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Editor-in-Chief

Prof. Henryk Skarzynski, M.D., Ph.D., Dr. h.c. multi

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Journal of Hearing Science®

Scope and Purpose: **Journal of Hearing Science®** is a peer-reviewed scientific journal that publishes original contributions to knowledge in all areas of **Otolaryngology**, **Audiology**, **Phoniatrics**, and **Rhinology**. Its primary mission is to offer an international forum for professionals; a secondary aim is to assist hearing practitioners by providing important knowledge helpful to patients with hearing, voice, speech, and balance disorders. J Hear Sci has a distinguished Editorial Board, ensuring that the journal produces multidisciplinary papers of the highest quality. The broad international membership promotes fair and thorough assessment. The journal is an open access type of publication which allows all readers free access to articles. We impose no publication fees or page charges.

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Dear colleagues,

I invite you to read the second number of the *Journal of Hearing Science* for 2021. The issue starts with two review papers, one devoted to how deaf children with cochlear implants develop a theory of mind – that is, how they model and react to other people as thinking beings – and the second to endoscopy of the cerebello-pontine angle, which is important for surgical treatment of certain hearing problems. The next interesting paper belongs to a special section of our journal devoted to hypothesis papers, where contentious areas of auditory research are discussed. This paper deals with 'type C' tympanograms, which occur when a tympanometer displays a negative middle ear pressure. Conventionally, this is interpreted as gas absorption in the middle ear cavity, but as the title of this challenging paper suggests, this cannot always be the case, and perhaps we are seeing the result of contraction of the middle ear muscles. No doubt this paper will provoke lively discussion.

At this point I would like to encourage our field to submit more hypothesis-style papers, for I believe there is an important role for scientific debate as well as experimental investigation. At least in our disciplines of otorhinolaryngology and audiology there is a scarcity of letters to the editor and hypothesis papers. In my view, such pieces make important contributions to scientific progress and should be encouraged.

The issue follows with research papers related to some musical aspects of hearing and also to an evaluation of tinnitus patients and what therapy might be most appropriate. The issue concludes with papers related to vestibular potentials in migraine patients and a case study of evaluation of speech in a case with dysarthria. The papers related to music are especially close to my heart as for the seventh time we are organizing the International Music Festival for Children, Youths, and Adults with Hearing Disorders called "Beats of Cochlea" (https://festiwal.ifps.org.pl/en/). One part of it will be an International Scientific Conference "Music in Human Auditory Development" and various workshops. It will conclude with a concert that will feature performances by people from Poland and elsewhere, as well as groups of musicians, music therapists, and workshop instructors. The festival will be broadcast online, and I invite you to tune in between the 12th and 15th of July. Afterwards, a recording of the concert will also be made available for those who were unable to listen to it live.

With kind regards and greetings,

Prof. Henryk Skarzynski, M.D., Ph.D., Dr.h.c.multi

Review papers

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B Data collection/entry
C Data analysis/statistics
D Data interpretation
E Preparation of manuscript
F Literature analysis/search
G Funds collection

THEORY OF MIND DEVELOPMENT IN DEAF CHILDREN WITH COCHLEAR IMPLANTS: LITERATURE REVIEW

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Abstract

Theory of mind (ToM) is the mental capacity that allows us to represent the mental states (beliefs, desires, emotions) of other people, infer them from situational cues, and predict their behavior. According to the standard view, the most important milestone in ToM development – the ability to pass the false belief test (FBT) – emerges around four years of age. FBT requires one to understand that the beliefs of others are independent from reality and from one's own beliefs, and that their behavior can be predicted by their mental states. Previous research has indicated that deaf and hard-of-hearing children born into hearing families (DoH) are at risk of delayed ToM development due to restricted social interactions. However, these findings are unclear for DoH children who receive cochlear implants (CIs) and whose hearing is partially restored. In this review, we summarize research on the development of ToM in DoH children with cochlear implants (CIs). We describe how language (vocabulary and syntax) influences ToM. Finally, we discuss the nature of social interactions that facilitate ToM development in children with typical hearing as well as in DoH children with CIs.

