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ORIGINAL ARTICLE

Aberrant Course of the Intratemporal Facial Nerve in Children with Congenital Hearing Loss

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Abstract
Congenital facial nerve (FN) malformations are uncommon. Our aim is to determine the clinical and radiological features of FN malformations along with the associated cochlear and vestibular malformations. We conducted a retrospective study including children with a profound sensorineural hearing loss who were candidates for cochlear implantation. We evaluated the presence of FN malformations through temporal bone computed tomography scan and magnetic resonance imaging. We recorded an aberrant FN course in five out of 165 patients in a total of 9 ears. They consisted of a bifurcation of the mastoid segment, an anterior or posterior displacement of the labyrinthine segment and a hypoplasia of the geniculate ganglion. Associated inner ear malformations included vestibular aqueduct dilation, cochlear hypoplasia and total labyrinthine aplasia. We noted a bilateral agenesis of the cochlear nerve in three patients. Facial nerve malformations should be suspected in patients presenting a congenital hearing loss especially in association with other temporal bone malformations. Their pre-operative discovery is helpful in planning the surgical procedure.

Keywords Facial Nerve · Temporal bone · Malformation · Hearing loss

Introduction

Congenital malformations of the facial nerve (FN) are uncommon occurrences. They can be explained by various embryological factors [1]. Abnormalities along the course of the FN mainly occur in association with other inner and middle ear malformations and can involve all segments of the intra temporal FN [2]. The labyrinthine segment (LS) was reported as the most affected portion [3]. The identification of any aberrant FN malformation is important prior to middle ear surgery to decrease the risk of intra-operative injury to the nerve. Computed tomography (CT) scan identifies the presence of Fallopian canal variations [4, 5]. However, not performing CT scan pre-operatively can result in determining the FN aberration only during surgery, which puts the nerve at a high risk of injury [6]. The reported malformations consist mainly of FN Fallopian canal dehiscence, anteromedial displacement of the FN or less commonly bifurcation of the nerve [7].

Here we review the cases of children referred to our department for a congenital, profound and bilateral hearing loss in whom the diagnosis of a FN aberration was established based on CT scan. Our aim is to determine the clinical and radiological features of FN malformations, the associated vestibulocochlear nerve (VCN) malformations and their surgical implications.

Materials and methods

We conducted a retrospective study that concerned children referred to our department for a profound congenital sensorineural hearing loss (SNHL) in the period between January 2016 and June 2022.

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A CT scan and a magnetic resonance imaging (MRI) were performed in all patients prior to the cochlear implant surgery. CT scans were conducted using a Siemens scanner. Images through the petrous temporal bone were obtained in both axial and direct coronal planes. The minimal slide thickness was 0.3 mm for coronal slides and 0.6 mm for axial slides. MRI was performed using a 1.5 Tesla MRI machine, and the following sequences were obtained:

- Axial T2 high resolution (CISS) with a 0.7 to 1 mm slide thickness.
- Axial and coronal T2 and T1 slides with gadolinium injection.
- Diffusion-weighted images.

The analysis of the CT scan and MRI included the following parameters:

- External, middle and inner ear malformation: To define inner ear malformations, we utilized the classification published by Sennaroğlu et al. ,which includes the following possible malformations: Complete Labyrinthine Aplasia (Michel Deformity), Rudimentary Otocyst, Cochlear Aplasia, Common cavity, Cochlear hypoplasia, Incomplete Partition of the Cochlea, Enlarged Vestibular Aqueduct and Cochlear Aperture Abnormalities [8, 9].
- The diameter of the internal auditory canal (IAC) and its components focusing on the VCN and the FN: A diameter of the IAC lower than 3 mm was considered indicative of IAC hypoplasia of the IAC, while a normal cochlear nerve (CN) diameter was considered to be 1 mm. Hypoplasia of the CN when its diameter was lower than that of the contralateral CN [9, 10].
- FN abnormalities along its temporal bone course: For each malformation, we determined the exact abnormal segments and specific abnormalities, either positional or structural. Positional abnormalities were determined based on anatomical landmarks within the temporal bone (cochlea, oval window, round window, lateral semi-circular canals). Regarding abnormal divisions of the FN, we determined the number of divisions and their localizations [9].

