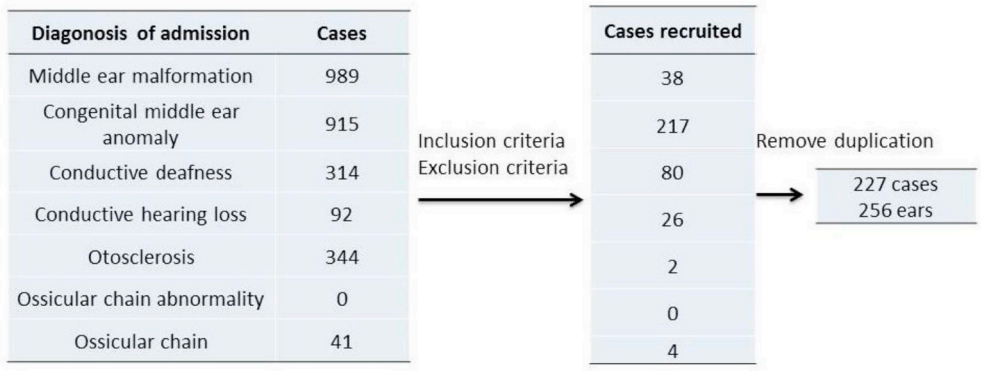

Fig. 1. Flow chart of case selection for the diagnosis of "congenital malformation of middle ear". The diagnosis was confirmed by intraoperative findings.

identification of relationships between the fallopian canal to the surrounding bony structures, CT has its advantages (Gupta et al., 2013). High-resolution computed tomography (HRCT) with multi-planar reconstruction (MPR) has been used to evaluate the whole intratemporal facial nerve from the labyrinthine segment to the mastoid segment (Murai et al., 2013). For cases with abnormal facial nerve courses together with OW atresia and malformed stapes, HRCT with MPR is helpful in surgical planning (Liu and Yang, 2015).

In this study, we compared intraoperative findings of middle ear anatomy with preoperative CT findings in order to optimize imaging and clinical evaluation of aberrant courses of intratemporal facial nerve in congenital malformation of middle ear. Accordingly, the choice of surgery for hearing improvement in such cases was also discussed.

2. Methods

A retrospective case review study was conducted. Electronic charts from January 2000 until December 2017 in the Department of Otolaryngology, Beijing Tongren Hospital of Capital Medical University, were reviewed. Diagnosis of "congenital malformation of middle ear" of the cases included in the study was confirmed by operation records and videos. During chart searching, "admission diagnosis" was screened for "middle ear malformation", "congenital middle ear anomaly", "conductive deafness", "conductive hearing loss", "otosclerosis", "ossicular chain abnormality" and "ossicular chain". Charts with admission diagnosis of "otosclerosis" were also browsed for possible mis-diagnosis in order to minimize unnecessary omissions.

Inclusion criteria: Hearing loss since birth; tympanic membrane intact under otoscopy; conductive hearing loss by pure tone audiometry (PTA); if mixed hearing loss, dominated by conductive loss; ossicular chain abnormality confirmed on operation records and videos.

Exclusion criteria: Chronic suppurative otitis media, otitis media with effusion, traumatic ossicular fracture, microtia, aural atresia, stenosis of external auditory canal (EAC) with or without cholesteatoma, and otosclerosis. Cases with malformation of the inner ear with remarkable bone conduction threshold elevation were also excluded.

Two investigators reviewed operation records independently. When there was any disagreement, discussion was undertaken until an agreement was reached. Based on the inclusion and exclusion criteria, 227 cases (256 ears) of congenital malformation of middle ear were drawn from the electronic chart system after duplication removal (Fig. 1). The median ± SD age at surgery was 20.4 ± 10.7 years. The male to female ratio was 1.55:1. Among the 256 ears, the leading cause of operation was hearing loss (n = 248, 96.9%), suppurative discharge (n = 6, 2.3%), tinnitus (n = 1, 0.4%) and vertigo (n = 1, 0.4%). Facial paresis was found in one case before surgery. Involvement of tympanic segment and genu of facial nerve by cholesteatoma was indicated by CT scanning before surgery and confirmed in the operation.

