

Musical ear syndrome in adult cochlear implant patients

W-K LOW, C A THAM, V-D D'SOUZA, S-W TENG

Department of Otolaryngology, Singapore General Hospital, Singapore

Abstract

Objective: Except for a single case report, musical ear syndrome in cochlear implantees has not been studied. We aimed to study the prevalence and nature of musical ear syndrome among adult cochlear implant patients, as well as the effect on their emotional well-being.

Study design, patients and intervention: A cross-sectional survey of patients aged 18 years and above who had received cochlear implants for profound hearing loss between 1997 and 2010.

Results: Of the 82 patients studied, 18 (22 per cent) were found to have experienced musical ear syndrome. Seven and 11 patients had musical ear syndrome prior to and after cochlear implantation, respectively. The character of musical ear syndrome symptoms was described as instrumental music ($n = 2$), singing (6) or both (10). Fourteen patients reported an adverse emotional effect, with three expressing 'intolerance'.

Conclusions: In this study, 22 per cent of cochlear implantees experienced musical ear syndrome. These symptoms affected patients' emotional state, but most coped well. Musical ear syndrome can occur prior to and after cochlear implantation.

Key words: Music; Hallucinations; Cochlear Implants; Hearing Loss; Deafness

Introduction

Musical ear syndrome is a form of musical hallucination, and is defined as the subjective experience of hearing music, or aspects of music, when none is being played.¹ Such hallucinations may be intermittent or continuous and are generally not controllable.^{1–4} Musical hallucinations have been found in association with social isolation¹, advanced age, hearing impairment,^{5–8} brain lesions⁹ and mental disorders such as depression.⁴ The association between hearing impairment and musical hallucination is well documented; a comparison has been drawn between musical ear syndrome and Charles Bonnet syndrome, in which patients with visual loss experience complex visual hallucinations with preservation of insight. However, except for a single case report, musical ear syndrome in cochlear implantees has not been studied.¹⁰

Our study aimed to investigate the prevalence and nature of musical ear syndrome among adult patients who had undergone cochlear implantation (CI), and to assess the effect on their emotional well-being.

Materials and methods

The study consisted of a cross-sectional survey of adult patients (defined as patients aged 18 years and above) who had received cochlear implants for profound

hearing loss between 1997 and 2010 at our institution. All the patients were primarily managed by the senior author (WKL). Patients were sent survey forms to assess whether they were experiencing musical ear syndrome.

Among these patients, those who had experienced musical ear syndrome were identified. These patients' case records were reviewed and they were invited to complete a questionnaire. This gathered information on the nature, characteristics and frequency of symptoms experienced, and explored the effect musical ear syndrome had on patients' mood, sleep and work.

The study was approved by the hospital's research ethics committee, the Centralised Institution Review Board. All patients included in the study gave written, informed consent to study inclusion, and were judged competent and capable of doing so.

Results and analysis

We included in the study 82 adult patients who had received cochlear implants at our institution between 1997 and 2010. All these patients' hearing had improved after CI.

Of these 82 patients, 18 (22 per cent) were experiencing musical ear syndrome at the time of study: 10 men (56 per cent) and 8 women (44 per cent). Participants'

ages ranged from 18 to 61 years. No patient had any history of mental illness. All patients had insight into their musical ear syndrome and recognised that the music they heard was a perception without any corresponding external cause. With regard to medical history, one patient had a history of stroke and another had had a brain tumour.

Patients described the character of their musical ear syndrome symptoms as instrumental music ($n = 2$), singing (6) or both (10) (Table I). Of the 18 patients with musical ear syndrome, 7 reported that it had started before CI. In five of these seven patients, CI had improved their musical ear syndrome symptoms, in that the perceived sound had become much softer. The remaining two patients, however, felt that their symptoms had become louder after CI. Interestingly, 11 of the 18 patients who experienced musical ear syndrome indicated that the symptoms had developed only after CI. As shown in Table I, the frequency of episodes ranged from less than once a week to 5–7 times per week. In 9 of the 18 patients, episodes lasted less than an hour. Eleven out of the 18 patients indicated that they felt they were ‘in control’ and able to ignore the hallucinations.