We then included patients who met all the following criteria:

- Children with a bilateral profound sensorineural hearing loss who were candidates for cochlear implantation.
- Aberrant facial nerve course confirmed based on MRI and/or CT scan.

We excluded patients who met at least one amongst these criteria:

- Having a hearing loss without being candidates for cochlear implantation.
- Facial nerve abnormalities consisting of a dehiscence of the Fallopian canal.

Results

We included 165 patients in the study. We recorded an aberrant FN course in a total of 5 patients and 9 ears which accounted for 3% of the total patients. The median age of patients presenting an aberrant facial nerve course was 2.8 years old [2;3] with a male-to-female ratio of 1.5.

FN abnormalities consisted of an abnormal division in three nerves and an abnormal position in three nerves. The detailed FN abnormalities are summed-up in Table 1. FN malformation occurred in association with another inner ear malformation in four patients; they consisted of cochlear aplasia or hypoplasia, vestibular dilation, vestibular aqueduct dilation, posterior semicircular canal agenesis of rather total labyrinthine aplasia (Michel aplasia) Table 1.

We noted a hypoplasia of the IAC in four patients, associated to an agenesis or hypoplasia of the CN in all of them. On the other hand, we noted other associated abnormalities unrelated to the temporal bone in four patients. Three amongst our patients were asymptomatic, while two patients presented either a unilateral incomplete eyelid closure or a neonatal unilateral peripheral FN palsy at the 4th stage of the House and Brackmann classification (Table 2).

Two patients presented an abnormal FN division, consisting of a bifurcation of the mastoid segment (MS); it was bilateral in one of them. In the three abnormally bifurcated nerves, the MS was composed of an internal and external branch before reaching the stylo-mastoid foramen (SMF).

In the bilateral form of MS bifurcation, the two branches emerged from two different SMF and the corda tympani emerged from the external MS branch. The external branch had a more anterior position in comparison to the internal branch (Fig. 1). MRI revealed an absent and very thin CN on the right and left sides respectively. It also showed a hypoplasia of both IACs that measured 1 and 1.3 mm at the left and right sides respectively. Other clinical and radiological manifestations in this patient included: velo-palatal cleft, incomplete closure of the superior eyelids and a pyelocalyceal dilation.

In the unilateral form of FN bifurcation, we rather noted that both branches emerged from the same SMF and that the ipsilateral labyrinthine and tympanic segments of the

Table 1 Summary of the encountered facial nerve and inner ear malformations

Patient	Age	Sexe	Side	Facial nerve aberration	Inner ear
1	2 YA	M	Right	Bifurcation of the MS	None
			Left	Bifurcation of MS	
2	3 YA	F	Right	Anterior dislocation Geniculate ganglion hypoplasia	Vestibular aqueduct dilation Posterior semicircular canal agenesis
			Left	Geniculate ganglion hypoplasia	Cochlear hypoplasia type III (right) and type IV (left)
3	2 YA	F	Right	Anterior displacement of the LS Geniculate ganglion hypoplasia	Cochlear aplasia with a dilated vestibule
			Left	Geniculate ganglion hypoplasia	Cochlear aplasia with a dilated vestibule
4	3 YA	M	Right	Thinned diameter of LS and TS Bifurcation of the MS	Vestibular dilation
			Left	None	Vestibular dilation
5	4 YA	M	Right	Posterior displacement behind the middle ear	Total labyrinthine aplasia (Michel aplasia)
			Left	Absence of identification of the TS and LS FN passed directly from the IAC to the MS	Total labyrinthine aplasia (Michel aplasia)

YA: years old, F: female, M: male, MS: mastoid segment, TS: tympanic segment, LS: labyrinthine segment, FN: facial nerve, IAC: internal auditory canal

