Tinnitus

Clinical and Research Perspectives

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Tinnitus and Hyperacusis in Literature, Film, and Music

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Introduction

When a clinician or scientist specializes in a particular condition or symptom, he or she can become vigilant to mentions of that symptom in literature and music. The same can be true of patients, who may be hyper-alert to and troubled by mentions of their symptom in a negative or catastrophized way. Having worked with tinnitus patients for over 30 years, and more latterly with hyperacusis, I have spotted several mentions of these symptoms in literature, film, and music, and patients have drawn my attention to others. Here I recall those that come to mind, and reflect upon the contexts in which tinnitus, hyperacusis, or both are mentioned, and consider what this may tell us about the experiences people associate with those symptoms. In a compendium of depictions of neurological disease in fiction, Bogousslavsky and Dieguez (2013) note that the description of the symptoms may be "creative and humane" and allow the clinician to view them from a different perspective. My list of references to tinnitus and hyperacusis will certainly not be exhaustive (and is essentially restricted to work written in or translated into English) but perhaps will spark some interest.

Tinnitus in Literature

The novel *A Pair of Blue Eyes* (1873) by Thomas Hardy (1840–1928) is viewed as one of his minor works, and the structure reflects the book's initial serialization, with a sequence of melodramatic situations. One of the main characters is the local vicar Stephen Swancourt in the fictional village of Endelstow in the English West Country. He has an assistant, William Worm, who describes tinnitus:

> Worm said groaningly to Stephen, "I've got such a noise in my head that there's no living night nor day. 'Tis fir all the world like frying fish: fry, fry, fry all day long in my poor head, till I don't know whe'r I'm here or yonder, There, God A'mighty will find it out sooner or later, I hope, and relieve me."

> "Now, my deafness," said Mr. Swancourt impressively, "is a dead silence; but William Worm's is that of people frying fish in his head. Very remarkable, isn't it?"
>
> "I can hear the frying-pan a-frizzle as naterel as life," said Worm corroboratively. (Hardy, 1873, p. 24)

This high-frequency tinnitus plays no further part in the plot, however.

The main protagonist and narrator of the novel *Money* by Martin Amis (1984) is John Self, an amoral and dissolute man who aspires to being a film producer and thereby making and spending large amounts of money. He only succeeds in the latter, and it transpires that the film plan was an elaborate hoax, and that all whom Self had trusted were in fact duplicitous in many and various ways. As the novel opens, Self complains of recent-onset tinnitus:

> Owing to this fresh disease I have called tinnitus, my ears have started hearing things recently, things that aren't strictly auditory. Jet take-offs, breaking glass. Ice scratched from the tray. It happens mostly in the mornings . . . (Amis, 1984, p. 1)

> I'm drinking tax-exempt whisky from a toothmug, and listening to see if I'm still hearing things. The mornings are the worst. This morning was the worst yet. I heard computer fugues, Japanese jam sessions, didgeridoos. What is my head up to? I wish I had some idea what it's got in mind for me. (Amis, 1984, p. 5)

Self's tinnitus is thus a formless auditory hallucination rather than tinnitus as generally experienced, though such reports are heard in the clinic. The context of stress in which Self develops tinnitus is also commonly observed in clinic.

Transient tinnitus is mentioned as an aside several times in the Harry Potter series, associated with Harry being angry, and with Hermione Granger's use of time travel in order to study harder. A more substantial reference is in the fourth book, *Harry Potter and the Half-blood Prince* (Rowling, 2005), wherein Harry finds that his allocated textbook for his Potions class has been annotated by a previous student, who turns out to have been his teacher Severus Snape. Among the scribbled notes are some spells, including, "perhaps most useful of all, *Muffliato*, a spell that fills the ears of anyone nearby with an unidentifiable buzzing, so that lengthy conversations could be held in class without being overheard" (Rowling, 2005, p. 224). Hermione refuses to use the spell, but Harry finds it of value when needing to have conversations un-overheard as the plot of the book unfolds.

Another children's book of interest is *Clicking Vicky* (Freud, 1980) (Figure 1–1). The eponymous heroine has an unusual trait:

> She clicked. Every night. Practically all the time it was dark and sometimes in the afternoon as well. . . . Absolutely regular, every nine seconds there would come from the dozing or sleeping child a small, precise and rather well-bred noise such as you would get if you struck the end of your little fingernail hard against your front tooth. . . . (Freud, 1980, p. 2)

In the book, Vicky is diagnosed with congenital "epiglottal prontosis" (which may be an error by the author, Clement Freud, as this is not a term in medical use), and the regularity of her clicks proves useful when all the clocks in the world fail. A more common finding in the audiology context would be middle ear myoclonus (Chapter 9), in which the clicks would be less regular but more frequent (Bhimrao, Masterson, & Baguley, 2012).

Religious Literature

There are 20 instances of the word *deaf* in the Bible, usually as a metaphor for not listening to God. There are no references to tinnitus,

