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REVIEW

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Risk factors and management strategies of inadvertent facial nerve stimulation in cochlear implant recipients: A systematic review

Asma Alahmadi MD[1] | Yassin Abdelsamad PhD[2] | Medhat Yousef MD[1,3] | Salman F Alhabib MD[1] | Afrah Alshalan MD[4] | Nezar Hamed MD[1] | Farid Alzhrani MD[1]

[1]King Abdullah Ear Specialist Center (KAESC), College of Medicine, King Saud University Medical City (KSUMC), King Saud University, Riyadh, Saudi Arabia

[2]Research Department, MED-EL GmbH, Riyadh, Saudi Arabia

[3]Audiology Unit, ENT Department, Menoufia University, Menoufia, Egypt

[4]Department of Otolaryngology-Head and Neck Surgery, College of Medicine, Jouf University, Sakaka, Saudi Arabia

Correspondence
Asma Alahmadi, King Abdullah Ear Specialist Center, College of Medicine, King Saud University, P.O. Box: 245, Riyadh 11411, Saudi Arabia.
Email: asma0alahmadi@yahoo.com

Abstract

Objectives: To systematically review the prevalence and risk factors of inadvertent facial nerve stimulation (FNS) after cochlear implant (CI) surgery. And to report the different management strategies used for reducing and resolving FNS.

Data Source: Web of Science, Scopus, PubMed, Cochrane Library, and Virtual Health Library (VHL) of the World Health Organization (WHO).

Review Methods: A systematic review was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) on studies that reported FNS as a complication after CI. A comprehensive electronic search strategy was used to identify the relevant articles. We extracted the data on the prevalence of FNS after CI activation, the reported grades, and the management strategies. The number of associated electrodes; cause of deafness; co-anomalies; and duration of hearing loss and their relationships with FNS were also studied.

Results: Twenty-one relevant articles were included in this review. The prevalence of FNS among the CI populations was 5.29% (175/3306 patients). Among those whose ages were reported, 58.3% (95/163) were adults, and 41.7% (68/163) were pediatrics. Modifying the different fitting parameters was the most used strategy, as it successfully resolved FNS in 85.5% of the patients (142/166). The second commonly used management strategy was surgical intervention (reimplantation or explantation), which was reported in seven studies for 23 patients.

Conclusion: FNS after CI activation could be controlled and resolved with many advances that range from readjusting the fitting parameters to surgical intervention. However, further studies are required to validate the efficacy of each management strategy and its impact on patients' performance. Our findings demonstrate that CI recipients with FNS could still benefit from the CI devices and their FNS could be controlled.

KEYWORDS
Cochlear implant, diagnosis, facial nerve stimulation, management, risk factors

1 | INTRODUCTION

Cochlear implant (CI) surgery has become the standard technique for treating patients with severe-to-profound deafness who receive little or no benefit from traditional acoustic amplification.[1] Although this surgery has many favorable audiological and speech outcomes, previous studies have revealed both major and minor complications. A potentially minor complication of CI is the inadvertent facial nerve stimulation (FNS).[2]

The prevalence of FNS significantly varies among patients undergoing CI; studies have reported prevalence ranging from 1% to 15%.[1,3–8] FNS is also accompanied by specific conditions such as cochlear ossification, inner ear malformation, temporal bone fractures, meningitis, and otosclerosis.[9] In this context, approximately 75% of patients with otosclerosis eventually develop FNS following CI.[5,7–11] However, they can still benefit from CI, and FNS can be controlled or minimized.[1,9,12,13]

According to Berrettini et al.,[1] FNS after CI is caused by mid-array electrodes in straight and perimodiolar electrode arrays. They attributed this to the proximity of the labyrinthine segment of the facial nerve to the superior segment of the cochlear basal turn.[1,14–16] Bigelow et al.[8] further reported that the delayed onset of facial nerve paralysis is owing to the pressure exerted by the electrode array on the bony separation between the scala tympani and the facial nerve. Other causes of FNS could also be related to the increased conductivity in the soft remodeled bone of early otosclerosis.[7]

The grading system for FNS post-CI was proposed by Kelsall et al.,[2] where grade I refers to no stimulation and grade VI is the total stimulation, that is, the severe gross motion of the total facial musculature and/or severe pain. Furthermore, patients with FNS may present with severe pain and/or severe gross motion of the facial musculature due to total stimulation.[7,8] Contrastingly, mild stimulation may cause patients to present with slight motion in the nasolabial, mouth, eye, or forehead regions.[2] However, the practical application of this grading system might be challenging because of subjective interobserver variability in reporting grades.