Table 2 Summary of the other associated malformations

Patient	Side	Internal auditory canal	Cochlear nerve	Middle and external ear	Other associated malformations
1	Right	Hypoplasia (1.3 mm)	Agenesis	None	Velo-palatal cleft incomplete closure of the superior eyelids and a Pyelocalyceal dilation
	Left	Hypoplasia (1 mm)	Hypoplasia 0.7 mm		
2	Right	None	None	External auditory canal atresia Agenesis of the long incudus crus	Velar cleft Bilateral Preauricular and cervical fistulas
	Left	None	None	Stapes malformation (left)	
3	Right	Hypoplasia (2 mm)	Agenesis	None	None
	Left	Hypoplasia (2 mm)	Agenesis		
4	Right	Hypoplasia (2 mm)	Agenesis	None	Cleft lip and palate Right peripheral facial nerve palsy Congenital nystagmus Spastic tetra paresis heterochromia MRI: Enlargement of the superior cerebral pedoncule and hypoplasia of the superior cerebellar vermis (Joubert syndrome)
	Left	Hypoplasia (2 mm)	Agenesis		
5	Right	Hypoplasia (1.3 mm)	Agenesis	None	Archanoid cyst at the left cerebellum measuring 43 × 30 mm
	Left	Hypoplasia (1.3 mm)	Agenesis		

FN were abnormally thinned. (Fig. 2) In this patient, CT scan also revealed a bilateral vestibular dilation and a total absence of communication between the IAC and the cochlea bilaterally. MRI showed an enlargement of the superior cerebral pedoncule, a hypoplasia of the superior cerebellar vermis, a bilateral IACs and a bilateral CVN agenesis. Clinically, the patient presented a cleft lip and palate, a right side peripheral facial nerve palsy, a congenital nystagmus, a spastic tetra paresis and a heterochromia. Clinical and radiological manifestations in were attributed to a Joubert syndrome.

Positional abnormalities were observed in three patients; they consisted of either a unilateral anterior dislocation in two of them or rather a unilateral posterior displacement. (Figures 3 and 4)

Anterior FN displacement was observed in two patients. In the first patient, it was associated to a bilateral geniculate ganglion hypoplasia, cochlear hypoplasia, dilation of