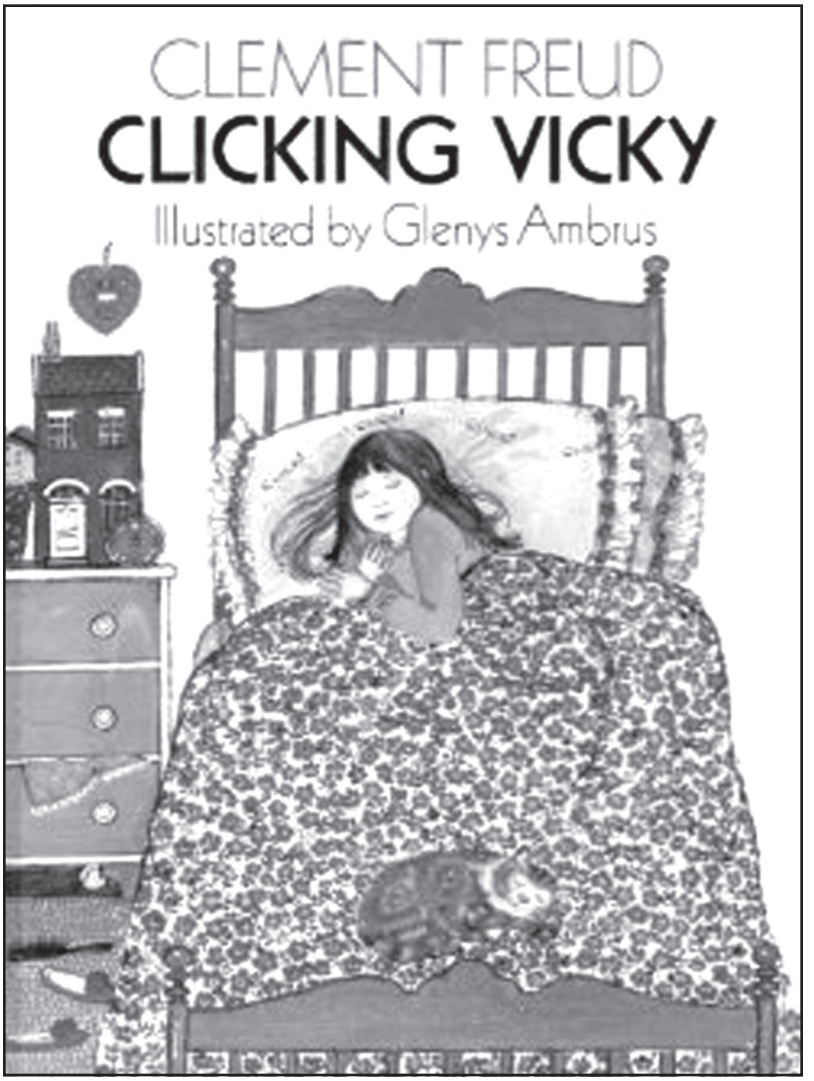

Figure 1–1. The cover of *Clicking Vicky* (Freud, 1980).

however. The Talmud is a Jewish Rabbinic work, compiling oral Torah (instruction) with expositions of the Hebrew Bible. It contains some historical references, and regarding tinnitus recounts the story of the Roman Emperor Titus (AD 39–81) who ordered the destruction of the Second Temple in Jerusalem (AD 70). The Talmud describes Titus as having tinnitus (Dan, 2005), and considers that as punishment for the fall of the Temple. The Talmud indicates that Titus had some short-lived relief:

> A gnat entered his nostril and pecked at his brain for seven years. One day Titus was passing by a blacksmith. He heard the sound of the sledgehammer and the gnat became silent. Titus thus said: "here is the remedy." Everyday he brought a blacksmith to bang in his presence . . . for thirty days this worked

> fine but then the gnat became accustomed and resumed pecking. (Quoted in Dan, 2005, p. 211)

This represents an early illustration of sound therapy, and the clinical observation that this may seem very effective initially but not result in long-term benefit. This is also an example of a person with tinnitus establishing an elaborate system of self-help (see Chapter 14).

Meditation is an integral part of Buddhist practice, and the experience of hearing tinnitus-like sounds while deeply meditating is mentioned quite often. An example is in the practice of nada yoga (*nada* being the Sanskrit word for "silence"), wherein one meditates on an inner sound, dubbed the "sound of silence." For example:

> To detect the nada sound, turn your attention toward your hearing. If you listen carefully to the sounds around you, you're likely to hear a continuous, high-pitched inner sound like white noise in the background. It is a sound that is beginningless and endless.
>
> There's no need to theorize about this inner vibration in an effort to figure out exactly what it might be. Just turn your attention to it. If you're able to hear this inner sound, you can use the simple act of listening to it as another form of meditation practice, in the same way one uses the breath as an object of awareness. Just bring your attention to the inner sound and allow it to fill the whole sphere of your awareness. (Amaro, 2012)

It is interesting to note that the modern use of Mindfulness Meditation calls upon such techniques to treat troublesome tinnitus (Roland et al., 2015).

Miscellaneous

Sir Francis Bacon (1561–1626) was a polymath, and celebrated as a statesman, scientist, and philosopher. In his posthumously published book *Sylva Sylvarum or a Natural History in Ten Centuries*, which compiled many of his writings, he mentions a personal experience of tinnitus in the context of exposure to intense sound and temporary threshold shift:

> A very great *Sound,* neare hand, hath stricken many *Deafe;* And at the Instant they have found, as it were, the breaking of a Skin or Parchment in theire Eare: and myself standing neare one that *Lured* loud, and shrill, had suddenly an Offence, as if somewhat had broken, or been dislocated in my *Eare;* And immediately after, a *loud Ringing;* (Not an ordinary Singing, or Hissing, but far louder, and differeing;) so as I feared some *Deafnesse.* But after some half Quarter of an Houre it vanished.

Following the introduction of tobacco to Europe it became very popular, and many benefits were described, including a reduction of tinnitus, in the work "Hymnus Tabaci: A Poem in Honour of Tobacco" (Thorios, 1651):

> The swelling of the *head* it drives away,
> And *bribes* the *Ears musicians* not to *play.*

A footnote indicates that the "Ears musicians" refers to "A whistling or singing in the head."

In a 1675 verse play (Aureng-zebe, Act II), John Dryden (1631–1700) describes tinnitus associated with a state of shock:

> My ears still ring with noise; I'm vext to death:
>
> Tongue-kill'd and have not yet recovered breath.