Key words: theory of mind • deafness • cochlear implants • social developmen

ROZWÓJ TEORII UMYSŁU U DZIECI GŁUCHYCH I NIEDOSŁYSZĄCYCH BĘDĄCYCH UŻYTKOWNIKAMI IMPLANTÓW ŚLIMAKOWYCH. PRZEGLĄD LITERATURY

Streszczenie

Teoria umysłu (theory of mind, ToM) to zdolność poznawcza, która pozwala reprezentować wewnętrzne, nieobserwowalne stany psychiczne (przekonania, pragnienia, emocje) innych ludzi, wywnioskować je ze wskazówek sytuacyjnych i na ich podstawie przewidywać zachowania innych osób. Zgodnie ze standardowym poglądem kamień milowy w rozwoju ToM – zdolność do przejścia testu fałszywych przekonań (false belief test, FBT) – dzieci osiągają w wieku około czterech lat. FBT wymaga zrozumienia przekonań innych osób jako niezależnych zarówno od rzeczywistości, jak i od prawidłowych przekonań obserwatora oraz zrozumienia, że zachowanie ludzi można przewidzieć na podstawie ich stanów psychicznych. Wcześniejsze badania wykazały, że dzieci niesłyszące i niedosłyszące urodzone w rodzinach słyszących (deaf of hearing parents, DoH) są narażone na opóźnienie w rozwoju ToM z powodu ograniczonych interakcji społeczno-komunikacyjnych. Jednak wyniki te są niejasne w przypadku dzieci DoH, które otrzymują implanty ślimakowe (CI), zatem ich słuch może być częściowo przywrócony. W tej recenzji prezentujemy badania dotyczące rozwoju ToM u dzieci DoH, które są użytkownikami implantów ślimakowych (CI). Opisujemy, jak język (słownictwo i składnia) wpływa na rozwój ToM. Na koniec omawiamy naturę interakcji społecznych, które ułatwiają rozwój ToM u dzieci z typowym słuchem, a także u dzieci DoH będących użytkownikami CI.

Słowa kluczowe: teoria umysłu • głuchota • implanty ślimakowe • rozwój społeczny

Introduction

Typically, at around age four, children undergo a striking developmental change in the way they perceive their social world. They start to understand that thoughts and feelings shape human behavior, enabling them to "enter the community of minds" [1]. This ability is called Theory of Mind (ToM) and is considered by developmental psychologists to be the "most important development in early childhood social cognition" [2]. It allows the child to understand that the knowledge and perspective of others can be different from their own [3]. Therefore, ToM is important for successful social functioning and affects many aspects of social life, including peer popularity (e.g. [4]). Moreover, it is worth noting that poor social perception poses adverse risks to mental health.

ToM ability progresses in typically-developing children according to a particular sequence based on age [5]. Around the age of four, children undergo a conceptual change and start to explicitly reason about the mental states of others, which manifests in the ability to pass the traditional false belief test (FBT) [3,6]. The FBT assesses whether the child can predict the behavior of another person, even if the other person is acting on a false belief that differs from objective reality and the child's own knowledge. The FBT is therefore considered to be the "litmus test of Theory of Mind" [7]; however, there are also other methods that assess

ToM ability in infants, children, and adults [8,9]. Furthermore, ToM development is a gradual process, reflected in several milestones that children reach in infancy and later childhood [10].

ToM is a multifaceted concept, requiring the integration of a number of components. It can be broken down into at least two subcomponents: affective ToM (inferring the feelings of another person) and cognitive ToM (understanding the beliefs of another person), which comprise both distinct as well as overlapping neural processes [11]. It has also been suggested that there are additional abilities connected with ToM development – in particular, language skills [12]. Furthermore, the quality of conversations about mental states with caregivers has also been suggested to be an important factor in ToM development [13]. Therefore, deaf children of hearing parents (DoH) – who might have less access to spontaneous mentalistic-type conversations and who frequently show delays in language acquisition – are at risk of slower ToM development [14,15]. At the same time, medical and technological advances provide more options for the treatment of children born with profound hearing loss. These children can benefit from cochlear implants (CIs), which, by restoring a degree of hearing, can allow them to acquire spoken language [16,17]. However, in terms of conversations with their hearing parents, deaf children with CIs might have qualitatively different experiences compared to their peers with typical hearing, since their parents are likely to simplify conversations to adjust to the assumed cognitive ability of their child [18]. Hence, deaf children with CIs can still experience limited conversational input early in life, they can manifest difficulties in some language tasks, and possibly experience delayed ToM development [14].

This line of argument highlights that specific features of the early social interactions with caregivers, along with language development, have important effects on the subsequent development of social cognition, but this remains underexplored in children with hearing loss who use CIs.