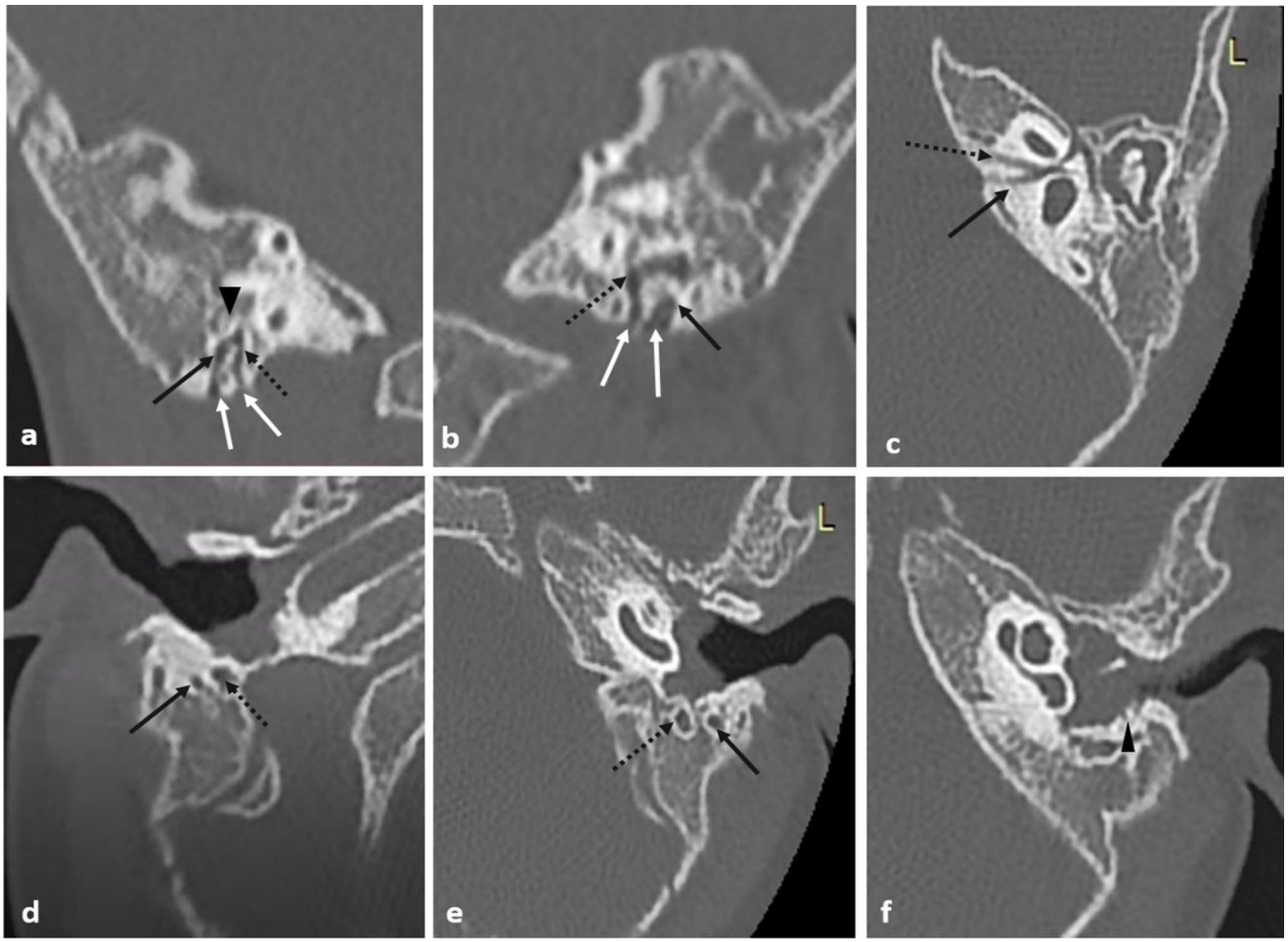

Fig. 1 Temporal bone CT scan of patient1: coronal (**a**, **b**) and axial (**c**–**f**) and slides showing: **a**–**e** Bifurcation of the mastoid segment of the right FN (**a**, **d**) and the left FN (**b**–**e**). We note the presence of an external branch (black arrow) and an internal branch (dashed black arrow) leaving the temporal bone from 2 different stylomastoid foramina (**a**, **b**: white arrows) **a**, **f**: the chorda tympani emerges from the external branch (arrow heads) **c** : Independent paths of the labyrinthine segment of FN (black arrow) and CVN (dashed black arrow)