3. Results

3.1. The most common aberrations of the facial nerve

Among the 256 ears with isolated congenital malformation of middle ear, the facial nerve was found to be abnormal in 82/256 ears (32.03%). In the 82 ears, 37 (45.12%) showed facial nerves with failed osseous closure of the fallopian canal, of which 23 (62.16%) showed facial nerves located anteriorly and inferiorly, partially or completely covering the OW. Facial nerve in the rest 14 of the 37 ears (37.84%) were in a normal anatomic location without ptosis.

Facial nerve displacement could exist alone or co-exist with dehiscence. Regardless of dehiscence, the most common displacement, found in 50/82 ears (60.98%), was anterior and inferior, partially or completely covering of the OW (Fig. 2, 1A-1C). Bifurcation of the facial nerve is one of the most confusing conditions surgeons may encounter. In our series, bifid facial nerve was found in 3/82 ears (3.66%). The divergent branches encircled the OW (Fig. 2, 2A-2C). During the surgery, facial nerve stimulation confirmed the anomalous split facial nerve configuration. When facial nerve was located even further anteriorly and inferiorly, it could be found crossing the promontory (3/82 ears, 3.66%). We also described this anatomic variation as crossing between the OW and the round window (RW) (Figs. 2,3A-3C).

Taken together, following the dehiscence of facial nerve (37/82 ears, 45.12%), the second most common aberration of facial nerve in this series of congenital malformation of middle ear was displacement (56/82 ears, 68.29%), including hanging over the OW (50/82 ears, 60.98%), bifurcation (3/82 ears, 3.66%) and crossing over the promontory (3/82 ears, 3.66%) (Table 1).

3.2. Concomitant malformation of stapes and OW

Among the 82 ears with aberrant facial nerve, 76 (92.68%) were also found to have concomitant malformation of stapes and 27 (32.93%) had atresia or stenosis of OW. Absence of both stapes and OW occurred in 9/82 ears (10.98%).

Among malformations of stapes, isolated fixation of stapes footplate is the least severe, for which pre-operative CT scanning provides hardly any useful information. Among our 82 ears with aberrant facial nerve, isolated fixation of stapes footplate was found in 13/82 ears (15.85%) according to operative records, of which preoperative CT was normal in 7 ears (Ear 73#, 111#, 157#, 174#, 2-2#, 2–29#, 2–41#), and suggested otosclerosis 5 ears (Ear 34#, 103#, 126#, 183#, 2–30#) due to low density around the OW area and "probable" stapes disconnection from the OW in 1 ear (Ear 130#).

Atresia of the OW is relatively easy to be identified on CT scans, especially on coronal projection. OW stenosis, however, is somewhat challenging. In our series, stenosis of the OW was found in 4/82 ears (4.88%) during the operation. Preoperative CT reported narrow OW