The main aim of this article is to review the existing literature on ToM development and assess the environmental factors that might affect mentalizing in profoundly deaf children of hearing parents (DoH) who use cochlear implants. We decided to focus exclusively on DoH children with CIs who use oral language as their preferred mode of communication. The reason is that previous research has shown that deaf children of deaf parents, who communicate in sign language, develop ToM in line with the timetable of children with typical hearing [19,20] due to a typical linguistic experience and access to conversational interaction with their caregivers. However, studies on prelingually DoH children who use CIs are still scarce and provide inconsistent results regarding ToM competencies.

The first section outlines the concept of ToM, including theoretical and methodological frameworks behind its development, ending with the relationship between ToM and language. The following section focuses on profoundly deaf children who use CIs, further expanding on the relationship between ToM, language, and conversation. Finally, we set out research questions that need to be explored by future studies on social cognition development in DoH.

Theoretical and methodological framework of ToM

There are various theoretical accounts that explain the development and mechanisms of ToM and they focus on different aspects of this ability.

According to “theory–theory” accounts, ToM development is similar to building a scientific theory. Children are compared to scientists who, through learning and experimentation, reorganize their theories in order to explain the causal relationships between mental states and behavior [21]. Simulation accounts of ToM posit that it is rooted in a mechanism of adopting the perspective of others and imagining their mental states, relying on a system of mirror neurons [22]. Modular accounts of ToM, such as that of Leslie et al. [23], argue that there is an innate mechanism corresponding to the ability to reason about mental states.

However, ToM could also be viewed in social constructivist terms, as proposed by Carpendale and Lewis [24]. Their account emphasizes the role of social interactions and communication in ToM development. They propose that “children gradually construct social understanding through the regularities they experience in interacting with others” [24]. They also outline the role of language competency and the quality of conversations in the development of a child’s social understanding. Their account corresponds with studies of deaf children of hearing parents, as the delay in development of ToM in this group can be explained by limited conversational input [14].

There are several methods available to study ToM. Task variations might depend on the age of the participant or the ToM system being studied (explicit or verbal; implicit or automatic, nonverbal), since ToM can be divided into two systems which differ in terms of cognitive control and awareness [25]. Some measures also allow to assess how the parents perceive their child’s social understanding [26,27]. The most common method of assessing explicit ToM is the false belief test. One version of this test is the change-of-location task [6]. This paradigm was devised by Wimmer and Perner [28] in the form of a puppet play, and was later modified by Baron-Cohen et al. [29] during their research with children with autism spectrum disorder. In this task, participants observe a scene with two protagonists named Sally and Anne. Sally places a marble in a basket and leaves the room. During her absence, Anne transfers Sally’s marble to a box. When Sally returns, the participants are asked a question concerning the beliefs of the protagonist: “Where will Sally look for her marble?” [29]. In order to pass the Sally–Anne task, participants have to acknowledge the protagonist’s false belief and point to the previous location of the marble (the basket). Studies have shown that typically developing children are able to successfully pass this test around the age of four [6]. Children aged three and younger fail this test and point to the actual location of the object, not accounting for the lack of knowledge of the protagonist [6].

In addition to the change-of-location task, there are also other methods of assessing false belief understanding, such as the unexpected contents tasks (the “Smarties” box task) or unexpected identity tasks [30]. In the unexpected contents task, the child is shown an object with something unexpected inside (e.g., a box of sweets filled with pencils). Next, the child is asked what a child who had not been shown the unexpected contents would say is in the box. To succeed, children have to understand that the other child could not know the truth and would therefore have a false belief. Similarly, in the unexpected identity task, the child is also presented with a surprising object, but the properties of the object are unexpected (e.g., a sponge that looks like a stone) [31]. These tasks require a direct description of another person's mental state and consequently engage an explicit and intentional system of ToM.

Although most ToM studies focus on children, this ability can also be studied in adults. There are special paradigms adjusted for adult studies that also enable more ecological assessments of ToM and focus on the understanding of more advanced concepts, such as metaphors or faux pas [9].

Finally, some measures also assess how caregivers perceive their child's understanding of mental states [26,27]. For example, the Theory of Mind Inventory has been used in studies of typically developing children as well as of children with autism spectrum disorder or children with hearing loss to identify challenges specific to these populations [26,27]. The advantage of this type of measure is that they include everyday manifestations of ToM and take into account parental observations and expertise regarding their child.

Thus, reasoning about mental states can be studied in various ways and in a wide range of participants of different ages, giving measures of the developmental stage of ToM ability.