We explored the effects of musical ear syndrome on an array of emotional states ranging from helplessness and unhappiness to anger (Table II). As shown in Table II, 14 patients had on occasion experienced at least one adverse emotional effect because of their musical ear syndrome. Fortunately, most were able to cope with these emotions, with only 3 out of the 14 patients expressing ‘intolerance’ overall. Interestingly, none of these three patients had complained about experiencing musical ear syndrome symptoms or emotional ill effects during their regular post-CI follow-up medical visits.

Discussion

Musical ear syndrome typically causes affected individuals to hear instrumental music or singing, and is more common in women.^{1,7} The hallucinatory experiences differ in character from those commonly experienced in psychotic disorders, and have different neural correlates.¹¹ The most important distinction is the preservation of insight. There should not be any other psychotic symptoms present, especially hallucinations in other modalities. There should be an absence of serious mental illness, although mild cognitive impairment is often associated with musical ear syndrome. In our series, there was no gender preponderance. None of our patients had any symptoms of psychosis, and all were fully aware that the music they perceived was coming from ‘within their heads’.

Musical ear syndrome has been hypothesised to be secondary to sensory deprivation of the auditory cortex, akin to the effects of visual sensory deprivation seen in Charles Bonnet syndrome. Cope and Baguley¹² conducted a literature review on musical hallucination and found that existing literature supported the proposal that the otological system played a role in the pathogenesis of musical hallucination. Griffiths⁷ proposed that sensory deprivation of the auditory cortex in hearing-impaired individuals leads to relative overactivity of the auditory association cortex, and thence to musical ear syndrome. Seven of our patients (39 per cent) experienced musical ear syndrome prior to receiving their cochlear implants. Of these, five (71 per cent) experienced improvement of their musical ear syndrome symptoms after receiving their cochlear implant. This finding supports the hypothesis that musical ear syndrome is due to sensory deprivation of the auditory cortex. Following CI, there is restoration of auditory stimuli to the auditory cortices, which

TABLE I
PATIENT CHARACTERISTICS OF MUSICAL EAR SYNDROME

Pt no	Age (y)	Gender	Musical ear syndrome episodes			
			Frequency (per wk)	Duration (hr)	Onset	Character
1	56	Male	<1	<1	Post-CI	M
2	25	Male	5–7	>8	Post-CI	M & S
3	70	Female	5–7	<1	Post-CI	S
4	58	Female	1–4	1–3	Pre-CI	M & S
5	54	Male	5–7	>8	Post-CI	M & S
6	43	Male	5–7	>8	Post-CI	M & S
7	23	Male	<1	<1	Post-CI	S
8	22	Male	1–4	1–3	Pre-CI	M & S
9	61	Female	<1	<1	Pre-CI	S
10	23	Male	1–4	5–7	Post-CI	M
11	18	Male	<1	<1	Post-CI	S
12	30	Female	1–4	<1	Post-CI	S
13	57	Female	5–7	1–3	Post-CI	S
14	57	Female	5–7	>8	Pre-CI	M & S
15	45	Female	1–4	<1	Pre-CI	M & S
16	43	Female	1–4	<1	Pre-CI	M & S
17	31	Male	<1	<1	Post-CI	M & S
18	47	Male	5–7	>8	Pre-CI	M & S

Pt no = patient number; y = years; wk = week; hr = hours; CI = cochlear implantation; M = instrumental music; S = singing

TABLE II
EMOTIONAL EFFECTS OF MUSICAL EAR SYNDROME

Pt no	Tolerance of MES?	Emotional effect										
		Unhappy or depressed	Tense or unable to relax	Angry	Confused	Helpless	Insecure	Tired	'Drives me crazy' or 'Frustrated'	Unable to concentrate or work	Social phobia	Interferes with sleep
1	Yes	0	0	0	0	0	0	0	0	0	0	0
2	Yes	1	0	0	0	2	0	0	0	0	0	2
3	Yes	0	0	0	0	0	0	0	0	0	0	0
4	Yes	0	0	0	1	0	0	1	0	1	0	1
5	No	2	2	1	2	3	1	3	2	1	0	1
6	Yes	2	2	2	2	1	1	1	1	1	1	1
7	Yes	0	2	1	2	0	0	0	0	0	0	0
8	Yes	1	1	1	2	2	1	1	1	2	0	1
9	Yes	0	0	0	0	0	0	0	0	0	0	0
10	No	2	2	3	3	3	3	3	3	3	3	2
11	Yes	0	0	0	0	0	0	0	0	0	0	1
12	Yes	3	2	2	3	0	0	3	0	2	0	0
13	Yes	0	0	0	0	0	0	2	2	1	0	2
14	No	3	3	3	2	3	2	3	2	2	2	2
15	Yes	2	2	1	2	1	1	1	0	1	1	0
16	Yes	3	0	0	0	2	0	2	2	2	2	3
17	Yes	0	0	0	0	0	0	0	0	0	0	0
18	Yes	2	2	2	2	2	2	2	2	2	2	2