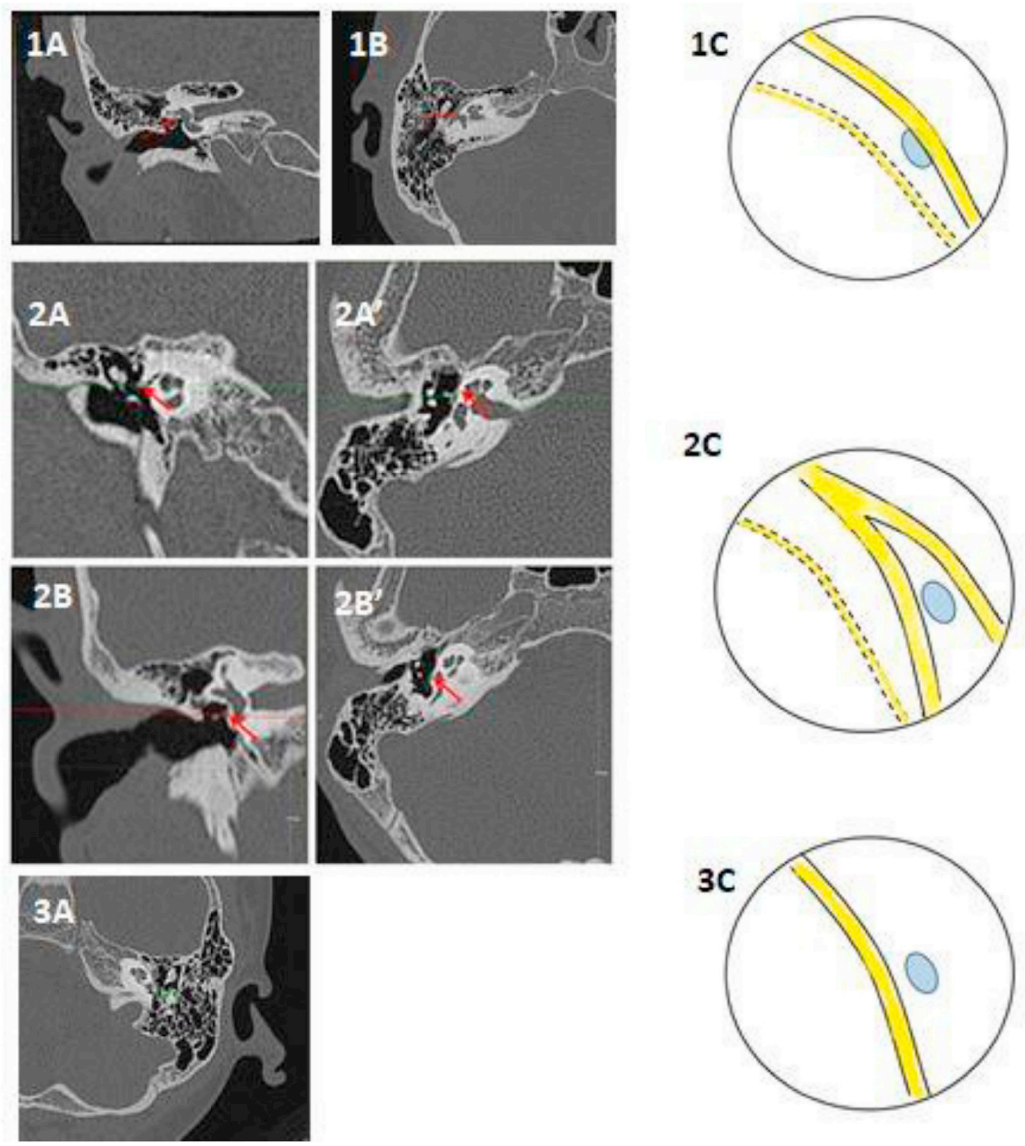

Fig. 2. CT images and schematic diagram of displacement of the facial nerve. 1A and 1B are coronal and axial CT images of the facial nerve partially covering the OW (Red arrows). **2A and 2A′** show one branch of a bifid facial nerve. with the other branch seen on **2B and 2B**' (Red arrows). **2C** is a schematic diagram in the operation record of bifurcation of the case with the location of stapedotomy and piston implantation. **3A and 3C** show a displaced facial nerve crossing over the promontary (Green arrow). Solid line - facial nerve; dotted line - chorda tympani.

niche in only one ear (Ear 148#). In the other 3 ears, CT revealed malformation of the ossicular chain but no description of the OW.

3.3. Choice of surgery for hearing improvement

Since the leading cause of operation was hearing loss (248/256 cases, 96.9%), hearing improvement procedures were performed in the majority of cases after middle ear exploration (Table 2). Cholesteatoma hindered primary stage hearing restoration in 4 ears and secondary stage hearing improvement surgery was expected when clearance of cholesteatoma could be assured. After releasing adhesion, the ossicular chain was found to be intact with good mobility in 14 ears, allowing immediate. tympanoplasty.

Base on the condition of stapes and mobility of the foot plate, partial ossicular replacement prosthesis (PORP), total ossicular replacement prosthesis (TORP) or piston was chosen for hearing reconstruction. Autogenous bone prosthesis was preferably chosen by two otologists from 2002 to 2005 and used sporadically afterwards until 2009 (24 ears). Fenestration of semicircular canal was used in 2 ears. Vibrant sound bridge (VSB) was used in 6 ears. Floating mass transducer (FMT) was installed on the head of stapes in 2 ears. Due to OW atresia and/or blockage of OW by displaced facial nerve, RW-vibroplasty was performed in 4 ears. Pre-operative CT scanning in Patient CX (Ears 3–11# and 3–12#) indicated absent OW bilaterally (Fig. 3A). Fenestration of semicircular canal was performed on the left ear when he was 5 as VSB was yet available at that time. The parents chose VSB implantation for the child on the right side 10 years later (Fig. 3B). Thickening and fixation of the footplate was found during operation in Ear 3–14#, with the OW partially blocked by the displaced facial nerve. Stapedotomy was performed with piston implantation with dissatisfactory hearing outcomes. After one year, VSB was implanted with RW-vibroplasty in the same ear. Due to severe malformation of the middle ear, no hearing improvement surgery could be performed after exploration in 12 ears.