Development of ToM

Development of ToM can be viewed as a gradual process [5,24]. Passing the standard false belief test around the age of four is a pivotal point for ToM; however, there are several social development milestones that children reach before this point. These skills include joint attention (which is the shared focus of two individuals on an object), the ability to recognize others' emotional states, the knowledge that people act according to their intentions, understanding the causes and consequences of emotions, and pretend play. Moreover, social development also continues as children progress into middle and later childhood when they develop more advanced ToM concepts and can reason about mental states in a more refined manner [5,10].

Infants already demonstrate a considerable interest in social interaction. Even 6-month-olds distinguish between the motion of inanimate and animate objects and "interact dyadically" with objects and people [32]. Around their first birthday, infants undergo what Tomasello (e.g. [32]) has described as a "nine-month revolution", as at this stage they start to engage in joint attention. Joint attention refers to the ability of an infant to share a focus on an object or event with an adult in a triadic interaction. For example, a child might point to a toy on the table to draw the parent's attention or follow the parent's gaze to the toy. Therefore, joint attention is based on the understanding that both the infant and adult share a focus on the object, but from different perspectives [3]. Children develop pretend play around 18–24 months (e.g. [33]). Leslie [33] argued that the ability to engage in a "shared pretense" (e.g., understanding that someone imagines that a banana is a telephone) relies on the same representational mechanism as later understanding of explicit false beliefs. Subsequently, toddlers and young preschoolers are able to understand desires and intentions, which are precursors to false belief understanding [5]. Finally, around the age of four, children start to understand that someone can hold false beliefs about the world; they can then pass the standard false belief test (FBT), which is considered critical in ToM development [6]. There is also evidence that children pass the FBT around the same age across different cultures [34]. The critical age for false belief understanding is a matter of some debate, as the test itself may be too difficult for younger children due to linguistic and executive demands. Thus, simplified versions of the FBT can improve a child's performance [8]. However, a meta-analysis by Wellman et al. [6] supported the claim that a conceptual change in the understanding of beliefs occurs around preschool age, questioning the view that it only appears around this age due to test difficulty. Currently, there is still discussion about whether success on simplified tasks represents early false belief understanding or a distinct competence [3]. Later, children are able to understand that someone can have a belief about another person's belief (known as a second-order belief) [35]. Finally, during later childhood children are able to understand irony, metaphors, and faux pas, which can be described as "advanced ToM" [10,36].

The emergence of ToM concepts during subsequent stages of life supports the idea that this ability is multifaceted and not limited to false belief understanding. Furthermore, this developmental process can be viewed in a much broader context. Linguistic and family factors, such as the child's level of language competency or the frequency and quality of talk about mental states in parent–child interactions, may contribute to variance in ToM [12,13]. Furthermore, progress in ToM might be altered or delayed in populations with atypical language development [5]. Several studies have highlighted the case of deaf children of hearing parents, as these children can display different trajectories of acquiring ToM concepts – mainly false belief understanding – in comparison to their peers with typical hearing (e.g. [5,14,37,38]). These results can be explained in terms of the relationship between language and ToM, which will now be discussed.

Language and ToM

Bretherton et al. [39] have previously emphasized the connection between the emergence of explicit ToM and a child's ability to verbally refer to mental states, suggesting the importance of language development for ToM. Since then, studies have confirmed the existence of a relationship between language and ToM ability – in particular false belief understanding [12,40]. The role of language in ToM development can also be conceptualized in terms of social constructivist accounts, since the quality of everyday conversations has been shown to contribute to a child's later understanding of mental states [13].

There is ongoing discussion on the nature of the relationship between language and false belief understanding. As summarized by Farrar et al. [40], some studies emphasize the role of general language competency, including syntactic and semantic ability (e.g. [41,42]), while others indicate that there are specific aspects of language that might be more important for false belief understanding, such as complement syntax (e.g. [43]). According to the second account, complementation is necessary for the representation of false beliefs [43]. Understanding special structures such as "Sally thinks that the marble is in the basket" might be especially important for false belief understanding, as they reflect a subjective perspective of the situation [40,44]. However, other studies argue that for typically developing children, complementation is not always uniquely related to false belief understanding and point towards the role of general language measures such as semantic or syntactic abilities [40]. General syntax, which includes various syntactic forms, enables the child to represent and track the relations and changes in the classic change-of-location task, thereby contributing to false belief understanding [42,45]. For example, de Rosnay et al. [41] measured children's syntactic understanding with the Test for the Reception of Grammar and found that it was significantly related to their performance on the FBT. Also, according to a meta-analysis by Milligan et al. [12], syntax, among other language measures, was significantly related to false belief understanding.