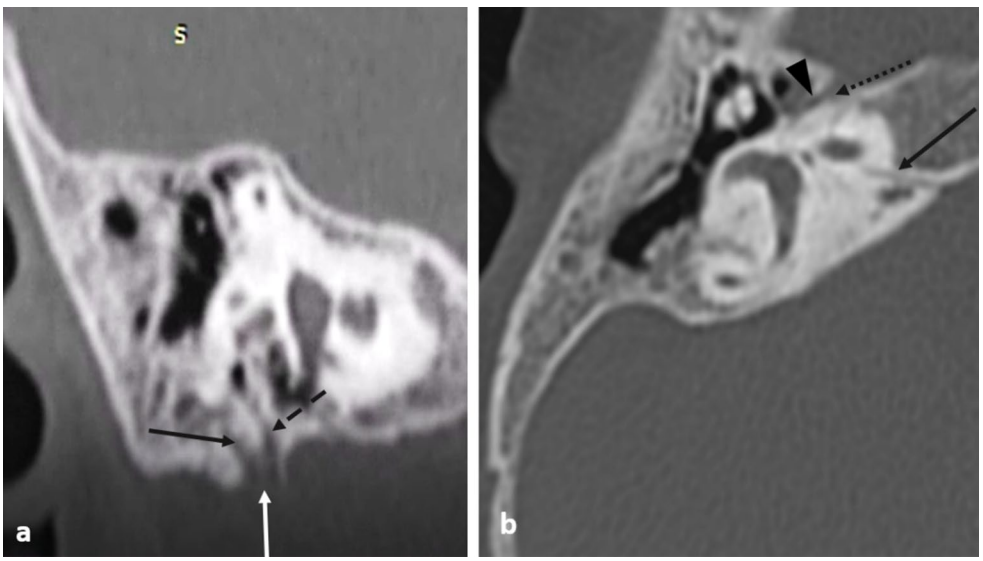

Fig. 2 CT scan of the right temporal bone (patient4), coronal (**a**) and axial (**b**) slides showing: **a** bifurcation of the mastoid segment of the FN with the presence of an external branch (dashed black arrow) and an internal branch (black arrow) that emerge from the same stylo-mastoid foramen (white arrow) **b** hypoplasia of the IAC (black arrow) with thinned labyrinthine (dashed black arrow) and tympanic (arrow head) segments of the FN

