PAROXYSMAL POSITIONAL VERTIGO AFTER COCHLEAR IMPLANTATION

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Running head: PPV after cochlear implant

ABSTRACT

CONCLUSIONS - The observed 4 cases of PPV in our series of adult cochlear implant (CI) recipients occurred in the implanted ear without chronological relation with the surgical procedure or the implant's activation. All of them relapsed within 3 months from an initially successful repositioning maneuver, and finally recovered after the second one. None of the patients showed labyrinthine weakness in the implanted ear. The outbreak of PPV did not affect the patients' speech perception performances.

OBJECTIVES - To report and discuss the occurrence of PPV after cochlear implantation.

METHODS - Among 32 adult patients who received a Nucleus® CI at the ORL Dept. of the University of Brescia, 4 suffered of acute vertigo spells, diagnosed as PPV on the basis of Dix-Hallpike's maneuvers. After a Semont's repositioning maneuver, recurrences were similarly addressed. An ENG recording of bithermal caloric irrigation tests were obtained once the symptoms definitively subsided.

RESULTS - The observed incidence of 12.5% exceeds the figures reported in the literature. Since no anatomic abnormalities were identified in these patients, nor were any intra- or post-operative complications reported, we can only speculate about the possible ethiopatogenetic role of the array's insertional trauma. PPV developed 1 to 12 months after CI surgery in the posterior canal on the side of the implant and relapsed within 3 months. ENG showed a normal reflectivity in 2 patients, while the 2 others had a significant prevalence of the labyrinth in the implanted ear. The outcomes of the speech perception tests after CI in all the 4 patients did not differ from the results achieved by other patients who belong to corresponding categories. At the longest follow-up (6 to 24 months) all patients are free from vertigo.

INTRODUCTION

Cochlear implantation (CI) performed by experienced operators is considered a safe surgical procedure, with a low rate of both major and/or minor complications.

Among the latter, post-operative dizziness is a frequent consequence of the decrease of the vestibular function that may occur in deaf patients submitted to CI. The incidence of post-operative vestibular disturbance ranges between 20 and 60% [1,2,3,4,5] and is mainly related to the pre-operative status of the labyrinthine function [1,2,3] but also to a number of co-factors, such as age, cardiovascular or metabolic risk factors and pre-implantation vestibular symptoms, especially in Meniere's disease-related deafness [1,2,3,4]. Thus, it can manifest with many different clinical presentations, but in most cases it is mild and self-limiting within a few days [3].

Paroxysmal positional vertigo (PPV) after CI is rarely described, although it is the most frequent peripheral vestibular disorder among non-implanted people [6,7]. It is well known that the main clinical sign of PPV determined by lithiasis in the posterior semicircular canal (PSC) is the nystagmus (Ny), observable with Frenzel glasses, typically evoked by the Dix-Hallpike or Semont's maneuvers. Its main features are: latency of onset: from 2 to 20 seconds; rotatory geotropic direction; fast build-up and exhaustion; short duration (5-30 seconds); typical "rebound" (change of direction) when returning to the sitting position.

During the last 3 years we observed 4 cases of PPV among 62 patients who underwent a cochlear implant procedure. In the present study we analyzed the clinical, audiological and otoneurological features of these patients, in order to assess if the correlation between the CI and the occurrence of PPV was casual or determined.

MATERIALS AND METHODS

Between January 2002 and December 2005, sixty-two patients underwent cochlear implantation at the Otolaryngology Department of the University of Brescia. Thirty-two were adults (age range 18-79 years, 17 males and 15 females) and 30 children (age range 2-15 years, 12 males and 18 females).

After appropriate audiologic assessment, all candidates to CI underwent a radiological study by means of high resolution CT scan of the temporal bones (post-verbal patients) combined with an MRI of the brain (pre- and peri-verbal children). A neuropsychiatric evaluation was always included in the diagnostic protocol, as well as clinical vestibular examination. Patients with a positive clinical history for vestibular disorders underwent also instrumental vestibular testing, i.e. electro-nystagmography (ENG). After the activation of the cochlear implant, that usually occurred

at the 3rd or 4th post-operative week, all patients started an individualized habilitative training with speech therapists. During this period the implants' maps were re-fitted as frequently as needed by a trained Audiometrist (S.P.) or by the Audiologist (C.C.). Perceptive performances were tested at scheduled intervals, during the fitting sessions.