In all cases with displacement of facial nerve, there was partial or complete blockage of the OW by the nerve from highly variable malformations. Stapedotomy with piston implantation was performed on the fixed footplate between the divergent branches of facial nerve in 3 ears. In the 3 ears with facial nerve crossing over the promontory, the

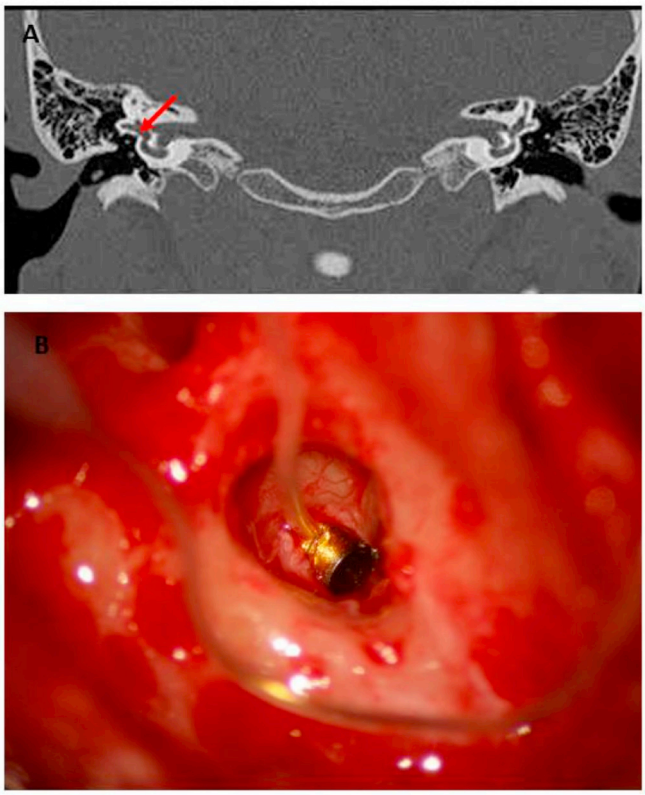

Fig. 3. Coronal CT scan (A, red arrow) and placement of floating mass transducer (FMT) of VSB on the round window membrane (B) in the right ear of Patient CX.

Table 1
The most common aberration of facial nerve in congenital middle ear anomaly.

The most common aberrations of facial nerve	
1.Dehiscence	
With anterior/inferior displacement	23 ears
At normal anatomic location	14 ears
	37/82 ears (45.12%)
2.Displacement	
Covering the oval window	50 ears
Bifurcation	3 ears
Crossing over the promontory	3 ears
	56/82 ears (68.29%)

Table 2
Types and number of hearing improvement procedures performed.

The surgery for hearing improvement	Cases
No implantation due to cholesteatoma	4
Intact ossicular chain with good activity after adhesion removal	14
PORP implantation	37
TORP implantation	54
Piston implantation	103
Autogenous bone prosthesis implantation	24
Fenestration of semicircular canal	2
VSB implantation	6
Operation ceased due to severe malformation	12
Total	238

foot plates were fixed and piston prosthesis was used in 2 ears and autogenous bone prosthesis in 1 ear.

4. Discussion

In this series of congenital middle ear anomaly (227 cases, 256 ears), aberrant facial nerve was found in 82/256 ears (32.03%), with dehiscence of the fallopian canal in 37/82 ears (45.12%), which was slightly lower than the previously reported 55% (Baxter, 1971). Instead, the incidence of displacement of facial nerve reached 68.29% (56/82 ears), much higher than the previously reported 24% (Jahrsdoerfer, 1981). This may be partially explained by better observation with improved microscopy equipment in addition to intraoperative facial nerve monitoring. The degree of aberrant facial nerve in congenital malformation of middle ear varies. But otologists should be mindful that the incidence of facial nerve malformation is not low.

Preoperative CT scanning failed to alert bifurcation of the facial nerve in all 3 ears in this study. Bifurcation of the facial nerve has been previously reported, which often accompanies other malformations of the outer-middle/inner ear (Durcan et al., 1967; McRakan et al., 2014), or is seen in syndromic conditions, such as Branchio-oto-renal (BOR) syndrome (Glastonbury et al., 2003). When enough space is left between the divergent branches encircling the oval window, stapedotomy with piston implantation could be performed to improve hearing as in our series.