FNS has a variable time of emergence; it may be immediate or may take up to 10 years after the CI surgery.[1,4,17] The diagnosis of FNS following CI surgery is difficult because it mainly relies on patient self-report.[9] Additionally, identifying pediatric cases is usually tricky, and these cases remain unreported or misdiagnosed.[18] In addition, the course of the condition is not steady, and its severity usually increases over time.[9]

Although FNS is difficult to diagnose and has a controversial etiology, it can be easily and effectively managed.[8] Several methods have been developed to reduce the incidence and severity of FNS after CI. Muckle and Levine (1994) suggested that the maturation of otosclerosis is signified by the use of fluoride, which reduces FNS.[11] Various reprogramming strategies are useful for managing FNS in CI recipients. Polak et al.[19] reported some of these strategies, such as switching off the offending electrodes, keeping the stimulus levels lower than the FNS threshold levels, and changing the fitting strategies. However, this may deteriorate patient outcomes.[17]

Consequently, changes in the fitting parameters of speech processors can be tried to resolve FNS.[1] However, it should be pointed out that these reprogramming approaches may not be practical in some cases that require reimplantation.[19]

Various studies have reported the risk factors, management approaches, and outcomes related to FNS after CI. However, their findings were inconsistent, and the prevalence of FNS was not clearly reported. In addition, there is a lack of standard practices and FNS management protocols that can be generated from current pools of evidence, and no established success rates have been reported for different management strategies.

Therefore, this systematic review was primarily conducted to report the prevalence and risk factors of FNS after CI. Furthermore, we aimed to provide comprehensive evidence, from the relevant studies in the literature, on the reported management options of FNS.

2 | METHODS

2.1 | Outcomes of interest

The main outcome of interest of the present systematic review was a discussion of the different approaches to managing CI-induced FNS that were reported in the current literature. Moreover, we aimed to identify the potential risk factors and follow-up outcomes. This might help the CI professionals prepare policies to enhance prognosis, prevention, and/or management of the FNS.

2.2 | Search strategy

This study was conducted in accordance with the recommendations of the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA). First, in November 2021, we identified all relevant articles, using a comprehensive online search strategy using relevant keywords based on our intended outcomes. The following databases were used to perform the search: Web of Science, Scopus, PubMed, Cochrane Library, and Virtual Health Library (VHL) of the World Health Organization (WHO). Then, we performed a manual search of the reference lists of relevant reviews and other related resources in different databases to identify potentially relevant studies.

2.3 | Inclusion and exclusion criteria

Based on the aforementioned outcomes, we included the studies that reported: (i) FNS as a complication after CI surgery; (ii) management of FNS; and (iii) diagnostic methods. All included studies were original studies, case reports, case series, and studies on human research with no restrictions on the country of research or year of publication. We excluded the following studies: (i) those that did not report sufficient data regarding diagnostic tools or management of CI-induced FNS; (ii) non-original studies (including poster publications, commentaries, letters, review articles, thesis, conference, book chapters); (iii) studies without available full-text; (iv) any papers with overlapping datasets; and (v) those that were not in English.

2.4 | Screening of relevant articles

After conducting the search using different databases, all relevant citations were imported into Endnote X9 to identify and omit all duplicates. Subsequently, all relevant articles were imported into a prepared Excel sheet that included the titles, DOIs, author lists, abstracts, journal names, and URLs of each citation. Each citation was also assigned an ID for easy identification and to prevent overlap. The screening strategy was performed in two steps: title/abstract screening and full-text screening. Both screening procedures were conducted by at least two authors under the supervision of the senior author, who resolved differences, if any, between the two authors via group discussions. Finally, all relevant articles were identified and prepared for data extraction.

2.5 | Data extraction

Before initiating the data extraction process, the senior author prepared a pilot form for data extraction based on the outcomes of interest. The form was agreed upon by all researchers and comprised three main parts: (i) baseline characteristics of participants in included studies (sample size, age, and sex), study design, and citation details, that is, last author's name, year of publication; (ii) outcomes of interest of the included studies, such as risk factors of CI-induced FNS, management processes, diagnostic approaches, and follow-up outcomes; and (iii) quality assessment. Data extraction of each included study was performed by at least two researchers who were blinded to the results, and the data were then compared publicly to identify differences and to reach a final decision about the included studies.

2.6 | Quality assessment

The CARE checklist for case studies and the National Institutes of Health Quality Assessment Tool for observational studies were used to assess the quality of the included studies. The domains of these tools were imported into the third part of the extraction sheet, and assessments were performed using a process similar to data extraction.