Jean-Jacques Rousseau (1712–1778) was an influential 18th century political philosopher. In "Confessions"(1782) he wrote:

> A great noise started up in my ears, a noise that was triple or rather quadruple, com-

> pounded of a low and muffled humming, a softer murmuring as though of running water, a piercing whistle. . . . This internal noise was so loud that it robbed me of the keen ear I had previously enjoyed and made me, not completely deaf, but hard of hearing . . . in spite of the throbbing in my arteries and the humming in my ears, which since that time, some thirty years ago now, have never left me for a moment. . . . The noise was irksome, but it caused me no suffering: it was not accompanied by any chronic affliction, apart from insomnia at night. (Rousseau, 1782, p. 222)

This description has a number of interesting aspects. First, the tinnitus described is complex, consisting of a variety of different sounds, some pulsatile. Second, the tinnitus is described as interfering with hearing, and finally, there was "no suffering"—Rousseau has tinnitus for sure, but no distress.

A breakthrough in the ability to read Egyptian hieroglyphics occurred with the discovery of the Rosetta Stone, and Jean-Francois Champollion (1790–1832) was instrumental in that process, though the pressure of work led to major health problems, including tinnitus. He died of a stroke at age 41 years. The book, *The Keys to Egypt* (Adkins & Adkins, 2001), contains this vignette:

> Depressed and feeling more and more unwell, Champollion now wrote to his brother about the damage done by the strain of work: "My poor head hurts, my tinnitus, the humming and buzzing noises, has worsened and leaves me neither day nor night. I have frequent spasms and am incapable of occupying myself seriously for more than a quarter hour . . . " (Adkins & Adkins, 2001)

More recently, in a moving and reflective book entitled, *The Train in the Night—A Story of Music and Loss*, music critic Nick Coleman (2013) describes his experience of a sudden unilateral sensorineural hearing loss, imbalance, and severe tinnitus. While his vestibular problems abated, Coleman relates how the single-sided deafness removed his ability to hear what he calls the "architecture" of music, rendering it flat and meaningless. He describes his tinnitus in emotional terms:

> It was the hullabaloo in my brain, which was getting louder and more insistent, more confident, like the bully who sees that resistance is dwindling and takes larger and larger handfuls of what he can get, because he can. (Coleman, 2013)

"Like the bully . . ." is a poignant description of tinnitus, implying some malicious intent on behalf of the percept—and patients may mention similar views.

Film

In the comedy-drama film *Hannah and Her Sisters* (Allen, 1986), Woody Allen (who wrote and directed the film) plays a character named Mickey who is the ex-husband of the eponymous Hannah. Mickey is a troubled character, with severe hypochondria. When he develops a unilateral hearing loss and tinnitus he becomes very concerned and seeks medical advice. He sees an ear, nose, and throat (ENT) doctor, and the film shows Mickey undergoing a tuning fork test (Figure 1–2). The asymmetry in his hearing is thus confirmed, and further investigation is performed in the form of auditory brainstem responses and a computed tomography (CT) scan (both shown in the film), presumably to exclude a vestibular schwannoma. This possible diagnosis is very distressing for the character, and his relief when given the all clear is immense. This leads Mickey to an existential crisis, and to re-evaluate his life, drawing him to spiritual exploration (Catholicism and Krishna) and

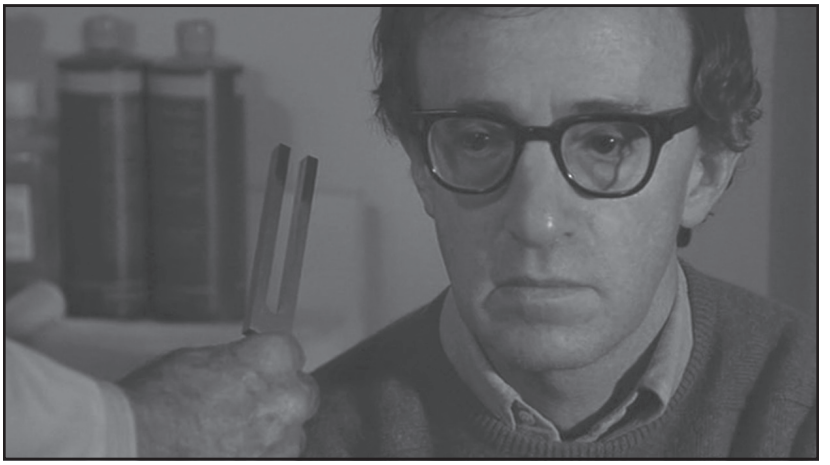

Figure 1–2. Woody Allen undergoes a tuning fork test in *Hannah and Her Sisters* (Allen, 1986).

eventually to a relationship with his ex-wife Hannah's sister Holly. While the film contains other story lines, Mickey's journey is a major element in the humor and warm-hearted feel of the film. Tinnitus is portrayed as a significant symptom, the film indicating that it can be a substantial burden for an individual.

In the film *Children of Men* (Cuarón, 2006), which is based on a novel by the English writer P.D. James (1992), tinnitus is described as "hair cells dying" by the (female) character Julian, talking with the lead male character Theo who has been exposed to a close-proximity blast. The plot addresses worldwide male infertility, leading to a dystopian population crisis, and the search for the one woman in the world who is pregnant. The book and film are rich with religious overtones and metaphors, and some critics have noted that in the film tinnitus is used as a metaphor for loss (see http://www.criticalcommons.org/Members/thecinefiles/clips/julians-monologue-about-tinnitus-from-children-of).

Another film explosion leading to tinnitus is found in the movie *Hunger Games* (Ross, 2012), and in the book on which the film is based (Collins, 2009). One of the main characters is a young woman named Katniss Everdeen, who in the course of one of the eponymous contests is exposed to a blast injury. Completely deaf in her left ear, Katniss is comforted by ringing tinnitus in her right ear, which she believes is indicative of some hope of hearing recovery, which proves to be the case.

The technique of playing high-frequency sounds in a film soundtrack following an explosion to emphasize the intensity of the explosion (as in *Saving Private Ryan*, Speilberg, 1998) has been named "the tinnitus trophe" by Haddon (2015), who cites 33 instances. While not having time or energy to check this claim, it is interesting that this has become a standard technique in film soundtracks.