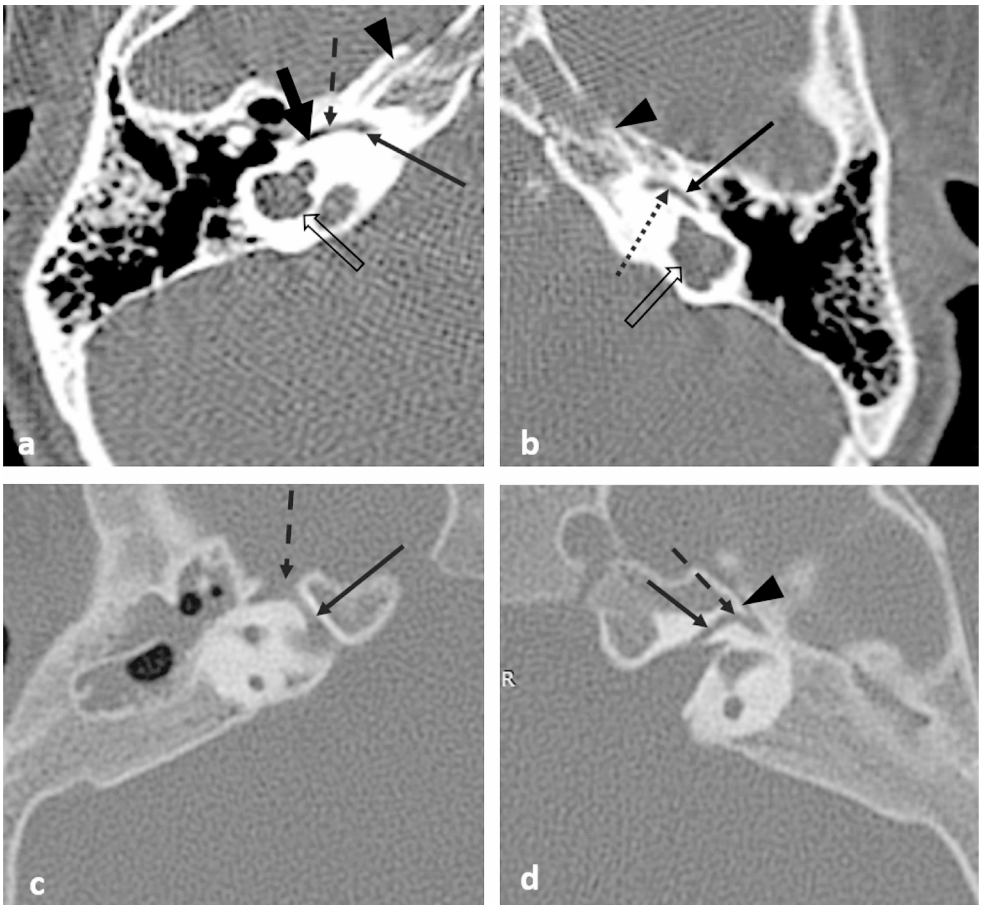

Fig. 3 temporal bone CT scan bone of patient2 (**a**, **b**) and patient3 (**c**, **d**),axial slides **a**, **b** Anterior dislocation of the right labyrinthine segment of the right FN (**a**: black arrow), with a bilateral hypoplasia of the geniculate ganglion (dashed black arrow), we note a bilateral cochlear aplasia with a dilated vestibule (empty arrows), the right tympanic segment (thick black arrow), and the greater petrosal nerve (arrow head). **c**, **d**: anterior dislocation of the labyrinthine segment of the right FN (**c**: black arrow), we note a hypoplasia of the geniculate ganglion (dashed black arrows). The left labyrinthine segment is represented by a black arrow and the greater petrosal nerve emerging from the tympanic segment of the FN is shown by an arrow head

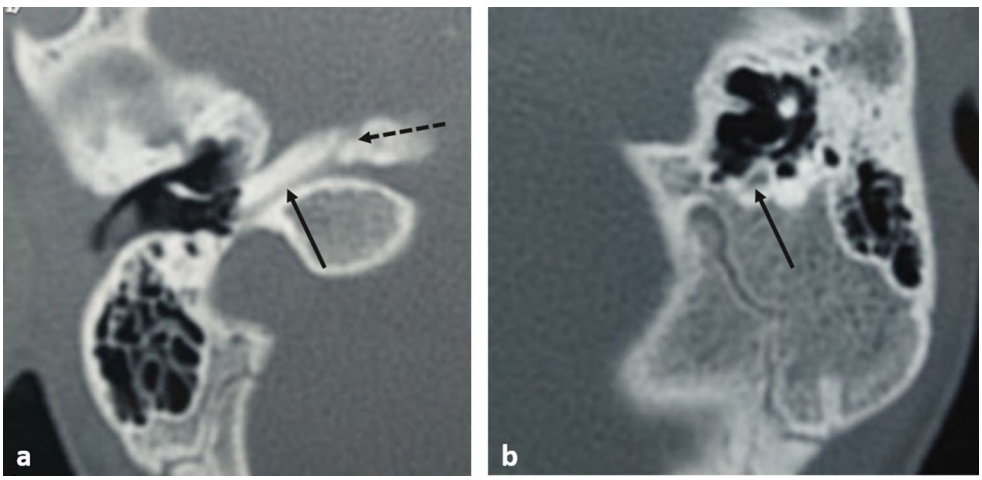

Fig. 4 Temporal bone CT scan of patient5, axial slides showing: **a**: at the right side: a posterior course of the labyrinthine segment of the FN (black arrow), the greater petrosal nerve is represented by the dashed black arrow. **b**: at the left side, the only visualized portion is the mastoid segment (black arrow)

the vestibular aqueduct and agenesis of the posterior semicircular canal. (Figs. 3) Other associated malformations consisted of a right external auditory canal atresia and a left stapes malformation with footplate thickening. Physical examination revealed the presence of preauricular and cervical fistulas bilaterally.

In the second case of anterior FN displacement, we observed a bilateral geniculate ganglion hypoplasia along with a bilateral hypoplasia of the IAC, cochlear aplasia and vestibular dilation. (Figs. 3) MRI confirmed a common hypoplasic VCN and the absence of the CN bilaterally. The clinical examination in this patient was also normal.

In the patient presenting a unilateral posterior FN displacement, the FN coursed posterior to the middle ear. The course of the contralateral FN was also abnormal; it passed directly from the IAC to the mastoid portion while the tympanic and labyrinthine segments were not identified. (Fig. 4) These FN abnormalities were associated to a Michel

Aplasia bilaterally. In addition, MRI revealed showed the absence of the VCN and a hypoplasia of the IAC bilaterally. Neither Cardiac nor ophtalmic and neurological abnormalities were observed in this patient.