Existence and proper location of the stapes and oval window are vital for hearing restoration. In our study, absence of both stapes and oval window occurred in 9/82 ears (10.98%) combined with aberration of facial nerve. The close anatomic relation among facial nerve and stapes and oval window derives from its embryology. The facial nerve originates from a collection of neural crest cells in close caudal proximity to the otic placode, which is known as the facioacoustic primordium. The facial and auditory parts of the primordium demarcate by the end of the fourth week of gestation. In the fifth week of gestation, the facial primordium has differentiated distally into the main trunk of the facial nerve and the chorda tympani nerve. Meanwhile, the facial motor nucleus can be identified in the developing brain stem. During the sixth week of gestation, the facial nerve bends caudally at the level of the geniculated ganglion, forming the external genu. A short horizontal segment then passes between the developing membranous labyrinth and the upper end of Reichert's cartilage, which will become the blastema of the stapes. The facial nerve then bends vertically before passing into the substance of the second branchial arch. The chorda tympani nerve by the seventh week has assumed a ventrally concave configuration—as in the normal ear—and enters the first (mandibular) branchial arch. By the eighth week, the orientation of the facial nerve within the temporal bone has been established (Jahrsdoerfer, 1988).

The most common displacement of the facial nerve is in the anterior and inferior direction, therefore covering the OW. Why? Gerhardt and Otto explained that anterior displacement of the facial nerve results from possible underdevelopment and shortening of the first branchial arch, which in turn allows the second branchial arch to overshift in a rostral direction. The facial nerve, being the nerve of the second branchial arch, follows this shift (Gerhardt and Otto, 1981).

In terms of the embryonic relation of facial nerve to the stapes, there are two explanations. One, the stapes develops slightly later than the facial nerve. In the early part of the seventh week, Reichert's cartilage first appears, but by this time the horizontal portion of the facial nerve has already been established. If extrinsic forces (e.g., hyoid arch shifting) have occurred, the nerve may be displaced anteriorly before the stapes has developed. This would explain those rare middle ear anomalies wherein a bare facial nerve is located anterior and inferior to an intact stapes and oval window, as the facial nerve crossing over the promontory in our study. A more common scenario results from a time-related migration of the facial nerve anteriorly while the stapes is forming. The nerve may then be interposed between the stapes blastema and the labyrinth, preventing the formation of the footplate and oval window. Additionally, one or both

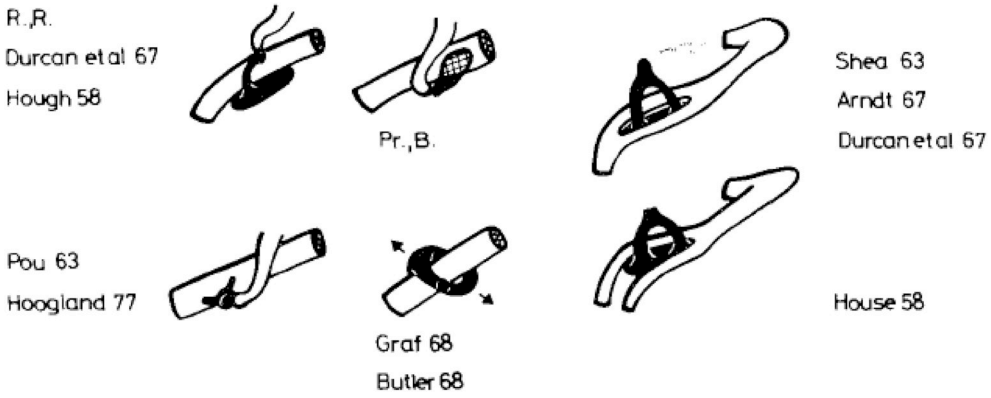

Fig. 4. Dysplasias of the stapes may be due to developmental disturbances caused by a dystopic nerve (Gerhardt and Otto, 1981).

stapes crura may fail to develop, or the crura may be small and the rudimentary stapes free-hanging (Fig. 4). Jarhsdoefer believed that the development of the facial nerve influences the development of the stapes, rather than the reverse (Gerhardt and Otto, 1981; Jahrsdoerfer, 1988). The knowledge of embryonic derivation of facial nerve and its relation with stapes and oval window may suggest the relevant malformation during the middle ear surgery.