While not explicitly mentioned, the film *The Passion of the Christ* contains an auditory representation of tinnitus. Directed by Mel Gibson (2004), the film portrays the arrest, trial, and crucifixion of Jesus, and depicts events in a particularly bloody and graphic manner. The story follows the account given in the Gospels, fusing the narrative of John

with the synoptic gospels (Matthew, Mark, and Luke), and close to the opening of the film the arrest of Jesus is depicted (John 18). Simon Peter draws his sword and strikes off the ear of the High Priest's servant (unnamed in the film, but the Gospel of John names him as Malchus [John 18:10]). As he does so the film soundtrack contains a strident high-pitched note, which resolves as Jesus heals and restores the ear to its owner.

Music

The story of the deafness of Beethoven is widely known, but the Czech composer Smetana (1824–1884) also became profoundly deaf (aged 50) and suffered with tinnitus. He wrote his tinnitus into a composition: during the last movement of his String Quartet No. 1 (1876), "From My Life," the second violin, viola, and cello start by playing bass notes quietly. After two bars, the first violin in contrast plays a striking, high-pitched note (a high E) which is sustained for six bars. The other instruments then start playing louder, but despite this crescendo, the high-pitched note cannot be drowned out. The passage in question is illustrated in Figure 1–3.

Given the association between amplified music and tinnitus (Chapter 8) there are surprisingly few mentions of tinnitus in popular song. In an outtake from "Blood on the Tracks" eventually released as part of his Bootleg Series, Bob Dylan (1974) describes hearing tinnitus in the song "Call Letter Blues":

> My ears are ringin'
> Ringin' like empty shells
> Well, it can't be no guitar player
> It must be convent bells

(the sound of church bells having been mentioned earlier in the song as an image of loneliness and melancholy).

The British performer Steven Adams, then recording with the Broken Family Band, mentions tinnitus in his song "John Belushi" (2005). In the song he expresses reasons for not taking drugs that evening, singing

> Don't feel like getting high this time
> You go ahead I just need to rest my mind

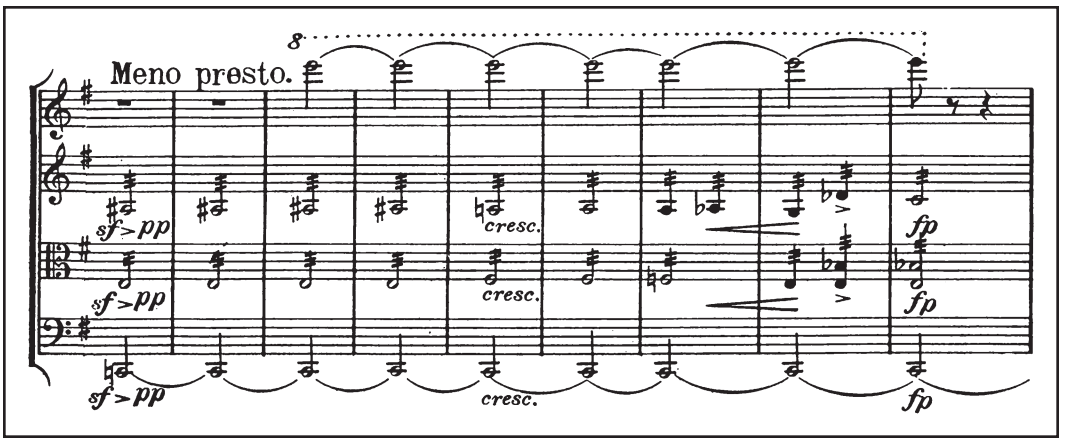

Figure 1–3. An excerpt from the score of String Quartet No. 1 by Smetana (1876) illustrating the strident high-frequency representation of tinnitus.

> I don't need that buzzing in my ear
> I listen to my heart and baby it's bringing a tear to my eye

Further in the song "Man Down," recorded solo, Adams (2014) sings,

> I sleep with my ears still ringing,
> it's a comforting sound.

and while most people with tinnitus would say the opposite, some do express this idea.

In lists of musicians with tinnitus, Thom Yorke from Radiohead often features prominently. The band released two versions of their song "Stupid Car" in 2009: one being a straightforward take and the other a "Tinnitus Mix," with a continuous white noise underlying the music.

Some artists use tinnitus imagery in their song titles. Examples include the lovelorn song "I've got all this ringing in my ears and none on my fingers" by Fall Out Boy (2006), and the impressively titled instrumental "Spectrograph reading of the varying phantom frequencies of chronic, incurable tinnitus" by Man or Astro-Man? (2000).

There is also a band named Tinnitus, formed in the United States in 1996, and playing hardcore punk to this day. They took their name from a quote in the film *Singles* (directed by Cameron Crowe, 1993), namely, "Sorry, I can't hear you . . . I have tinnitus, you know, club disease."

While not an explicit depiction of tinnitus, some music deliberately uses dissonance and noise, and it is paradoxical that when such sensations are perceived as tinnitus they evoke aversion and distress, but for some that percept as music is compelling. Examples would be the dissonant classical music of Messiaen, or the feedback sounds in the music of the Jesus and Mary Chain (who famously enjoyed 24 track recording as it allowed 4 tracks for instruments, and 20 tracks for feedback). There is an analogy with the patient deeply concerned about symptoms of imbalance, but whose friends and acquaintances are delighted to ride on the fairground rollercoaster. The patient experiences involuntary and long-term sensation, whereas the listener or fairground rider has voluntary control, and the experience will be transitory.

Hyperacusis

References to hyperacusis (decreased sound tolerance) in literature are sparse. In the novel *The Woman in White* (1860), Wilkie Collins uses hyperacusis as a plot device. The heroine of the noel, Laura Fairlie, is the niece and ward of Frederick Fairlie, who fails to protect her after she is orphaned, and she falls prey to the scheming Count Fosco. Mr. Fairlie's situation is that he has collapsed sound tolerance, telling Laura when he meets her for the first time:

> Pray excuse me. But could you contrive to speak in a lower key? In the wretched state of my nerves, loud sound of any kind is indescribable torture to me. You will pardon an invalid?