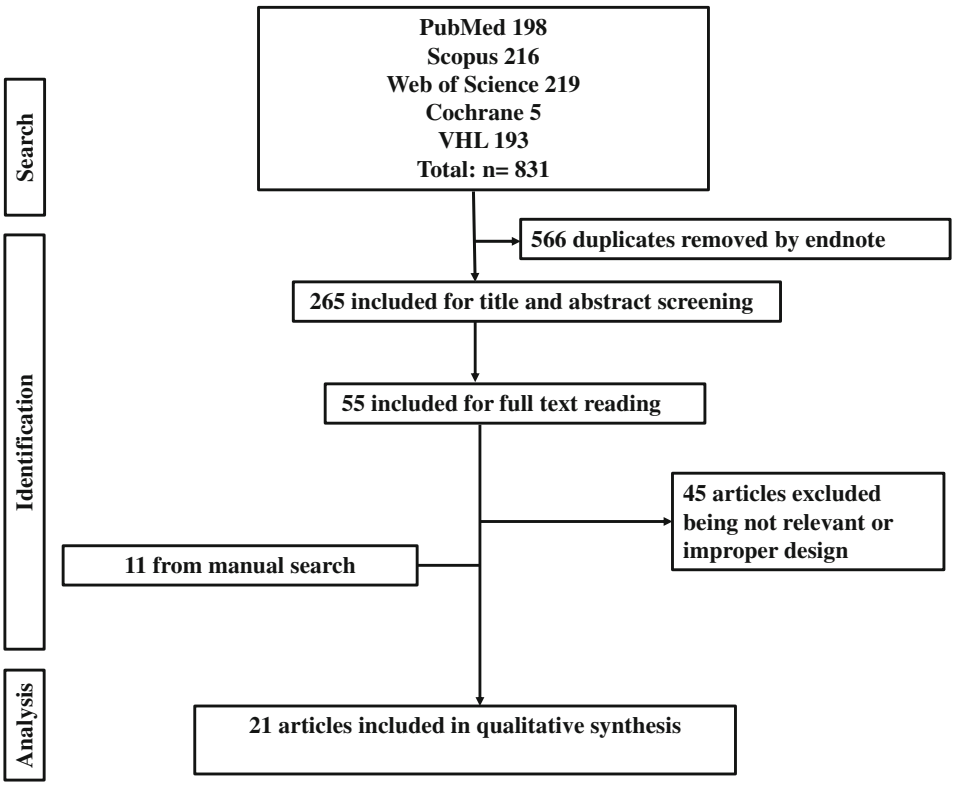

FIGURE 1 Flowchart of study selection and screening process.

TABLE 1 Baseline characteristics of the studies and their populations included in this review.

Author/year	Reference number	Country of residence of the patients	Study design	Follow-up period (months)	Sample size (*N*)	Patients with FNS after CI, *N* (%)	Age (years), mean (SD)	Sex (male), *N* (%)	CI Device (Manufacturer, Model or Electrode Type)	QA score
Alhabib et al., 2021	35	Saudi Arabia	ROS	17.5 (5–24)	16	16	2–56	NR	MED-EL, NR	10
Alharbi et al. 2012	33	Germany	Case report	NR		1	75	1	Cochlear, Nucleus 24 Contour Advance	10
Alzhrani et al., 2021	27	Saudi Arabia	ROS	NR	1100	32	Adults, 33 (12.6); pediatrics, 4 (2.2)	NR	MED-EL, (15 ears) SYNCHRONY (10 ears) CONCERTO (6 ears) SYNCHRONY-P	8
Atanasova-Koch et al., 2021	12	Germany	ROS	NR	287	4	31–83	NR	NR	8
Battmer et al., 2006	32	Germany	ROS	12	4	4	35–72	NR	Cochlear, Nucleus Mini22 device with a Nucleus Straight electrode	7
Berrettini et al., 2011	1	Italy	ROS	>12	119	11	41 (27 m-67 years)	NR	Cochlear, Nucleus 24 Ci	8
Braun et al., 2019	9	Germany	ROS	NR	19	19	44.68 (21.93)	6 (40)	MED-EL, (8) Concerto, (1) PULSARci100, (3) SONATAti100, (2) Synchrony, (1) CMD Concerto	7
Chen et al., 2021	40	China	Case report	NR	1	1	36	1 (100)	MED-EL, Flex28	10
Fang et al. 2017	36	USA	Case report	36	3	2	81, 34	2 (100)	Cochlear, Nucleus Freedom device	11
Fernandez-Vega et al. 2008	13	USA	ROS	8	2	2	60, 64	2 (100)	Cochlear, Nucleus 22 devices	8
Gold et al., 1998	20	USA	Case report	–	2	2	73, 51	1 (50)	Cochlear, Nucleus 22 devices	9
Nassiri et al., 2018	29	USA	ROS	–	14	5	18.2 (0.6–55)	2 (40)	(1) Cochlear, CI24M double array, (2) AAB HiRes Ultra with HiFocus, (1) Cochlear Nucleus 5 Contour	7
Niparko et al., 1991	3	USA	ROS	6–24	82	12	NR	NR	(6) Multiple-channel implants, (6) Single-channel electrodes (3MNlennadevices)	8
Ozkan et al., 2021	31	Turkey	Case report	96	1	1	4	1 (100)	MED-EL, FORM 24 prototype electrode	10
Pires et al., 2018	6	Portugal	ROS	24	448	13	20–78	8 (61.5)	NR	8
Polak et al., 2006	19	USA	Case report	15	2	2	60, 64	2 (100)	Cochlear, Nucleus 22 devices	10
Rah et al., 2016	37	Korea	ROS	–	881	32	3 (1–7)	12 (37.5)	NR	8
Rath et al., 2022	38	USA	Case report	NR	1	1	2	1 Female	Advanced Bionics, HiRes90K with the HiFocus 1j electrode on the right ear and advanced Bionics HiRes90K with HiFocus Mid-Scala electrode in the left ear	11
Rayner et al., 2003	5	USA	ROS	6	147	12	(41–75)	5 (41.6)	(11) Cochlear, Nucleus 22; (1) Advanced Bionics, Clarion 1.2	7
Sefien et al., 2019	17	Egypt	ROS	>13	175	2	11, 6	2 (100)	(1) MED-EL, (Sonata TI100 titanium implant footprint with Form 24 electrode); (1) Advanced Bionics, (HiFocus midscala electrode)	8
Wester et al., 2016	39	USA	Case report	NR	1	1	9	1 (100)	21-electrode device	11

Abbreviations: FNS, facial nerve stimulation; ROS, retrospective cohort review; QA score, quality assessment; NR, not reported.