The primary goal of the operation for most of the patients (248/256 cases, 96.9%) in our study was to improve hearing. But among so many procedures listed in Table 2, how do the otologists choose the optimal surgery for each case?

A general principle of otologic surgery is to eradicate cholesteatoma lesions before hearing restoration. Congenital cholesteatoma can be present in cases of congenital malformation of middle ear. When the middle ear cavity is involved by cholesteatoma, clearance of cholesteatoma is the primary goal of surgery in the initial stage. The chance of hearing restoration is reserved in the “second look” surgery. Large amount of cholesteatoma was found in the only case with facial paresis before surgery in our series. Malleus and Incus were absent and the footplate fixed without suprastructure. In accordance to the above principle, we chose clearance of cholesteatoma + “second look” as the management for this case. When the middle ear is not involved by cholesteatoma, hearing restoration is then the primary goal of surgery. In 14 ears in our series, intact ossicular chain with clear middle ear activity was found after adhesion removal, leading to subsequent tympanoplasty (Fig. 5).

The anatomic relationship between the aberrant facial nerve and the stapes/oval window plays a key role in hearing restoration. If the stapes is not or only partially blocked by the facial nerve, adjustable and unadjustable prostheses with variable heights and designs, including PORP, TORP and piston, may be implanted based on the integrity and mobility of the stapes. Even though bifurcation of the facial nerve was found in 3 cases in our series, piston was implanted through the fixed footplate of stapes because it was not blocked by the divergent branches of facial nerve. Regarding the material of implant, titanium provides good biological compatibility and auditory performance. As a result, autogenous bone prosthesis implantation was abandoned.

When the aberrant facial nerve blocks the stapes/OW, or the OW is absent (Jahrsdoefer's type III) (Jahrsdoerfer, 1981), classic ossicular chain restoration is not possible. In our series, surgery was aborted due to severe malformation in 12 ears. Fenestration is a choice for hearing improvement. In our series, fenestration of semicircular canal was performed in 2 ears. Hasegawa et al. reported surgical treatment in 3 cases of congenital

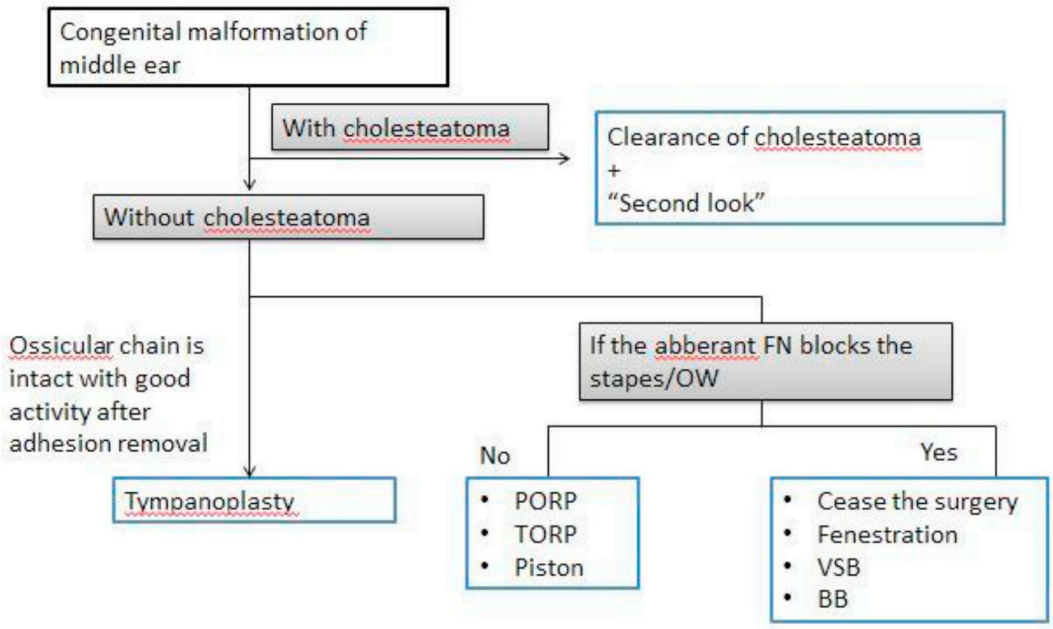

Fig. 5. Proposed algorithm for the management of congenital malformation of the middle ear. BB, bone bridge; FN, facial nerve; OW, oval window; PORP, partial ossicular replacement prosthesis; TORP, total ossicular replacement prosthesis; VSB, Vibrant sound bridge.