During every training session the speech therapist was instructed to take thorough notes of every comment or complaint of the patients and to report to the Otologist responsible for the CI program (D.Z.). All patients who claimed to suffer of vertigo or dizziness were referred for vestibular examination. Post-operative X-rays with the modified Stenvers projection [8] were obtained in all patients in order to verify the correct insertion of the intra-cochlear electrode array.

Among the 62 implanted patients, 4 suffered of an acute vertigo spell that was diagnosed as PPV, on the basis of Dix-Hallpike's diagnostic maneuvers.

Table I summarizes some of their relevant clinical features.

In 3 patients the onset of deafness was post-verbal, while in one it was pre-verbal. The etiology of deafness in the post-verbal subjects was chronic otitis media (n=1) and progressive degenerative cochlear loss of microvascular nature (n=2). The hearing loss was bilateral in all cases, with slight asymmetry in two. It was profound in 3 and severe to profound in one. The post-verbal candidates underwent the CI after 3 to 10 years of auditory deprivation in the affected ear. All patients received a Cochlear device, Nucleus CI 24R "Contour" (n=2), CI24CA "Contour Advance" (n=2).

All implant procedures were performed by the same surgeon (D.Z.) with the "soft surgery technique", recently described by Roland [9]

Once PPV was diagnosed, each patient was immediately treated with a Semont's repositioning maneuver [10], followed by a 48 hours period of rest and subsequent repetition of the Dix-Hallpike diagnostic maneuver. Eventual recurrences were recorded and treated with the same modalities. An ENG recording of bithermal caloric irrigation tests were obtained once the symptoms definitively subsided.

RESULTS

The 4 patients who manifested an attack of PPV after CI represent 6.4 % of the patients implanted at our institution. Excluding children (n=30), the rate in adult CI recipient becomes 12.5% (4/32). No anatomic abnormalities were identified in these patients by the pre-operative imaging studies, nor were any intra-operative complications reported. All 4 patients had an uneventful post-operative course; none of them complained of dizziness or vertigo. The electrodes array's insertion appeared correct by intraoperative electric impedance testing and Neural Response Telemetry (NRT), as well as by the post-operative modified Stenvers x-rays.

No patient were complaining of vertigo at the moment of the surgical procedure. Patient #4 (B.L.) had suffered three years earlier of an undefined episode of long-lasting vertigo, with a diagnosis of acute left labyrinthine deficit, on the same side later implanted. Another one had suffered from vertiginous spells with the characters of PPV in the past (pt. # 2, A.S.) but he could not recall which side had been affected.

Table II shows the features of the vertigo observed in the described patients. They developed PPV at an average of 6 months after CI surgery (range 1-12 months). In every one of them posterior canal was involved on the same side of the implanted ear. Nystagmus was in each case rotatory, geotropic and exhaustible, with duration between 2 and 10 seconds.

In no instance the onset of the paroxysmal vertigo was related with the switch-on of the device or with any of the following fitting sessions.

All patients felt subjectively relieved after the Semont's repositioning maneuver; the control maneuver performed 48 hours later did not provoke subjective vertigo nor further elicited nystagmus in any of them. Rather surprisingly, though, all patients developed a relapse with a mean delay of 2.25 months from the first episode (range 2-3 months). A second therapeutic maneuver was then administered with the same modalities, and this time all patients were definitively cured.

An ENG recording of bithermal caloric irrigation tests was obtained once the symptoms disappeared. These postoperative ENG showed vestibular symmetry with a normal reflectivity in 2 patients, while the 2 others had a significant prevalence of the labyrinth in the ear receiving the implant. As one would expect a deficient function on the side of the implant, our findings could be explained by the presence of an "irritative" state of the labyrinth in the implanted ear.

The outcomes of the speech perception tests after CI in all the 4 patients suffering from PPV did not differ from the results achieved by other patients who belong to corresponding categories at our Cochlear Implant centre (Figure 1). None of the 4 patients had residual hearing preserved in the implanted ear.

At the longest follow-up (6 to 24 months) all patients are free from vertigo and they are carrying out a normal social life.

DISCUSSION

The fate of vestibular function in deaf patients undergoing CI is controversial. A cochlear implant in the only hearing ear could cause a bilateral vestibular areflexia, with chronic disequilibrium and oscillopsia [2]. Conversely, an old study by Eisenberg et al [11] found an improvement of postural stability in CI recipients who preoperatively suffered from vestibular deficit, in agreement with the

more recent results of Buchman et al [5]. The effects of CI on the posterior labyrinth depends on its pre-operative status and on the extent of surgical trauma.