Moreover, in line with social constructivist accounts, conversations with parents might also be important for the child's ToM development [13,24]. Devine and Hughes [13] found that talk about mental states predicted the child's false belief understanding after one year. Taumoepeau and Ruffman [46] suggested that parents could "scaffold" their child's mental state understanding through the use of mentalistic language. Parental input can be assessed by self-report (e.g. [47]) or, more qualitatively, through the analysis of real-life conversations (e.g. [38,44]). Peterson and Slaughter [47] devised the Maternal Mental State Input Inventory (MMSII) in order to measure a mother's preference for explanations of everyday interactions (e.g., preparing a birthday surprise). They found that mothers who preferred more elaborate explanations with references to mental states had children who exhibited higher false belief understanding than their peers (e.g., by explaining that dad will be surprised with his birthday present because he does not know what is inside the box). Tompkins [44] studied conversations between parents and children during shared storybook reading and also found that references to mental states were positively related to the child's performance on the FBT. Furthermore, a recent meta-analysis of family correlates of ToM by Devine and Hughes [48] also confirmed the impact of parental mental state talk on the development of children's false belief understanding.

Finally, the relationship between language and false belief understanding can be elucidated by studying specific populations in which language development differs from the typical model, such as deaf children raised by hearing parents [49]. The experiential view of cognitive development assumes that the child's language skills affect how much they can access and understand conversations that refer to the mind. Consequently, delayed acquisition of language – which is frequent in deaf children of hearing parents – might hamper the later development of ToM [14,20]. On the contrary, deaf children who use sign language and whose parents are native signers do not show delays in explicit ToM, as their experience in conversations has not been constrained [20].

Language and conversation in deaf children with CIs

In deaf children of hearing parents, the hearing loss can have a significant impact on speech, and indirectly affect academic achievement or other aspects of life, such as social functioning [20,50–53]. However, the spread of hearing screening programs has contributed to earlier diagnoses and interventions for hearing loss [54,55]. Furthermore, the invention of cochlear implants (CIs), an electrical hearing prosthesis that provides access to environmental sounds and spoken language, has provided the opportunity for habilitation of children with profound hearing loss [56]. CIs provide access to sensory input, improving the perception of sound and acquisition of spoken language, leading to a larger proportion of children approaching the spoken language levels of their peers with typical hearing [51,52,56]. Improvements in communication abilities after implantation are also reported to positively affect the child's relationship with family members and peers [57].

Nevertheless, it should be borne in mind that it takes substantial time for young children or infants to adjust to a CI as an aid to hearing – and so CIs may not be sufficient to overcome slower development in several aspects of spoken language (e.g. [38,58]). Indeed, it has been reported that deaf children with CIs experience delays in various domains of spoken language development, including grammar and pragmatic skills [59] and lexical comprehension [60]. The variability in spoken language outcomes in children following cochlear implantation remains quite high. Previous studies have shown that spoken language performance in children following cochlear implantation is influenced by the age at implantation and access to conversations [58]. The main idea behind early cochlear implantation is that children who are implanted earlier have better spoken language outcomes because they will have experienced a shorter period of auditory deprivation and had more opportunities to engage in vocal interaction with their caregivers than children who are implanted later [61].

Geers and Sedey [52] assessed children's spoken language skills first during elementary school and then at high school, finding that deaf children who had earlier implantation – and who had thus experienced a shorter period of auditory deprivation – had better spoken language skills later in life. However, a number of those children still encountered difficulties in connected discourse and abstract reasoning tasks.

The importance of early implantation for spoken language acquisition can be explained by the hypothesis of sensitive or critical periods in language development, an idea supposing that there is a biologically determined period of life when language can be acquired more easily and after which language becomes increasingly difficult to acquire. Although it is difficult to precisely identify the optimal time for language acquisition, there is a general consensus that earlier implantation is better than later implantation, as it minimizes the gap between the child's chronological age and their linguistic age [58,61]. However, despite conflicting results of studies comparing language development in deaf children who received implants before the age of 1 to those who received CIs between 1 and 2 years of age (which suggest a small or nonexistent advantage in the case of younger children), there are consistent results showing that deaf children implanted later than 2 years of age lag behind their peers with typical hearing in language development. We can therefore conclude that the period before 2 years of age is critical for language development [62].

Over the years, the consensus regarding the critical period for cochlear implantation has changed – from up to 5 years previously to 12 months of age now [58]. However, in a recent review and meta-analysis, Duchesne and Marschark [58] emphasized the variability in studies examining the relationships between age at cochlear implantation and vocabulary and grammar outcomes. While many of the reviewed studies found significant relationships between early implantation and better language outcomes, the authors conclude that other factors, including the family environment, should also be considered when trying to explain the observed variability.