3 | RESULTS

3.1 | Search results

A total of 831 articles were identified using the electronic search strategy. Endnote identified 566 duplicate articles, leaving 265 articles for the screening process. After title/abstract screening, 55 articles were eligible for full-text screening. Furthermore, 11 articles were identified through the manual search. After the full-text screening of all these articles, we included 21 studies that met the inclusion criteria. A PRISMA flowchart of this process is presented in Figure 1.

3.2 | Baseline characteristics of included studies

A total of 21 relevant articles were included in the present systematic review. These studies were published between 1991 and 2021. Most studies ($n = 13$) were retrospective studies, whereas the remainder ($n = 8$) were case reports. Some of the included studies reported only FNS cases, whereas others estimated the prevalence of this condition within the study. The total population of included studies was 3306 patients, and the prevalence of FNS in this population was 5.29% (175/3306). The follow-up period also varied among the included studies, ranging from 5 to 96 months. The total quality assessment scores for most of the retrospective observational studies and case reports were 8 and 10, respectively. Table 1 illustrates the detailed characteristics of each included study and other variables, including age, sex, device type, and quality assessment.

Of the 21 included studies, one did not provide data on the age groups of the patients who were reported to have FNS.[3] In the remaining studies, which reported a total number of 163 cases of FNS, 68/163 (41.7%) were pediatric cases and 95/163 (58.3%) were adults. The details of these studies in terms of reported age groups (adults vs. pediatric patients) are presented in Figure 2.

3.3 | Characteristics and risk factors of FNS

Different risk factors for developing CI-induced FNS have been reported in the literature, also the time of onset of FNS after CI device activation (in months), FNS grade, number of electrodes associated with FNS, cause of deafness, co-anomalies, and duration of hearing loss. Table 2 summarizes the characteristics and the risk factors reported by each study included in this systematic review. For instance, the onset of FNS after CI activation varied, ranging from immediate onset to up to 48 months post-activation. Moreover, different comorbidities have been reported as risk factors for the development of FNS post-CI: meningitis, osteosclerosis, cochlear facial dehiscence, narrow bony cochlear nerve canal, and labyrinthitis ossificans.

3.4 | Management strategies and outcomes

Numerous FNS management strategies have been reported in the reviewed studies, such as audio processor reprogramming, device reimplantation, and explantation. Twenty studies reported reprogramming as a primary and major management strategy approach in 166 patients. Among them, 142 (85.5%) had FNS, which completely resolved after readjusting the fitting parameters, while the rest ($n = 24$, 14.5%) had recurrent FNS; details of these studies are presented in Supplementary Table S1. Moreover, some studies ($n = 13$)

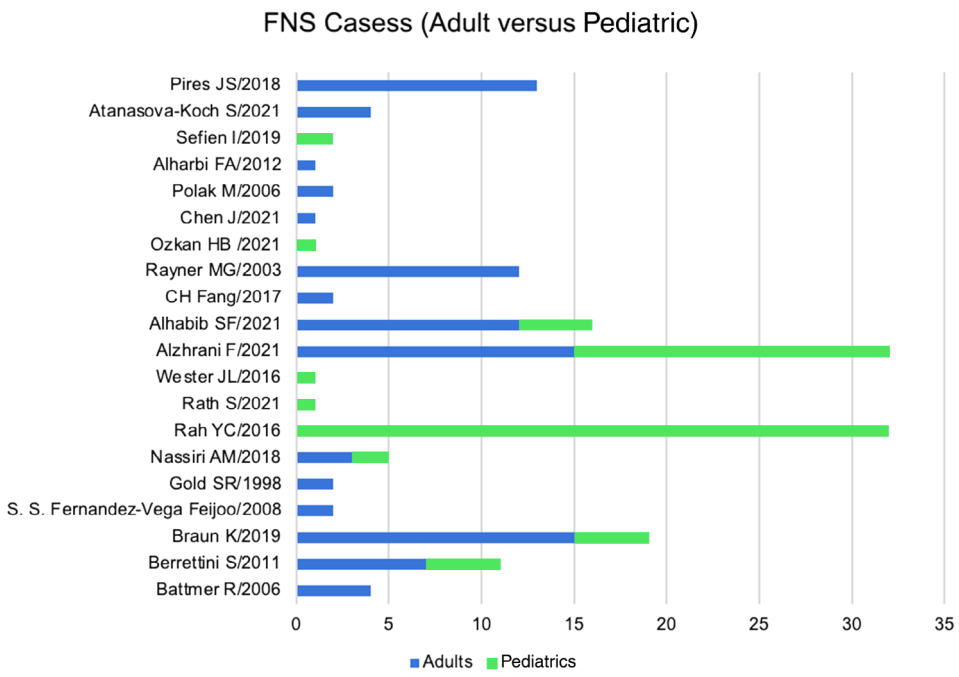

FIGURE 2 Characterization of FNS according to adult and pediatric patient groups. FNS, facial nerve stimulation.