Before implantation the vestibular function may vary from normal to absent [1]. The insertion of the electrode array into the scala tympani can disrupt the membranous cochlear lateral wall and also endanger the posterior labyrinthine compartment if the basilar membrane is torn [12].

Several authors suggest a vestibular pre-operative evaluation in order to offer a better counseling about the post-operative course [1] while others do not deem it a valuable prognostic indicator [13].

In our Department, similarly to other centers, we do not routinely perform vestibular testing in preparation for CI, unless indicated for specific reasons, i.e. positive clinical history or syndromic patients. As generally accepted, we do not request as well an instrumental vestibular testing to obtain a diagnosis of PPV.

The rate of postoperative vestibular deficit induced by the CI ranges in literature between 20 and 60% [1,2,3,4,5].

Migliaccio et al [14] found a low incidence (9%) of post-operative decrease of vestibular function and attributed it to the surgical preservation of membranous labyrinth.

Rossi et al [15] remarked that the surgical procedure does not affect the vestibular response in the majority of patients, suggesting that the choice of the side to implant should not be influenced by the outcome of the vestibular examination.

Age is considered a relevant factor by some Authors. Patients below 60 years of age seem to be less exposed to the risk of post-surgical labyrinthine dysfunction [1,2,3,4].

Recently Fina et al [4] reported a study concerning 75 patients, with a mean age of 57 ys (20-86) and a prevalence of postoperative dizziness of 39%. The majority of them perceived it as imbalance or vertigo, delayed in its onset; four (5,3%) complained of early perioperative transient attacks induced by head position changes. The Authors identified some risk factors for post-CI vertigo: age at implantation, abnormal preoperative dynamic posturography, preoperative symptoms especially in Meniere's disease.

Ito [2] classified the post-implant vestibular disorders in: early (onset within 2 weeks); prolonged (early onset but duration greater than 2 weeks); delayed (starting after 2 weeks at least). Early transient vertigo is the most frequent manifestation, theoretically caused by the surgically-induced perilymphatic fistula or by spread of electric current to the adjacent vestibular labyrinth.

The explanation for the prolonged or delayed vestibular dysfunction, apparently not related with the surgical trauma, is less obvious. In a Japanese study [3] the delayed vertigo was assumed to depend on an endolymphatic hydrops induced by the array's insertion or by chronic indirect electrical stimulation of the vestibular nerve.

The hypothesis of perilymphatic fistula has been recently confirmed by Kusuma et al. [16] by means of CT scan, which detected the presence of air in the vestibule. Significantly, in their patient the dizziness started 2 weeks after implantation, it was increased by head's movements, and it lasted for several months. The patient had immediate and complete resolution of symptoms after revision tympanotomy. A similar case was quoted by Hempel et al [17]: the onset was 8 months after cochlear surgery and after patient blew his nose hardly; in this case the symptoms subsided without re-exploration of the middle ear.

A case of post-implant acute rotational vertigo associated with fluctuating electro-acoustic thresholds was described as autoimmune-mediated endolymphatic hydrops [18].

Different hypothesis have been postulated in the literature to explain the post-CI vertigo:

-electrical stimulation of the vestibular receptors [2];

-perilymphatic fistula due to imperfectly sealed cochleostomy [16];

-mechanical trauma to membranous labyrinth [19];

-obstruction of endolymphatic flow [3,4,5].

To date, unfortunately, there are no pathological or electrophysiological studies confirming or excluding either hypothesis.

Opposed to the high incidence of PPV as a cause of peripheral vertigo in the general population, cupulo-canalolithiasis post-implantation is very rarely reported (Table III). Di Girolamo et al [7] described a case of lateral canal PPV originating in the implanted ear; it occurred 3 days after the activation of the device, and the patient had a normal pre-operative vestibular function. The same Authors hypothesized an otolithic dislocation due to electrical stimulation during initial fitting, but they did not exclude a casual association.

Fina et al [4] noted only 4 cases of PPV out of 75 CI recipients (5.3%); another 39% of their patients complained of unspecified dizziness in the postoperative period.

Limb et al [13] reported a PPV rate of 2.2% (12 out of 540 adult patients) corresponding to an incidence of 159 cases in 100.000 persons per year. This figure is significantly higher than that of 64/100.000/year in the general (non-implanted) population [20]. They did not observe an impairment of the audiologic performance in their patients complaining of PPV.

Only Steenerson et al [21] found that PPV after CI was the prevalent condition among their patients complaining of post-implant vertigo (23 out of 82, i.e. 28%), in each case involving the implant side.