Hyperacusis is not named, and in fact the word was first used clinically in 1938 (Perlman, 1938), but is indicated here, and Mr. Fairlie's life as a recluse means that he cannot be the protector that Laura requires.

Temple Grandin is a U.S.-based professor of animal science and has written extensively about her experiences as a person on the autistic spectrum. This includes descriptions of her variable and often reduced ability to tolerate sound, describing her hearing as follows:

> My hearing is like having a hearing aid with the volume control stuck on "super loud." It is like an open microphone that picks up everything. I have two choices: turn the mike on and get deluged with sound, or

> shut it off. Mother reported that sometimes I acted like I was deaf. Hearing tests indicated that my hearing was normal. I can't modulate incoming auditory stimulation. (Grandin, 1992)

Her experience is one of the auditory environment being intensely loud, but also over-complex:

> I still dislike places with confusing noise, such as shopping malls. High-pitched continuous noises such as bathroom vent fans or hair dryers are annoying. I can shut down my hearing and withdraw from most noise, but certain frequencies cannot be shut out. It is impossible for an autistic child to concentrate in a classroom if he is bombarded with noises that blast through his brain like a jet engine. High, shrill noises were the worst. A low rumble has no effect, but an exploding firecracker hurts my ears. As a child, my governess used to punish me by popping a paper bag. The sudden, loud noise was torture. (Grandin, 1992)

Autobiographic accounts like this help us understand the auditory world of an autistic person. It seems that for some the sound tolerance issues are variable, unlike general hyperacusis, and that there is an element of hyperreactivity to novel sound events, there being some neuroscientific evidence to support this (Gomot et al., 2008). The description of "shutting down" from sound may explain why problems with hearing are often an early complaint in a child's journey to a diagnosis of autism. Much more research is needed in this area.

Conclusion

While I have noticed a number of mentions of tinnitus and hyperacusis in the arts, these are not huge in number and are potentially at odds with epidemiological data that suggest that 5% of the adult population have moderate to severe distress with tinnitus (reviewed in Baguley, Andersson, McFerran, & McKenna, 2013). There are two possible factors at work here. The first is that the literature informally surveyed predates the industrialization of the developed world, and it may be that is only relatively recently that tinnitus has become this prevalent, due to noise exposure. The second factor may be that tinnitus is generally viewed as a rather banal experience, and so is no more likely to receive a literary reference than doing the washing up, or cleaning the bathroom, or other nonspecific symptoms such as a headache or a stomach ache.

However, there are ways in which the literary and artistic references to tinnitus and hyperacusis resonate with the experiences described by patients. These are summarized in Table 1–1. In a sense, considering these artistic references reminds us as clinicians of the massive heterogeneity of responses to tinnitus in the population, and not to oversimplify the situation.

While not a common approach, some clinicians dealing with tinnitus (and latterly hyperacusis) have used art and literature in therapy. Palm and Goebel (1998) described the use of art therapy in "chronic complex tinnitus" wherein patients draw images of themselves or their context, or an abstract image, to express their situation, and an art therapist (from a psychologist background) helps the patient interpret the unconscious feelings and cognitions they have expressed. As tinnitus therapy continued, the art therapy was used to indicate improvements in mood and coping. The technique of drawing pictures has been used in children with tinnitus (Edwards & Crocker, 2008) and hyperacusis (Baguley, 2014). Goebel (1998) also described patients writing letters to their tinnitus, arguing with it, complaining about it, and bemoaning their situation, but as therapy took hold, describing improvements over time.

Table 1–1. Links Between Patient Reported and Artistic Representations of Tinnitus

Tinnitus Characteristic	Title	Author/Composer (date)
Relentlessness	*A Pair of Blue Eyes*	Hardy (1875)
Formless, pseudohallucination	*Money*	Amis (1985)
Interference with hearing	*Harry Potter and the Half Blood Prince*	Rowling (2005)
Sound therapy	Babylonian Talmud	Unknown
Tinnitus appears antagonistic	*Train in the Night*	Coleman (2013)
Association with health anxiety	*Hannah and Her Sisters*	Allen (1986)
Tinnitus overcoming other sounds	String Quartet No. 1	Smetana (1876)
Comforting	Man Down	Adams (2014)
Social isolation and hyperacusis	*Woman in White*	Collins (1860)
Cochlear damage/degeneration	*Children of Men*	Cuarón (2006)
Blast-related tinnitus	*Hunger Games* *Saving Private Ryan*	Collins (2009) Spielberg (1998)

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16

Musical Hallucinations

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Case Study

An 80-year-old diabetic gentleman presented to hospital with reduced conscious level. He was found to have elevated blood glucose concentration, and the rest of the physical examination and laboratory blood analyses were normal. After correction of his blood glucose with insulin infusion, he recovered back to his normal level of functioning and was suitable for early discharge with an escalation of his regular treatment for glycemic control.

His admission medications included amisulpride, an atypical antipsychotic. When asked, the patient reported that he was taking this because he had previously suffered musical hallucinations.

He had long-standing significant sensorineural hearing loss but had always refused to wear his hearing aids because he found them uncomfortable and unhelpful. A few years ago, he was his frail wife's main carer. This consumed the majority of his time, such that he rarely had any other social contact. One day he began to sing "God Save the King." At the time, he was under the impression that he was joining in with his neighbor. He thought this a little odd, as the contemporary monarch was female, but ascribed this discrepancy to his neighbor's well-known eccentricity. It was only when he began to hear a brass band playing that he realized he was experiencing hallucinations. The band played mainly old favorites that he had known since his youth, and especially, "She'll Be Coming 'Round the Mountain." He quite enjoyed the music and made no attempt to contact medical services until a few weeks later when he "made the mistake of mentioning it to [his] daughter." She insisted that he saw a psychiatrist, and amisulpride therapy was commenced on the first visit. After this, he reported that the volume of the musical hallucinations was reduced, but their frequency and character were unchanged.