Pt no = patient number; MES = musical ear syndrome. Degree of emotional effect: 0 = nil; 1 = mild; 2 = moderate; 3 = severe.

would be expected to result in overall improvement of musical ear syndrome symptoms.

The possibility of musical ear syndrome developing after CI is interesting. Auffarth *et al.*¹⁰ reported the case of a 66-year-old woman who developed musical hallucinations after CI. She had no history of mental illness. She described her symptoms as hearing music and rhythmic humming, which began softly and then became progressively louder. The symptoms persisted even when the cochlear implant was active. As the symptoms had developed only after hearing restoration with the cochlear implant, the authors questioned the theory that auditory sensory deprivation was the cause of musical ear syndrome. However, they offered no explanation as to how CI could have resulted in musical ear syndrome. As CI could have destroyed residual cochlear hair cells, the development of musical ear syndrome in cases of hearing loss may well be linked to disruption of inhibitory neural feedback mechanisms originating from cochlear hair cells.

In our series, 11 patients (61 per cent) reported that symptoms of musical ear syndrome developed only after CI. This observed prevalence should be interpreted with caution, as recall bias could have been a contributing factor in some patients; musical ear syndrome could possibly have been present even before CI. Nonetheless, the fact remains that 18 out of the 82 cochlear implant recipients studied (22 per cent) were experiencing symptoms at the time of study. These symptoms could negatively affect emotional states, but they were generally mild. Although three patients had expressed intolerance of their symptoms, the possibility of an exaggerated response could not be excluded. None of these three had volunteered information relating to musical ear syndrome experience during their routine post-CI follow-up medical visits, which was surprising if their symptoms had indeed been intolerable.

- **Musical ear syndrome is a form of musical hallucination**
- **It is associated with social isolation, old age, hearing impairment, brain lesions and mental disorders**
- **In this study, it was experienced by 22 per cent of cochlear implant recipients**
- **Although common, emotional effects were generally mild and well coped with**
- **Cochlear implantation can both improve and trigger symptoms**
- **Pathogenesis is probably more complex than simple sensory deprivation of the auditory cortex**

Currently, there is little consensus regarding the optimal treatment for musical ear syndrome.

Treatment of musical ear syndrome has only been successful in cases with a clear underlying cause. For example, depressed patients have been successfully treated with anti-depressants,⁵ while hearing-impaired patients have experienced relief with improved amplification.⁷ For many patients with musical ear syndrome of unclear aetiology, pharmacological therapy (including anti-epileptics and anti-psychotics) has been instigated but results have been mixed,^{5,7,13} with idiosyncratic responses. Hence, in the absence of data from randomised controlled trials, pharmacological therapy should ideally be reserved for patients with identifiable, treatable causes. Patient counselling remains the best option at this point in time.

Although this study was limited by its retrospective nature and relatively small number of subjects, the results suggest that the pathogenesis of musical ear syndrome is probably more complex than just sensory deprivation of the auditory cortex. A prospective, longitudinal study on a larger scale is warranted to better reveal the natural history and progression of musical ear syndrome in profoundly deafened patients treated with CI.

Conclusion

In this study, 22 per cent of cochlear implant recipients were found to be experiencing symptoms of musical ear syndrome. Although these symptoms commonly affected emotional states, they were generally mild, with most patients coping well. Although the symptoms of musical ear syndrome were improved by CI in some patients, in others they developed only after implantation. The pathogenesis of musical ear syndrome is therefore likely to be more complex than just sensory deprivation of the auditory cortex; disruption of inhibitory neural feedback mechanisms originating from the cochlear hair cells may possibly play a role.

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Address for correspondence:

Dr Wong-Kein Low,
Visiting Consultant,
Department of Otolaryngology,
Singapore General Hospital,
Singapore, 169608

Fax: +65 62262079

E-mail: low.wong.kein@gmail.com

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