TABLE 2 Characteristics and risk factors of the occurrence of cochlear implant-related facial nerve stimulation.

Author/year	Reference Number	Time of onset of FNS after CI activation (in months)	Grade of FNS	No. of electrodes associated with FNS	Cause of deafness	Co-anomalies	Duration of hearing loss	Others
Alhabib et al., 2021	35	6 Immediate 10 within (1–6 months)	–	6	–	–	–	3 Ears showed partially inserted electrodes
Alharbi et al. 2012	33	1 After 48 months	–	–	–	–	–	–
Alzhrani et al., 2021	27	9 Immediate 23 within 16 months	(8 ears) Grade 2 (13 ears) Grade 3 (1 ear) Grade 4 (7 ears) Grade 5 (3 ears) Grade 6	8 Electrodes cause mild stimulation, 13 electrodes cause moderate stimulation, 1 electrode causes moderate to severe stimulation, 7 electrodes cause severe stimulation	2 Sudden SNHL, 21 congenital, 7 progressives	–	–	–
Atanasova-Koch et al., 2021	12	–	4 Grade 2	–	–	3 Otosclerosis	–	–
Battmer et al., 2006	32	–	–	8	4 Osteomyelitis	3 Otosclerosis	1–25 years	1 Partial insertion due to fibrosed tissue inside the cochlea
Berrettini et al., 2011	1	4 Immediate, 1 within the 12 months, 6 after 12 months	–	–	4 Inner ear anomalies, 1 Central Nervous System Superficial Siderosis (CnSSS), 2 genetic deafness, 1 idiopathic, 1 post-high fever deafness	1 Malformed cochlea with hypoplasia of the cochlear nerve, 2 cochlear otosclerosis, 1 cochlear post-meningitis ossification	–	–
Braun et al., 2019	9	–	15 Severe	–	2 Otosclerosis, 5 congenital hearing loss, 6 Unknown, 1 temporal bone fracture, 1 sudden hearing loss	2 Otosclerosis, 5 congenital hearing loss	–	–
Chen et al., 2021	40	–	1 Very severe	–	–	1 Cochlear–facial dehiscence	–	–

TABLE 2 (Continued)

Author/year	Reference Number	Time of onset of FNS after CI activation (in months)	Grade of FNS	No. of electrodes associated with FNS	Cause of deafness	Co-anomalies	Duration of hearing loss	Others
Fang et al. 2017	36	1 Immediate, 1 1 month	–	5	2 SNHL	2 Cochlear-facial dehiscence	–	–
Fernandez-Vega et al. 2008	13	1 After 36 month, 1 after 24 months	–	1, 2	2 Otosclerosis	2 Otosclerosis	–	–
Gold et al., 1998	20	1 After 0.5 month, 1 after 30 months	–	–	1 Progressive hearing loss, 1 sensorineural	1 Otosclerosis	–	–
Nassiri et al., 2018	29	–	–	–	2 Meningitis, 1 sudden SNHL, 1 prematurity	4 Labyrinthitis ossificans	5.5 months (2–17 months)	–
Niparko et al., 1991	3	2 Immediate, 4 within 3 months, 6 within 18 months	–	–	–	–	–	–
Ozkan et al., 2021	31	1 After 6 m	–	–	–	1 Common cavity	–	–
Pires et al., 2018	6	12 Immediate, 1 within the first 12 month	–	–	5 Otosclerosis, 1 Meniere's disease, 2 head traumas, 5 idiopathic	–	–	–
Polak et al., 2006	19	1 After 36 m	–	2	2 SNHL	1 Otosclerosis	–	–
Rah et al., 2016	37	15 Immediate, 17 late onsets	–	–	–	23 Narrow bony cochlear nerve canals	–	–
Rath et al., 2022	38	1 After 21 months	–	–	–	–	–	–
Rayner et al., 2003	5	–	–	–	–	7 Otosclerosis	–	–
Sefien et al., 2019	17	4, 15 months	–	–	1 Meningitis, 1 congenital hearing loss	–	–	–
Wester et al., 2016	39	1 After 48 months	–	3	–	–	–	–

Abbreviations: CI, cochlear implant; FNS, facial nerve stimulation; SNHL, sensory neuronal hearing loss; –, not reported.

have reported hearing outcomes in patients ($n = 107$); 45.8% (49/107) had poor hearing outcomes (Table 3). Reprogramming of patients' audio processors is performed by combining many fitting parameters or by using only one.