The relationship between CIs and language might also depend on the language domain of interest, as some language skills might be more difficult than others. For example, Geers et al. [63] compared the performance of 5- and 6-year-old deaf children with CIs to their peers with typical hearing on various language measures. They found that syntactic tasks were more difficult than vocabulary tasks. Conversely, Boons et al. [64] did not find any strong or weak aspects of language development in school-aged deaf children with CIs compared to their peers with typical hearing. However, after a qualitative analysis of systematic errors in language tasks, they found that deaf children with CIs made more severe errors in syntactic tasks than the control group.

There could also be differences in terms of the quality of conversations displayed by parents of deaf children with CIs and parents of children with typical hearing, especially when it comes to the discussion of topics that can't be seen or pointed to [18]. Conversations are important for language acquisition, but they also affect the development of social perceptions, since parents tend to guide their children's perception of the environment and improve their reasoning about the cognitive and emotional states of others. For instance, a study by Morgan et al. [18] found that hearing mothers of deaf infants (using CIs or hearing aids) engaged in less effective exchanges with their children and made fewer references to mental states than the mothers of hearing infants. The authors hypothesized that hearing parents of deaf children might try to adjust the quality of their conversations to the perceived level of understanding of their child.

Current research on deaf children who use CIs has extended beyond functional communication, as improved language acquisition can also affect social development and ToM abilities [65]. In order to fully understand the relationship between cochlear implantation and ToM, different types of predictors should be included in the studies: age at implantation, level of specific language abilities, and family factors, including the quality of parental conversational input and the parents' perceptions of their child's social functioning.

ToM development in deaf children with CIs

The development of ToM in deaf children of hearing parents and, more specifically, in DoH who use CIs, has gained increased attention. The majority of studies conducted in the field aim to answer the question of whether deaf children with CIs who are raised by hearing parents struggle with ToM compared to their peers with typical hearing. The results are inconsistent and the underlying mechanisms will now be highlighted.

The first study on ToM development in deaf children of hearing parents was performed by Peterson and Siegal in 1995 [37]. They found that the majority of school-aged signing deaf children (who were raised by hearing parents) failed the standard false belief test. Peterson [14] examined deaf children with cochlear implants separately, making a novel contribution to the study of deafness and ToM. She found that deaf children with CIs performed worse on the FBT in comparison to preschoolers with typical hearing and suggested that deaf children with CIs could have delayed ToM reasoning. Even though CIs improve development in terms of communication and socialization [66], other researchers have also found that deaf children with CIs exhibit delayed ToM development, in particular false belief understanding, in comparison to their peers with typical hearing [67–71]. For example, Yu et al. [71] reported that only 3% of deaf preschoolers with CIs (aged 3–6) succeeded in the false belief task, a dramatically low success rate since the majority of hearing preschoolers pass the false belief task around the age of 5 [6].

Similar results were shown by Ketelaar et al. [67] who demonstrated that 2–4 year-old deaf children with CIs lagged in false belief understanding behind their peers with typical hearing. Nevertheless, the ability to ascribe early ToM concepts (understanding desires and intentions) to others was intact in this group of deaf children with CIs. A different pattern was demonstrated by Meristo et al. [72], who showed that deaf children with CIs did not differ from their peers with typical hearing on the verbal false belief task. However, they failed the implicit false belief task, meaning they were unable to spontaneously anticipate another person's belief.

In contrast, Remmel and Peters [73] did not find differences in ToM ability between deaf children with CIs and their peers with typical hearing. Ziv et al. [74] also reported no difference between a group of 20 deaf children with CIs who used spoken language as their main mode of communication (mean age = 6.6 years; mean age at implantation = 2.5 years) and 23 peers with typical hearing in either affective perspective-taking or in change-of-location false belief understanding. Although the study revealed a relatively high average success rate of deaf children with CIs on tasks measuring different domains of social development, the authors highlighted greater heterogeneity in ToM performance among deaf children with CIs than in children with typical hearing. This means that high rates of variability in ToM skills are widespread in deaf children with CIs even after many years of CI experience.

Delay in false belief understanding might also affect the development of advanced ToM concepts, as suggested by the recent study of Figueroa et al. [75]. They showed that deaf adolescents aged 12–16 years with CIs had a lower understanding of second- or higher-order beliefs and of understanding multiple perspectives requiring mentalizing. Marschark et al. [69] have even reported differences in advanced ToM in deaf adults with CIs.