Discussion

Malformations of the intratemporal FN are very rare occurrences that are important to recognize prior to any middle ear surgery. Due to its complex intratemporal course, any anatomical variation of the FN, especially when it is unrecognized pre-operatively, can put the nerve at a higher risk of injury during drilling at middle ear surgery leading to a post-operative morbidity [4, 5].

FN aberrations were seldom reported in association with otherwise normal temporal bones, with an incidence of 0.3% [1, 2]. They mainly occur along with other inner or middle ear malformations. Thus, a confirmed malformation of the outer, inner or middle ear should alert surgeons to the possibility of an associated FN aberration [12]. This association was reported to vary between 0.7 and 8.3% of cochlear implant cases [5, 7, 13–16]. In our study, the FN malformations were present 3% of our cochlear implant candidates.

The exact relationship between inner ear malformations and FN aberrations was explained by multiple embryological reasons. The FN derives from the otic capsule and the Reichert's cartilage from the second branchial arch. Hence, its malformations can be associated with an abnormal development of the first and second branchial arches structures including the bony canal of the FN, stapes, styloid process and external auditory canal. FN course is also determined by other surrounding temporal bone structures especially the inner ear, which is considered as the most important factor determining the FN final position [13]. On the other hand, a normally developed cochlea is suggested to prevent an abnormal migration of the FN [7, 12, 13]. Sennaroğlu et al. defined 6 groups of cochlear malformations having a higher risk of an aberrant FN: complete labyrinthine aplasia, rudimentary otocyst, cochlear aplasia, common cavity, cochlear hypoplasia and incomplete partition of the cochlea type III [7, 8, 17]. In our study cochlear malformations were reported in three patients consisting of either a cochlear aplasia or a cochlear hypoplasia.

CT scan is precise in determining aberrations of the FN course, associated middle and inner ear malformations and the relationship between the FN and adjacent temporal bone structures [5]. However, some aberrations can be misdiagnosed, as the presence of other temporal bone abnormalities may alter the standard landmarks making the determination of the exact course of the FN tougher [12]. Hence, CT scan can underestimate FN abnormalities leading to a possible intra-operative diagnosis of the FN malformation [12, 18]. In our patients, all cases were diagnosed through CT scan that was performed to explore congenital hearing loss.

Malformations of the FN involve any of its temporal bone segments. They concern its position, length and angles and can even consist in the presence of abnormal divisions of the nerve [19].

Divisional FN malformations consist of either a bifurcation or trifurcation. Bifurcation is defined as a focal splitting of the nerve at one or more of its segments [4]. In our study, we noted a bifurcation of the FN at the level of the MS in a total of 3 ears. Hoe et al. reported 3 cases (3.66%) associated to a middle ear malformation [5]. Its diagnosis is based on a temporal bone CT while few cases of an intraoperative discovery of a FN bifurcation were reported [5, 29, 30]. The causes of bifurcation are not known; It may be explained by an early bifurcation of the nerve at the embryological development [1, 26]. These abnormalities can be either asymptomatic or can manifest by a facial nerve palsy [22]. In our study, a peripheral facial nerve palsy was recorded in two out of the three FN bifrucations that we noted.

The bifurcation can affect all FN segments predominantly the tympanic segment (TS) at the anterior and posterior parts of the oval window [19, 31]. TS duplication can be associated to oval window atresia causing a fixation of the footplate and stapes [32]. LS bifurcation is rather very rare occurrence and was considered as one of the rarest malformations of the temporal bone. It was limited in the literature to case reports, occurring unilaterally in all reported cases [1, 2, 31]. It can be either associated to vestibular and cochlear malformations or a normal temporal bone [1, 32]. However, when a duplication occurs at this level, it was suggested that the second branch rather represents the nervus intermedius. But, in this situation, the two branches would have different diameters which was not the case of the reported duplicated LS [1, 2].