As shown in Table 3, the success rate and frequency of use of the different reprogramming techniques varied. For instance, a triphasic pulse has been used in 32 patients in three studies, with a success rate of 84.4% in resolving the FNS and good outcomes in 100% of the patients. Electrode deactivation was reported in seven studies, which helped in resolving the FNS in 90.9% of the patients (20/22) and good outcomes in 77.3%. Stimulation Levels were changed in 19 patients in three studies, with a success rate of 73.7% in minimizing FNS. However, the majority of the patients (68.4%) showed poor outcomes with this technique alone. The term reprogramming was used in two studies without including a methodical description, nevertheless, good outcomes were reported with a success rate of 100%.

Combining more than one fitting technique has been reported in many studies to completely resolve FNS with a success rate of 100% and good patient outcomes. As shown in Table 3, examples of these combinations are as follows: (i) triphasic pulse stimulation + electrode deactivation; (ii) electrode deactivation + oral fluorocal; (iii) electrode deactivation + readjusting the stimulation levels and thresholds + pulse width; and (iv) strategy + phase duration. In contrast, some combinations, such as (i) stimulation levels + electrode deactivation + biphasic + stimulation mode + phase duration; (ii) biphasic + stimulation mode + electrode deactivation; and (iii) pulse width + stimulation mode, neither resolved FNS nor demonstrated good outcomes. In one study, the use of combined techniques, including triphasic pulse, changing the pulse width, and changing the coding strategy resolved FNS but showed poor outcomes. The use of different fitting strategies has resulted in poor outcomes in most patients in some studies; however, it proved successful in resolving FNS in most cases. For instance, with the following combined techniques, (i) stimulation level + electrode deactivation, (ii) stimulation level + pulse width, and (iii) stimulation level + pulse width + electrode deactivation, the success rates were 100%, 70%, and 50%, respectively. Reducing the gain was effective in one study and changing the coding strategy was effective in another.

Regarding surgical explantation and reimplantation, seven studies reported that 12 patients in all of their FNS populations ($n = 23$, 52.17%) underwent reimplantation after failed reprogramming. Four of these studies reported that hearing outcomes improved in 5/6 patients (83.33%), while only one patient (16.67%) had poor hearing outcomes. In addition, FNS was resolved in all 12 patients. Gold et al.[20] reported that their management approach for FNS post-CI was with the application of oral Fluorocal course in addition to reprogramming. In the two included patients, hearing outcomes significantly improved and there was no evidence of FNS, except when the modality was stopped. These variables are listed in Table 4.

4 | DISCUSSION

FNS remains one of the expected complications of CI; it is bothersome to the affected patients and negatively affects their quality of

TABLE 3 Reported reprogramming strategies and their outcomes.

	Numbers		FNS		Outcomes	
Technique	Studies	Patients	Resolved	Failed	Good	Poor
Triphasic pulse	3	32	27	5	32	0
Electrode deactivation	7	22	20	2	17	1[a]
Stimulation levels	3	19	14	5	6	13
Reprograming	2	14	14	0	14	0
Stimulation levels + electrode deactivation	3	10	10	0	3	7
Stimulation levels + pulse width	1	10	7	3	0	10
Stimulation levels + pulse width + electrode deactivation	1	7	5	2	0	7
Triphasic + electrode deactivation	1	3	3	0	3	0
Electrode eactivation + oral fluorocal	1	2	2	0	2	0
Reduce gain	1	2	2	0	–	–
Stimulation levels + electrode deactivation + biphasic + stimulation mode + phase duration	1	1	0	1	0	1
Biphasic + stimulation mode + electrode deactivation	1	1	0	1	0	1
Electrode deactivation + Stimulation levels and thresholds + pulse width	1	1	1	0	1	0
Pulse width + stimulation mode	1	1	1	0	0	1
Strategy	1	1	1	0	1	0
Strategy + phase duration	1	1	1	0	1	0
Triphasic + pulse width + strategy	1	1	1	0	0	1

[a]Four not reported.
Abbreviation: –, Not reported.

TABLE 4 Surgical explantation or reimplantation. and other approaches and hearing and FNS outcomes per each study.

		Management		Outcome after management		
Author/year	Reference Number	Surgical explantation or reimplantation	Other	Hearing outcome	FNS incidence outcome	Outcome at last follow-up
Alharbi et al., 2012	33	1 Reimplantation with Nucleus CI512 and reprogramming after failed reprogramming of the first implant	–	1 Improvement in hearing tests with a score of perception of 81% in the German HSM Sentence Test in quiet	1 FNS was reported after reimplantation and disappeared after modified reprogramming	1 (100) no FNS incidence
Battmer et al., 2006	32	4 Reimplantation with a Nucleus24R and a Contour electrode	–	–	4 No FNS was observed even with the highest stimulation levels of all electrodes	–
Fernandez-Vega et al., 2008	13	2 reimplantation with a Nucleus 24 Nucleus 24 Contour and Corp after failed reprogramming	–	2 immediate improvements in speech discrimination tests	2 No FNS was observed	2 (100) improvements in hearing tests with the new re-implanted device with no FNS incidence
Gold et al., 1998	20	–	2 Oral Fluorocal courses with reprogramming	2 Improvements in hearing tests after failed reprogramming	2 1 No FNS was observed except when stopped	–
Niparko et al., 1991	3	1 Reimplantation with Nucleus multiple-channel device after failed reprogramming	–	–	1 No FNS was observed	–
Ozkan et al., 2021	31	1 Reimplantation after failed programming	–	1 Poor hearing tests outcome	1 No FNS	–
Polak et al., 2006	19	2 Reimplantation with a Nucleus 24 straight array and reprogramming after failed reprogramming of the first implant	–	2 Improvements in hearing tests	2 No FNS was observed	2 Improvements in hearing tests with no FNS incidence
Rath et el., 2021	38	1 Reimplantation after failed reprogramming	–	–	1 No FNS was observed	–