Results of studies of ToM in deaf children with CIs are mixed, but the majority of studies report delayed performance on ToM tasks in comparison to peers with typical hearing. Some show that there are partial deficits depending on the task. Few indicate no differences between these groups. However, there are various factors that might contribute to the performance on ToM tasks in deaf children with CIs, such as language skills and access to conversations about mental states (see [49] for a review of ToM and language development in DoH), age at implantation, family correlates, and executive functions. These factors are now described.

Spoken language abilities have been found to be associated with false belief understanding in deaf children with CIs (e.g. [14,43,73]). There are few studies aiming to determine which domains of language account for much of the success in ToM tasks in deaf children with CIs (for a review of the relationship between false belief understanding and language skills, including children who are deaf, see [40]). For example, Schick et al. [20] demonstrated that both vocabulary and comprehension of syntactic complements were significant independent predictors of success on ToM tasks. Remmel and Peters [73] showed a higher correlation between ToM performance and general syntactic proficiency than between ToM score and measures of complement syntax in deaf children with CIs.

In general, research with deaf children with CIs supports the idea that language experience affects ToM development and that delayed language acquisition is the key predictor of hampered ToM development of deaf children with CIs. There are a number of studies demonstrating this relationship (e.g. [14,43,71,73]). For example, Yu et al. [71] showed in a longitudinal study that deaf children with CIs who had more advanced language ability had better ToM growth. However, in the previously mentioned study by Ketelaar et al. [67], the authors demonstrated that despite having the same level of spoken language skills, deaf children with CIs still lagged behind their peers with typical hearing in false belief understanding. Furthermore, there might be a possible confounding effect of linguistic demands on the standard explicit false belief test, as it requires that the child follows the course of a story and answers test questions. The issue of linguistic demands on the standard FBT has also been raised by researchers working on nonverbal (implicit) ToM in infants [8], who proposed that failure on these tasks might be due to processing difficulties. However, de Villiers and de Villiers [43] and Pluta et al. (in press) [76] found that deaf children with CIs had difficulties reasoning about false beliefs even when the linguistic demands of the task were minimized.

These results might also be attributed to limited conversational input early in life, as hearing parents could find it difficult to interact with their deaf child until he or she acquires a sufficient level of spoken language development after cochlear implantation and effective hearing habilitation. Consistently, ToM impairment has not been reported in deaf children whose parents are native signers and could freely communicate with their child, supporting the hypothesis that limited access to language could impact ToM development in profoundly deaf children who are raised in a hearing culture [49]. Some have also emphasized that better language skills might provide enhanced access to conversations about mental states (e.g. [73]). Peterson [38] proposed certain potential mechanisms underlying delayed ToM in DoH children with CIs. She explained that deaf children are deprived of exposure to mental state conversations until they manage to master enough spoken language to be able to follow their parents' spoken conversations. Subsequently, they can gradually start to acquire receptive and expressive language (words referring to mental states such as "know" or "believe") as well as the syntax (sentential complements and relative clauses) necessary for conversations about mental states [38]. Moreover, a study by Moeller and Schick [77] found that the frequency of maternal references to mental states was related to false belief understanding in deaf children. This resonates with studies of children with typical hearing, which also emphasized the importance of a cognitively and socially stimulating environment for ToM development [13,46].

Sundqvist et al. [68] found that children who were implanted earlier (before 27 months) performed better on emotional ToM tasks than children who were implanted later (after 27 months). However, this finding has not been confirmed in other studies and some have advised that age at implantation should not be treated as the only factor explaining the delay in ToM experienced by deaf children with CIs [65]. Moreover, as described earlier, age at implantation is related to language skills.

A recent review by Marschark et al. [65] suggested that in order to fully understand the relationship between ToM and CIs, other factors, such as family correlates should also be taken into account. Previous studies focusing on children with typical hearing found that the family's socioeconomic status and number of siblings contributed to the child's ToM and false belief understanding [48]. Other factors could also be attachment security or the parent's propensity to be attuned to their child's mental states – thoughts, desires, emotions, and intentions (also referred to as mind-mindedness) [48,78].

Another factor that could be important in terms of deaf children with CIs is the quality and quantity of joint attention between the child and caregiver [79]. For example, MacGowan et al. [80] found that joint attention scores between hearing mothers and deaf children aged from 17 to 41 months (including deaf children with CIs) were positively related to the child's social competence as reported by the mother. Interestingly, this relationship was not observed in the hearing dyads. Furthermore, including parental assessment of the child's ToM ability in everyday settings could also help provide further perspectives on the characteristics of deaf children with CIs [26].