When his wife passed away, he moved to live with his daughter. He rapidly reconnected with his social circle and engaged in a number of new group activities, such as lawn bowls. Over time, he noticed that the music gradually stopped. He says that he misses it in some ways but does not particularly want it back. We withdrew his amisulpride, and his musical hallucinations did not recrudesce.

Definition and Phenomenology

Hallucinations are perceptions with a compelling sense of reality experienced but without causal external stimuli. This compelling sense of reality is what distinguishes musical hallucination (MH) from simply having a tune "stuck in your head," a phenomenon known as an *earworm* (Beaman & Williams, 2010). It is not necessary for the sufferer to be unable to distinguish MH from heard external music (this can be done on the basis of content, context, or timbre) but, as in the case description, this confusion is relatively common at symptom onset. It is not uncommon for patients to be referred to medical services for MH after an unproductive encounter with police or local council services regarding their neighbors' antisocial music listening habits. In extreme causes, this has led to retaliation to the perceived sound. This is perhaps compounded by the observation that MH is more likely to first arise at times of stress or emotional distress.

MH can be experienced as originating from either the outside world or internal to the head (*pseudohallucinations*), and, as with other types of auditory hallucinations, this distinction appears to have no clinical or mechanistic significance (Copolov, Trauer, & Mackinnon, 2004). Some patients are able to consciously manipulate their MH, most commonly by "changing the tune," but this ability is not universal, and it is rare to be able to voluntarily suppress MH. External music has a variable effect on MH, with some patients reporting that external music suppresses internal music, while others report no effect or even that their MH volume increases in competition. This effect can persist for some time after the offset of the external music, a phenomenon also often present in tinnitus and known as *residual inhibition* (Feldmann, 1971; Kumar et al., 2014; Roberts, 2007).

The content of MH is usually familiar simple melodies heard regularly in youth, particularly often experienced as repeated short fragments of a longer hymn or carol (Griffiths, 2000; Warner & Aziz, 2005). Although lyrics are sometimes heard, it is important to emphasize that MH is different from verbal hallucination in its phenomenology and demographics, and has different neural correlates (Izumi, Terao, Ishino, & Nakamura, 2002). It differs from tinnitus in that it is a disorder of complex sound processing with emotive content, rather than of elementary percepts, and is much less common (Cole, Dowson, Dendukuri, & Belzile, 2002). In a clinical context, specialists in audiology and ENT working regularly with tinnitus commonly classify MH as a subset of tinnitus, while psychiatrists and neurologists generally see it as a distinct condition. This reflects to some extent the treatment strategies employed; those who view MH as a form of tinnitus commonly treat it in the same way, while neurologists and psychiatrists might generally be more likely to employ pharmacological interventions. The relative merits of these approaches and the evidence base for each are discussed later in this chapter.

Prevalence

There have been no large studies of prevalence of MH in unselected populations, but several have evaluated those who might be deemed "at risk." The largest study to date reported a very low prevalence (0.16%) in a sample of 3,678 admissions to a general psychiatric hospital (Fukunishi, Horikawa, & Onai, 1998). Of the six cases reported in this study, five were female, three used hearing aids, and three were elderly. Even in elderly individuals with hearing impairment, the group generally thought

at highest risk, prevalence in small samples varies from 0.8% (Cole et al., 2002) to 3.6% (Teunisse & Olde Rikkert, 2012).

Incidence

Studies of MH incidence are complicated by the lack of a clear referral speciality. Patients with MH are seen for the first time by neurologists, psychiatrists, audiologists, and ENT surgeons (Stewart, von Kriegstein, Warren, & Griffiths, 2006), and no study to date has attempted to monitor referrals through all of these routes. The largest study to date, in a Welsh old-age psychiatry service, estimated that one new clinically significant case occurs per 10,000 people per year (Warner & Aziz, 2005), which would make MH approximately five times more common than vestibular schwannoma (Stangerup et al., 2010) and five times less common than clinically significant tinnitus (Martinez, Wallenhorst, McFerran, & Hall, 2014). As in the case description, many of the patients in this study reported that their MH was not distressing, perhaps suggesting that further undeclared cases exist in the community.

Demographics

MH is more common in females, with a ratio of about 3:1 (Evers, 2006; Pasquini & Cole, 1997), even accounting for the fact that females live longer and are more likely to live alone (Berrios, 1990). This gender bias is far greater than that seen in other forms of auditory hallucination and tinnitus, even in the elderly (Cole et al., 2002).

The incidence of MH rises with age until 60 years (Evers, 2006), but there is no convincing evidence of a further increase beyond this. It is unclear whether age is an independent risk factor, or whether it is merely a surrogate marker for hearing loss, social isolation, polypharmacy, and vascular or other neurological pathology.

Risk Factors

There is no doubt that socially isolated individuals are more likely to experience MH. It is reported that 77% of sufferers live alone (Warner & Aziz, 2005), at least twice as many as would be expected from age and gender alone.

Hearing loss is frequently reported in association with MH (Hammeke, McQuillen, & Cohen, 1983; Klostermann, Vieregge, & Kompf, 1992), and is present in 40% to 70% of reported cases (Berrios, 1990; Evers & Ellger, 2004). It is unclear to what extent this represents reporting bias, as a lower percentage was reported in psychiatry cohorts (Warner & Aziz, 2005). *Deafness* in this context is variably defined, and it is difficult to relate to the prevalence of hearing loss in matched unaffected populations. While MH most commonly occurs years after the gradual onset of hearing loss, it has been reported acutely after sudden hearing loss due to surgical intervention (Iijima, Maeda, Tsubouchi, & Iwamoto, 2000) and gentamicin toxicity (Tanriverdi, Sayilgan, & Ozcurumez, 2001). Because of this, MH has been described as the auditory equivalent of the Charles Bonnet syndrome, in which visually impaired individuals experience complex, well-defined hallucinations (Menon, Rahman, Menon, & Dutton, 2003). While similarities certainly exist, and these conditions have been reported to coexist (Warner & Aziz, 2005), there are also phenomenological and prognostic differences (for a review, see Cope & Baguley, 2009).