Abbreviation: –, Not reported.

life. Previous studies reported the prevalence of FNS to vary widely between 1.1% and 15% among the CI patients.[4,8] The present review found a prevalence rate of 5.29% for FNS cases. Our review investigated both adult and pediatric patients to determine the management techniques adopted for FNS. Previous studies reported that the incidence of FNS in children is much lower than that in adults, calculated to be 3% by Kempf et al.[21] and 0.9% by Hoffman and Cohen.[22] According to the findings of the present review, the pediatrics and adults represented 41.7% and 58.3%, respectively of the total reported FNS cases. However, the incidence of FNS in adults and pediatric patients was not reported accurately by some of the included studies, as they segregated patients with FNS post-CI according to certain risk factors, which does not reflect the actual incidence of FNS among all CI patients.

Our study shows that the incidence of FNS in pediatric patients has been underestimated. There are several reasons for that; First, the identification of FNS in the majority of CI recipients is evaluated visually by the clinician or must be self-reported by the patients.[18] Another reason is the lack of objective measures to detect FNS post-CI automatically.

Previous studies have indicated that the rate of FNS after CI in patients with otosclerosis is much higher, ranging from 25% to 75%.[11,23] Nine of the 21 included studies identified osteosclerosis as a key risk factor for FNS in patients with CI.

FNS management can be performed by adjusting the audio processor fitting parameters, such as reducing the stimulation levels, changing the phase duration, and deactivating the electrodes that stimulate the FN.[24] In this systematic review, we discuss the management approaches and outcomes of CI-related FNS based on evidence from the literature. Our current evidence shows that reprogramming is usually effective in resolving FNS, as reported in >85% of involved cases. Surgical approaches were used in patients with failed attempts of reprogramming, and they were associated with good hearing and FNS outcomes.

Triphasic pulse stimulation and electrode deactivation were associated with stable and improved outcomes in controlling FNS and hearing. Triphasic pulse stimulation has also been suggested to be more favorable for patients with labyrinthitis ossificans. Triphasic stimulation pulse pattern (TPP) may be most effective in patients with higher-grade FNS.[9,25,26] It can decrease the impact and development of the FNS by distributing charge across one positive phase and two negative phases with double and same durations, respectively.[9] However, it should be noted that this approach when combined with other reprogramming techniques might be associated with a reduction in hearing abilities. This can be attributed to changing the coding strategy, changing the phase duration, or deactivating the channels, which can eventually lead to less-than-optimal fitting maps. However, another study by Alzhrani et al.[27] indicated that TPP was associated with favorable hearing outcomes. These differences may be attributed to the differences in population characteristics. Other studies showed that Bipolar pulse can be also beneficial for controlling FNS.[2,3] Therefore, further studies are warranted for additional clarification of the effect of different pulse patterns.

The exact pathophysiological mechanism of the occurrence of FNS following CI surgeries is not adequately comprehended in the current literature. However, evidence shows that the association between the facial nerve and the proximity of the electrodes might explain this phenomenon.[14–16,28] Previous studies have also suggested that the FNS is most likely affected by the upper basal turn electrodes, which are most proximal to the labyrinthine segment of the facial nerve,[2,28] and this was demonstrated in a case series by Nassiri et al.[29] The authors observed that the basal turn electrodes were most proximal to the facial nerve on postoperative imaging and consequently demonstrated that the incidence of FNS was remarkably reduced after reducing stimulation levels and basal electrode deactivation.

It should also be noted that the current data suggest that the FNS is associated with different electrode locations within the cochlea, not only with those within the basal turn. Based on these findings, current evidence suggests that some cases may require electrode deactivation within different regions of the cochlea. Therefore, the proximity of the electrodes to the facial nerve might not be the only factor associated with FNS. In patients with labyrinthitis ossificans, shifting of the electrical current may also be influenced by fibrosis and ossification, which may cause FNS in different regions, not only at the basal turn. Previous studies have also implied an association between FNS and intracochlear impedance patterns.[3,30] Notably, Ozkan et al.[31] reported that, even after the deactivation of different electrodes, FNS was still observed in a patient who experienced auditory deprivation secondary to these management approaches. As reported in some studies, combinations of more than one management strategy have been used to control FNS. The impact of these combinations on the patients' performance showed favorable outcomes. However, they require further validation, owing to the small number of included studies that recruited a small number of patients.