The 4 patients who manifested an attack of PPV after CI represent 6.4 % of the patients implanted at our institution. If we exclude children, according to the low rate of PPV in the pediatric population [22] and to the challenges involved in eliciting vestibular symptoms histories [13], the

incidence of PPV among our adult CI recipients is 12,5 % (4/32), a figure exceeding the average incidence of the disease both in the general and in the deaf population. The 4 patients who developed PPV are mildly older then the average non-PPV adult CI recipients in the present series (66.25 vs. 52.8 years of age).

Among the other 28 adult patients without signs of PPV, one only recalled an acute vestibular deficit in the past, and in two others the etiology of deafness was related to bilateral Meniere's disease. None complained of vestibular symptoms at the moment of the implantation nor did they suffer from vertigo in the post-operative period, up to the longest follow-up (3 years). Eight out of 28 had associated cardiovascular risk factors. Six of the 28 maintained some residual hearing after implantation, accounting for 21.4% of all implanted patients.

In the literature, different etiopathogenetic explanations have been proposed for post-CI positional vertigo:

1. Mechanical trauma associated with electrodes insertion as a stimulus for otolithic dislocation [23];
2. Bone dust production during cochleostomy: microscopic bone debris could pass through a micro rupture of the basilar membrane, determining an accretion in the posterior canal [13];
3. Vibration trauma during cochleostomy drilling [13].

It is acknowledged that a misplaced cochleostomy in the region of the round window can induce a variety of pathologic changes such as basilar membrane rupture, scala media transaction, obstruction by drillout debris of the ductus reuniens at the hook region of the cochlea and direct saccular damage [5], but there is no mention of correlation with otolithic dislodgement. Clark et al [24] in a post-mortem study demonstrated end organ vestibular damage caused by a wrong electrodes' array insertion.

In a recent histopathological analysis of temporal bones from implanted patients [25], the device was not found to be responsible of deafferentation of the peripheral vestibular system, nor there was evidence of damage to the vestibular end organs or to the primary vestibular neurons. There was no significant difference in Scarpa's ganglion cell counts and hair cell densities between the implanted and non-implanted sides. Conversely, cochlear hydrops was a common finding, leading to attacks of delayed vertigo resembling Meniere's syndrome.

On the basis of the observation of the 4 patients included in the present series, we can infer that the risk of developing PPV in the implanted ear is slightly higher than in the general population, and that the inclination to recur within the next 2-3 months despite a correct and effective repositioning maneuver is concrete.

Furthermore, the vestibular response, that was preserved also in the implanted ear in almost all the 28 non-PPV adult patients, was set at the upper normal limit or even increased in the 4 ears with PPV, as if the labyrinth was enduring an irritative condition.

At the present state, we are unable to provide a feasible explanation of the phenomena underlying PPV in CI recipients, except for theoretical speculations, as well as we cannot rule out a relation with the surgical trauma, although the onset is chronologically delayed.

CONCLUSIONS

The incidence of PPV in our population of adult implanted patients (12.5 %) exceeds the average rates reported in the literature, being also higher then in non-implanted people.

In all cases the PPV occurred in the implanted ear, and, significantly, it relapsed within 3 months on the same side. This would lead to the assumption that the surgical trauma or the electrode array itself could play a relevant pathogenetic role. Conversely, the absence of chronological relation between the outbreak of PPV and the surgical procedure or the implant's activation, would deny such a close relationship.

In our series of four patients the bi-thermal caloric testing demonstrated complete post-operative preservation of vestibular function in the implanted ear in 2 cases, and in the other 2 we observed a temporary contra lateral labyrinthine weakness.

We can postulate that the surgical maneuvers during a cochlear implantation procedure do not cause a deterioration of the labyrinthine function. No correlation can be established between post-operative PPV and type of device, etiology of deafness or concurrent risk factors. Unfortunately, no definite conclusions can be drawn, due to the limited series of patients.

Nevertheless, it is clear that the outbreak of PPV does not affect, per se, a good audiologic outcome of cochlear implantation.