The Contribution of Psychological Pathology

Depression is present in about 1 in 3 elderly sufferers of MH (Aizenberg, Modai, Roitman, Mendelson, & Wijsenbeek, 1987; Pasquini & Cole, 1997). It seems likely that, in most cases, depression is primarily acting to increase social isolation, although there are case reports of MH in association with psychotic depression (Izumi et al., 2002) in which it can sometimes be a soothing, compensatory phenomenon (Fukunishi et al., 1999).

Individuals with obsessive-compulsive disorder have approximately a 40% lifetime risk of developing MH (Hermesh et al., 2004), with earworms (involuntary repetitive musical imagery) being even more commonly experienced.

It is important to stress that isolated MH is not associated with schizophrenia, in which auditory verbal hallucinations are common but MH is rare (Hermesh et al., 2004). In cases of schizophrenia in which MH is experienced, it is usually a transitory phenomenon and is often congruent with overriding delusional beliefs (Baba, Hamada, & Kocha, 2003; Saba & Keshavan, 1997) or associated with obsessive-compulsive symptoms (Bleich-Cohen, Hendler, Pashinian, Faragian, & Poyurovsky, 2011).

Secondary MH

Rarely, MH can be the manifestation of underlying organic brain disease. There does not appear to be a critical brain region for a lesion to lead to MH, reflecting the network-level organization of brain areas responsible for analyzing the complexities of pitch, rhythm, timbre, recognition, and emotion (Stewart et al., 2006); cases of MH associated with lesions at every stage of the auditory pathway from brainstem (Cascino & Adams, 1986; Murata, Naritomi, & Sawada, 1994) to auditory association cortex (Musiek et al., 2007) have been reported. Similarly, the cause of the lesion is unimportant, with MH reported in the context of both ischemic (Inzelberg, Vishnievskaya, & Korczyn, 1993) and hemorrhagic (Murata et al., 1994) infarction, intracranial aneurysm (Roberts, Tatini, Zimmerman, Bortz, & Sirven, 2001) and tumor (Keshavan, Kahn, & Brar, 1988; Nagaratnam, Virk, & Brdarevic, 1996).

MH is rare as a feature of epilepsy but has been reported in partial seizures, most often as an aura (Couper, 1994; Rennie, 1964).

There is an association between MH and vascular insufficiency (Gilhuis, Dara, & Renier, 2007) and Alzheimer's disease (Mori et al., 2006), but causality is not clear.

MH has been reported in the context of a wide variety of medications (for a review, see Cope and Baguley, 2009), but only opioids have a strong and clear association (Davies & Quinn, 2005; Keeley, Foster, & Whitelaw, 2000; Moore, 2003). Both central and cochlear contributions to this phenomenon have been proposed, and all three opioid receptor types are present in the cochlea (Jongkamonwiwat et al., 2003).

Idiopathic (Primary) MH

In common with tinnitus, MH principally occurs in the presence of sensorineural hearing loss, and thus can be understood, at least in part, as a disorder of deafferentation (loss of auditory input), which in turn leads to spontaneous central activity that is interpreted by the brain as sound. However, for MH to occur, additional processing is required for

the emergent auditory percept to take the form of music as opposed to a simpler percept. In keeping with this idea, resting-state electroencephalography (EEG) shows similar resting-state abnormalities of brain activity in tinnitus and MH patients compared to controls (Vanneste, Song, & De Ridder, 2013), including a characteristic pattern of increased activity in core auditory cortex. Further, there are additional abnormalities in MH patients in auditory association, frontal and limbic areas, which likely relate to the musical content of the percept and additional demands on auditory memory. Positron emission tomography (PET) has highlighted a similar network of brain areas, activated in proportion to the perceived intensity of hallucinations (Griffiths et al., 2000). In addition to the areas already mentioned, this network included cerebellum and basal ganglia. Activity in primary auditory cortex did not increase with hallucination intensity, suggesting that hyperactivity in this area may be a precursor for MH, but that the processes critical for perception of the hallucinated music occur within auditory association and extra-auditory areas. Similarly, magnetoencephalography (MEG) recorded while using residual inhibition to modulate hallucination intensity demonstrated that short-term changes in hallucination intensity associated with strong brain activity changed in only two main brain areas: auditory association cortex and precuneus, a key global perceptual center (Kumar et al., 2014). Thus, even though an extensive brain network was linked to MH, the interplay between auditory association cortex and higher perceptual centers was a key process responsible for the perception of music. These particular centers might thus comprise a recurrent and recursive loop, in which musical representations are generated in the auditory part, and are reinforced and maintained by predictive influences from higher perceptual and memory centers.

It may well be the relatively predictable structure of music (compared to speech or other environmental sounds) that makes it the ideal subject of complex hallucination; in this manner, predictions reinforce percepts, which go on to further reinforce the (musical) predictions. Ordinarily, input to this loop from primary auditory cortex would override the erroneous perception of music, but in the presence of a compromised auditory input (due to hearing loss), the system might use the predictable structure of the music perception to become stable and self-reinforcing. This could explain how MH persists once it is established, but there remains debate about how the hallucinated music initially becomes sufficiently established to be self-reinforcing. In some cases, MH is described as slowly developing out of tinnitus or other rudimentary sounds into familiar melodies, suggesting it may start due to small fluctuations in the patient's perceptual state and build from there. In other cases, the hallucinations begin fully formed, and it is less clear whether such a recursive buildup of the phenomenon occurs at its onset. Although MH can occur due to no other lesion than hearing loss (resulting in peripheral deafferentation and concomitant changes to the auditory pathway), there is a well-recognized overrepresentation of focal brain lesions, including small vessel disease, in MH patients compared to matched controls (Berrios, 1990). Therefore, certain focal brain disease may either further predispose people to MH, or could even trigger its onset; the importance, in some cases, of "central deafferentation" due to such lesions has been previously highlighted (Evers, 2006). Disruption of brain structure or function may lead to deficits in input to musical centers in association auditory cortex, and therefore make them more susceptible to influence from higher-order predictive influences that reinforce the perception of music.