Additionally, the majority of authors who conducted surgical approaches for their patients did so after the failure of reprogramming. Battmer et al.[32] reported that none of their patients developed FNS, regardless of the stimulation levels of the electrodes after reimplantation, with no evidence of reprogramming, indicating a high success rate for these surgical approaches without the occurrence of FNS. Nevertheless, the surgical approach options are unfavorable as they have subsequent adverse effects and may require revision surgeries. Furthermore, the effect of these approaches on hearing outcomes could not be precisely estimated because of the small number of patients.

It is important to further assess if reimplantion has better outcomes compared to the reprogramming strategies. Battmer et al.[32] considered reimplantation for their patients, as many electrodes[8] were associated with FNS and some patients had otosclerosis. In another study, patients who preferred re-implantion were found to have otosclerosis.[13] Another factor that supported reimplantation was the failure of re-programming in reducing FNS.[31,33]

The appropriate selection of electrode array was reported as an important step for patients undergoing CI and enhancing their outcomes. According to Battemer et al.,[32] using perimodiolar electrode with contacts facing the modiolus is more often associated with

reduced FNS than using a straight electrode.[32] In this context, Jaekel et al.[34] compared the two types of electrodes and found that none of the patients for whom the perimodiolar electrode was used had FNS. Furthermore, they found that 4/6 patients in whom the straight electrode was used had FNS. However, some cases of FNS were reported with the usage of perimodiolar electrodes. Berrettini et al.[1] reported two cases of FNS in two patients with otosclerosis, one of them was implanted with a perimodiolar electrode and developed the FNS immediately, and the other case had a straight electrode and exhibited a delayed and more gradual onset FNS. Moreover, a study by Ahn et al.[24] showed that, in a normal cochlea, no difference in the prevalence of FNS between straight and perimodiolar electrode arrays. Therefore, the current review reveals that FNS might occur with different types of devices (Table 1). However, further studies are required to better understand the effects and influence of different types of electrode design on the FNS.

Our findings mainly discuss the three interventions adopted by the included studies to control FNS post-CI: reprogramming, surgical explantation, and re-implantation. Of these measures, 15 studies resorted to a management strategy of deactivating the electrodes causing the FNS. A previous study presented this view and claimed that the deactivation of electrodes is necessary when it causes non-auditory stimulation, malfunction, or incomplete insertion.[6] However, our findings further show that deactivation of the electrode may not be the optimum solution in some cases with congenital hearing loss, failed programming, multiple electrodes associated with FNS, or severe FNS, as it does not address the main cause of FNS and it can affect hearing.[1,9,17,27] One of the important findings of our review is that the hearing outcomes were significantly improved in the studies in which management techniques such as changing the pattern of stimulation pulse, re-implantations, oral Fluorocal course, and reprogramming.

Finally, we encourage clinicians to consider the different risk factors for the occurrence of FNS after the CI surgery, owing to the difficulty in arriving at the diagnosis and its late presentation, thereby improving the possibility of early diagnosis and improved management and prognostic outcomes.

The present review has certain limitations. First, data from the included studies are based on retrospective data or single case reports, and none of them had a control group, nor did they randomize patients to different intervention groups to enable the evaluation of the effect of available interventions. Furthermore, detailed population characteristics, etiology of FNS, demographics, interventions, specific reprogramming techniques, and device type were not clearly mentioned in many of the included studies, representing significant heterogeneity among these studies. Future research, in which complication of FNS post-CI is uniformly addressed, may indicate whether the factors found in our review are true risk factors.

5 | CONCLUSION

The present systematic review identified different risk factors for the development of CI-induced FNS, including the onset, grade, and associated number of electrodes. Our results also suggest that the incidence of FNS in pediatric CI patients with CI is not significantly lower than that in adults and requires due attention. Hence, regular follow-ups are recommended for the identification of FNS in children. In addition, there is a wide range of management approaches for FNS without affecting patients' performance. Readjusting the fitting parameters, deactivating the associated electrodes if they are in low numbers, and switching to a different pattern of stimulation are recommended before considering any further surgical interventions. Reporting the audiological and speech outcomes of managed patients is highly recommended for future research to accurately determine the accurate success rate of the available management approaches. This review sets the foundation for future studies that aim to characterize management strategies of CI-induced FNS to achieve favorable patient outcomes to allow the streamlining of approaches for tailoring care to the needs of individual patients.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

ORCID

Asma Alahmadi https://orcid.org/0000-0001-6603-1596
Yassin Abdelsamad https://orcid.org/0000-0001-8281-5825
Medhat Yousef https://orcid.org/0000-0003-2102-6704
Salman F Alhabib https://orcid.org/0000-0002-2646-0393
Afrah Alshalan https://orcid.org/0000-0003-0265-6049
Nezar Hamed https://orcid.org/0000-0002-8622-6195
Farid Alzhrani https://orcid.org/0000-0002-0564-7204

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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