The bifurcation of the FN at its MS is also uncommon; it takes place just after the second genu of the FN at the beginning of the MS [19]. It manifests at CT scan as a bifid MS with two branches emerging from two different SMF [32]. Kalaiarasi et al. reported a 4% occurrence of FN bifurcation that intereseted the MS with two branches emerging from the SMF and the petrotymapnic fissure while the chorda tympani emerged from the lateral branch [4]. In our study, the two branches emerged either from the same SMF or from different ones. The corda tympani emerged from the lateral branch in two cases. The bifrucation of the MS was associated in two patients to an agenesis of the cochlear nerve. To our knowledge, no other similar associated has been previoulsy reported in the literature.

Positional abnormalities mainly consisted of anteromedial displacement of the FN; it represents the commonest

FN congenital malformation especially when occurring at the LS, the most affected segment [5, 12, 20]. The antero-medial displacement of the LS occurs in association to cochlear malformations mainly cochlear aplasia as it results in an anterior migration of the LS at the normal level of the cochlea [8, 11, 13, 20–22]. A delay in the rostral development of the cochlea and the absence of the cochlear duct development were suggested to cause an anterior FN migration [13, 23, 28]. Sennaroğlu et al. concluded that a normal sized-cochlea is associated with a normal LS while a hypoplasia of the middle and apical cochlear turns are rather associated with an anterior and superior displacement of the LS [3]. However, not all cochlear malformations were reported to be associated with an antero-medial displacement of the FN such as Modini malformation where the abnormal cochlear development rather occurs at a delayed timing [13]. In our study, we recorded an anterior dislocation of the FN in two cases having either a cochlear aplasia or a type III cochlear hypoplasia.

The LS can also have a posterior and inferior course mainly reported in cases of Michel aplasia [24, 25]. In our study, a posterior displacement of the LS was reported in a patient presenting a bilateral Michel aplasia.

The occurrence of positional abnormalities of the tympanic segment (TS) was less commonly reported. They are related to oval window atresia [2]. A superior displacement of the TS was described in association with semicircular canal decrease in size, hypoplasia and aplasia while an inferior migration was seen in association with a cochlear hypoplasia [3]. Positional abnormalities of the MS are rare and were reported in association with external canal atresia [13, 26, 27].

Other rare abnormalities include IAC hypoplasia and VCN malformations, either hypoplasia or aplasia. However, only few studies focused on the association between VCN, CN and FN malformations [9, 19]. A hypoplasia or aplasia of the FN is also a possible congenital abnormality of the FN, but it was seldom reported [33].

Middle ear surgery can be challenging in the case of a confirmed FN aberration. An aberrant FN course can change the choice of the surgical technique. A bifurcation of the FN or a displacement of its TS can limit the access to the round window during cochlear implantation especially in the case of an anteromedial displacement of the nerve since it can make the facial recess approach not possible and then increases risk of FN injury. Similar situations required a retrofacial approach in order to access the round window while other authors opted for a transcanal cochleostomy or a wall down mastoidectomy. [12,34] These aberrations can even cause the FN stimulation at the activation of the CI [12].

Conclusion

The anatomy of the facial nerve is complex and important. Pre-operative knowledge of the FN variations and congenital malformations is important prior to any middle ear surgery to avoid post-operative esthetical, functional and psychological complications related to an intra-operative injury to the facial nerve. The presence of any inner ear malformation must alert the surgeon to the possible presence of a FN malformation. The determination of the FN course is based on CT scan while MRI is mainly helpful in determining other associated malformations. FN malformations can change the surgical procedure and the surgical technique in order to access the round window.

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Declarations

Ethical Approval Ethical approval in not required in our institution for retrospective studies.

Consent to Publish Written informed consent was obtained from the patients' legal parents for their anonymized information to be published in this article.

Conflict of interest The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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