Proposed Neural Mechanisms of Primary MH

The means by which this recurrent loop of communication between higher areas (auditory association areas and precuneus in the above case) is established is based on the way the brain manages uncertainty of the environment in its normative functioning. When confronted with incongruous inputs, the brain estimates not only the causes of sensory input but also the reliability (or precision) of those causes. Mechanistically, this calculation corresponds to postsynaptic gain of a population of neurons in a brain area. The extent to which activity from a lower (sensory) area updates the expectations generated at the higher (associative) depends on this gain (or estimated precision): higher gain in the sensory areas allows sensory input to access the higher areas, thereby correlating expectations to the sensory input.

With hearing loss, hearing becomes less reliable, and the brain responds by recalculating the precision of sensory information. This model of MH proposes that, in reestimating the precision of sensory information following hearing loss, the brain devalues the precision of the sensory areas, which results in a tilt of balance of precision such that higher centers are recognized as relatively more precise than lower sensory areas. A consequence of the lower precision in the sensory areas is that communication between the higher areas is decreasingly informed by the bottom-up sensory information (because of the low postsynaptic gain in the sensory areas, i.e., primary auditory cortex and areas lower in hierarchy to it).

Investigations

Because of the strong association between MH and hearing loss, measurement of audiometric thresholds is always indicated as part of a patient's initial examination. Similarly, because MH can be a manifestation of structural brain disease, some form of brain imaging should be performed, although this is usually unrevealing. There is insufficient evidence to make a strong recommendation on whether this should take the form of contrast-enhanced computed tomography (CT) or magnetic resonance imaging (MRI). More intensive investigation for organic brain disease should be triggered by transient visual disturbances, dizziness, severe headache, abnormal speech or neurological examination, or an audible carotid bruit.

EEG should only be performed if there are other clinical grounds on which to suspect epilepsy, as nonspecific abnormalities are present in more than 1 in 10 healthy elderly individuals (Leuchter, Daly, Rosenberg-Thompson, & Abrams, 1993).

Mental state examination should be part of the initial assessment of MH, given the strong association with depression, and consideration should be given to whether disorders of mood are likely to be cause or consequence of the MH.

Treatment Strategies

Treatment of MH depends on its underlying drivers. When they are present, the cornerstones of active intervention are the correction of hearing loss, reduction of social isolation, and treatment of low mood with psychological and pharmacological strategies (Pasquini & Cole, 1997; Wengel, Burke, & Holemon, 1989). Similarly, epilepsy-associated MH has been successfully treated with anti-epileptics (Terao & Tani, 1998) and drug-induced MH by drug withdrawal (Davies & Quinn, 2005; Moore, 2003).

A common concern among patients is that MH is a sign of dementia or psychosis,

especially when they have suffered embarrassment having complained to local services about perceived antisocial behavior. It should be stressed that this is not the case, and an explanation of the underlying mechanisms provided. MH is often not distressing and, once they understand that their symptoms are not harmful and do not represent a serious underlying pathology, patients are often content to accept ongoing musical experiences.

If pharmacological therapy is desired, and no underlying treatable cause is present, the literature most commonly reports success with antipsychotics (Gilhuis et al., 2007) and anticholinesterases (Ukai, Yamamoto, Tanaka, Shinosaki, & Takeda, 2007), but there are no randomized controlled trials, and, as in the case description, benefit from these drugs can be disappointing and side effects significant.

Future Directions

Some of the fundamental questions that could guide the future direction of research in MHs are (a) why hearing loss is more likely to produce hallucinations of music and not of any other auditory percept; (b) why hearing loss in some people, but not in others, leads to MH; and (c) what explains the different phenomenologies of MHs. Although each of these questions can be studied in isolation, the availability of a single conceptual framework within which specific hypotheses about different aspects of MH can be empirically tested would help accelerate the progress in research in MH. One such framework based on predictive coding was proposed by Kumar et al. (2014) and is described in the section on neural mechanisms above. Developing further on this, we now give one example of how recent evidence for acetylcholine-based pharmacological intervention as a treatment option (Blom, Coebergh, Lauw, & Sommer, 2015) for MH could be explained within this framework. As described in the previous section, reduced precision in the ascending sensory pathways following hearing loss contributes to the development of MH. It therefore follows that treatments that boost sensory precision should provide relief from MH. Evidence from animal neurophysiology (Goard & Dan, 2009) and computational modeling (Moran et al., 2013; Yu & Dayan, 2005) suggests that the neuromodulator acetylcholine enhances reliability of representations in the primary sensory cortex (i.e., the sensory precision). This provides a putative mechanism of action for cholinesterase inhibitors to treat MH by increasing the availability and duration of action of acetylcholine in the sensory cortex. However, it is clear that these drugs are not effective for all patients with MH, and further work is needed to account for these individual variations.

A few studies in the past have tested the efficacy of neurostimulation using transcranial magnetic stimulation (TMS) as a treatment option for hallucinations (for a review see Moseley, Fernyhough, & Ellison, 2013). In particular, pilot studies on a pattern of stimulation called *continuous theta bust stimulation* (cTBS) have provided early promise for this technique (Plewnia, Zwissler, Wasserka, Fallgatter, & Klingberg, 2014). However, all studies to date have concentrated on hallucinations in psychiatric disease, and none have looked at MH. Future studies should explore this area because neurostimulation, if successful, not only has the benefit of providing relief to the patients but also has the potential to illuminate the neural and cognitive mechanisms underlying MH.

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