absence of the oval window with facial nerve anomalies. Fenestration of the vestibule above the facial nerve was performed in 2 cases, one combined with piston implantation and one without. In the other patient, fenestration of the scala vestibule below the facial nerve was performed with piston implantation (Hasegawa et al., 2012). The optimal location of fenestration is controversial. Recently, Yang and his colleagues reported 3 cases with oval window atresia and malformed facial nerve that were treated with the scala tympani drill-out technique combined with TORP implantation. They regarded the initial part of scala tympani as the optimal location of fenestration, corresponding to the promontory wall anterior-inferior to the RW membrane (not the niche). At this location, vibration through the fenestration can be best conducted to the perilymph in the scala tympani. Anterior-superior to the RW membrane should be avoided (Yang et al., 2016).

Regardless of the location, fenestration increases the risk of labyrinthitis after all. Lately, new designs of hearing aids and implants may help avoid opening the inner ear and/or provide better audiological benefits. There are 3 main types: active middle ear implants, active bone conduction implants and passive bone conduction implants. All three types of implants improve speech perception, speech recognition, signal-to-noise ratio and directional hearing. Decision making on the type of implant depends on the extent of malformation and hence preoperative imaging (Frenzel, 2018). Imaging assessment of middle ear structures, particularly the anatomic relationship between facial nerve and stapes/OW, helps selection of the most optimal strategy of hearing improvement (Fig. 5).

In our series, VSB, an active middle ear implant, was implanted in 6 ears for hearing improvement. Among them, floating mass transducers were implanted on the head of stapes in 2 ears and in the other 4 ears VSB was implanted together with RW-vibroplasty. Due to its design, VSB can significantly improve hearing thresholds, especially at high frequencies, and speech recognition scores (Zhao et al., 2016). Therefore, application of VSB to the RW membrane can be an efficient therapy for congenital OW atresia or when OW is completely obscured by the aberrant facial nerve.

Baha systems and Ponto systems are percutaneous bone conduct implant systems whose disadvantages are high infection rate, fixture losses and need for revision surgery. An active transcutaneous bone conduct implant, Bonebridge system, has been developed to overcome the weaknesses of percutaneous systems. With its active transcutaneous design, the Bonebridge offers a lower complication rate and higher and more reliable hearing gain compared to other transcutaneous or percutaneous systems. Moreover, early activation of the implant system enables the recipient to benefit in a short time frame postoperatively from the intervention. Until the collection of clinical data in our series, Bonebridge has not been applied in congenital malformation of middle ear. Without taking the risk of inner ear fenestration or even middle ear exploration, patients with congenital malformation of the middle ear can benefit from Bonebridge with a more reliable audiological outcome (Sprinzl and Wolf-Magele, 2016).

Since this is a retrospective study, an obvious weakness is the lack of follow-up data regarding long term hearing improvement, and therefore incomplete evaluation of short- and long-term outcomes for the different procedures of choice used. In future studies, attention will be directed to not only pre-operative CT scans and operative findings but also post-operative audiologic outcomes. Apart from one case with preoperative facial nerve paresis due to massive cholesteatoma, no facial nerve paresis was recorded in the chart during hospital stay following surgery.

5. Conclusion

In congenital middle ear anomaly, aberration of facial nerve is not rare (32.03%). The most commonly encountered malformation of facial nerve is facial nerve displacement (68.29%) rather than the previously reported dehiscence of fallopian canal. Facial nerve displacement can include blockage of the oval window, bifurcation and crossing over the promontory. Facial nerve displacement and accompanying malformation of stapes and OW influence the choice of surgery for hearing improvement. New designs of hearing aids and implants provide new options for patients with potential benefits of more reliable and better audiological outcomes. Future investigations should be focused on the short- and long-term outcomes for different hearing improvement surgeries.

Declaration of conflict of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

Acknowledgements

J.H. initiated this study, reviewed the charts and wrote the manuscript. LS.L. was in charge of the assessment and processing of CT images. XX.F. was the other independent investigator who reviewed the charts. SQ.Z. did most of the surgeries recruited in this study. She offered operation videos for analysis. J.H. revised the manuscript.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.joto.2018.09.002.

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