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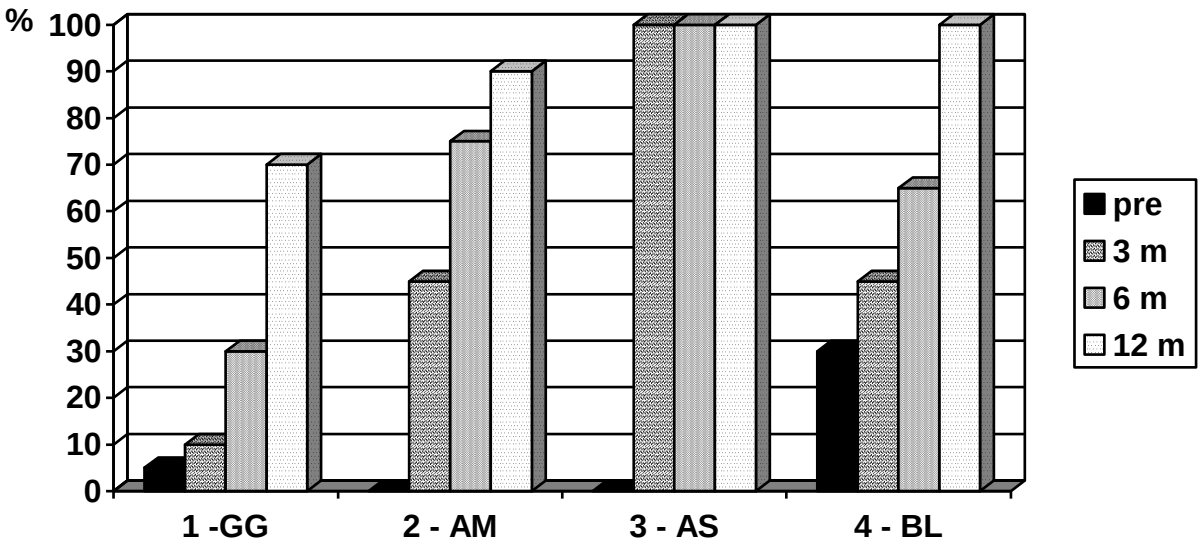
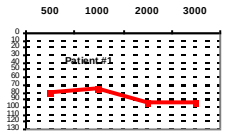
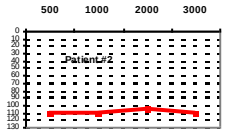
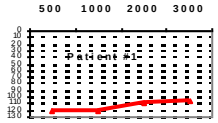
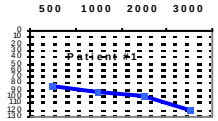


Fig 1: Recognition scores for phonetically balanced bisyllabic words for the CI patients with PPV (open set without lip-reading). Note that patient #1 is the only preverbal subject in the group.

Table I: Demographics and audiological assessment of implanted patients who developed PPV. PTA= pure tone average (0.5-1-2-3 kHz), WRS=word recognition scores

	Age	Sex	Side	Etiology	Co-factors	Pure tone audiometry	PTA	WRS	Device type
GG	54	F	Rt	Genetic	Hypertension		86,25 dB	5%	Nucleus CI24 R
AS	72	M	Rt	Progressive degenerative	Hypertension;		98,75 dB	0%	Nucleus CI24CA
AM	64	M	Rt	Progressive degenerative	Hypertension; myocardial infarction		112,5 dB	0%	Nucleus CI24R
BL	75	F	Lt	Bilateral chronic otitis	Antiplatelet drugs Chronic cerebrovascular disease		100 dB	45%	Nucleus CI24CA

Patient	CI side	Onset after CI	Caloric tests (ENG)	Previous vestibular dysfunction	Relapses
1. GG	Right	7 months	Simmetric; left prev 9%; right DP 46%	---	2 months
2. AS	Right	4 months	Right prev 24%	One PPV episode	2 months
3. AM	Right	12 months	Simmetric; right prev 6%; left DP 26%	---	3 months
4. BL	left	1 month	Left prev 82%; right DP 34%	Left acute labyrinthine deficit (3 ys. earlier)	2 months

Table 1I: characteristics of PPV in CI recipients.

Prev = labyrinthine prevalence; DP = directional preponderance of nystagmus

	# of PPV	Age, risk factors	Features of PPV	Onset latency	Vestibular tests	Recurrence	Side
Di Girolamo et al 1999	1	40 ys; none	Severe; recovery after 2 repositioning maneuver	6 weeks post-CI	Symmetric normal responses	No	Implanted ear
Steenerson et al 2001	23/82	Mean 50,3 ys; (no correlation with risk factors)	Recovery after 1 repositioning maneuver	n.s.	n.s.	No	Implanted ear
Fina et al 2003	4/75	n.s.	n.s.	Delayed (>24 hours post-CI)	n.s.	Yes	n.s.
Limb et al 2005	12/540	none	n.s.	Mean 29.2 days	Not performed	No	n.s.

Table III: literature review of positional vertigo after cochlear implantation.