Finally, apart from the linguistic, conversational, and family factors, executive functions have also been indicated as a possible factor contributing to the performance on ToM tasks (in particular, the FBT) [81]. A positive relationship between executive functions and ToM development has been confirmed in children with typical hearing as well as in deaf children with CIs [70].

It is also worth noting that the equivocal results of studies on ToM development in deaf children with CIs might partly result from differences in the recruitment procedures used, since groups are heterogeneous in terms of age at implantation and preferred mode of communication (spoken or signed language) (e.g. [67,73]). Moreover, Ziv et al. [74] highlighted the greater heterogeneity in ToM performance among deaf children with CIs than in controls with typical hearing. This is important, as it suggests that assessment of ToM should be routinely done in deaf children with CIs in order to identify individuals who need tailored interventions with additional ToM training.

Interventions to promote ToM in deaf children

Several studies have provided evidence that mentalizing abilities might be enhanced via specific intervention programs designed to focus a child's attention on the mental states of others. This could be achieved by training caregivers to talk more elaborately about past events with their children, or by teaching children the language used to talk about the mental states of others [82]. For example, the training designed by Wellman and Peterson [83], using cartoons with thought bubbles, helped school-aged deaf children understand that different people might have different representational mental states. After the training, children scored higher on false belief scales than did the control groups without training. It was also demonstrated that ToM can be scaffolded with explicit instructions [49] or through using fiction books in order to engage the child in exploring the topic of thoughts and feelings [84]. Additionally, interactions between deaf parents and deaf children could serve as a model for interactions in the population of DoH children. By mimicking deaf parents of deaf children, hearing parents might learn how they can adjust their behavior to effectively adapt to their child's needs in terms of visual input (in particular, prior to implantation when their children have no auditory input). This might facilitate the development of joint attention (prior to CI implantation), which is an important prerequisite of ToM ability [79]. Additionally, closer collaboration between practitioners working with deaf children and researchers is still needed. Beazley and Chilton [85] conducted qualitative interviews with five educators of deaf children (including deaf children with CIs) in terms of ToM development. In some parts of the interviews, practitioners described techniques they were using that could support ToM ability in children, such as book sharing, role play, or "speech" and "thought" bubbles. However, although most participants were familiar with the concept of ToM, they were unsure about its definition or implications for deaf children in their everyday practice. Nevertheless, practitioners expressed their expectations towards future ToM research in terms of supporting their work and strengthening the collaboration.

Summary

Understanding the development of ToM during childhood has practical relevance because success on tests of ToM correlates with many important aspects of social life, including mental health in general. To date, there have only been a few studies that focus on ToM development in deaf children with CIs, and the results of these studies are mixed due to confounding factors (e.g., participants varied substantially in terms of age at implantation and their preferred mode of communication, parental hearing status, or linguistic complexity of the ToM tasks [67,73]).

The majority of these studies report that false belief understanding is delayed in deaf children with CIs. Considering the fact that the protocol for when a child should receive a CI has changed recently (cochlear implantation is now performed in children under 12 months of age [86]), there is a pressing need to conduct studies on children who were implanted early in order to determine how severe ToM delays are (if any) in this unique population.

Studies of deaf children of hearing parents further confirm a positive relationship between language, conversations about mental states, and ToM ability, which has been previously indicated in studies of children with typical levels of hearing [12,13]. Thus, it is not deafness per se, but rather delayed spoken language development and restricted early access to abstract mind-related discourse, that are the key factors explaining ToM delays in DoH [38]. There are also different variables that might explain and/or mediate this relationship, such as the age at which the child started to receive auditory input, level of language ability and access to conversations about mental states, socioeconomic status, and parental education [18,26,65,82].

Furthermore, to the best of the authors' knowledge, no study of deaf children with CIs has yet adopted a social constructivist perspective and investigated the relationship between the quality of social interactions (including the propensity to use mental state talk) and ToM development in DoH children who received their implant in infancy.

Future studies could also further inform rehabilitation programs and provide practical guidelines for therapists and parents of deaf children with CIs. Moreover, based on existing studies on ToM in DoH, clinicians should consider including assessments of mentalizing ability in interventions offered to this population.

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ENDOSCOPY OF THE CEREBELLO-PONTINE ANGLE: AN OVERVIEW

Contributions:
A Study design/planning
B Data collection/entry
C Data analysis/statistics
D Data interpretation
E Preparation of manuscript
F Literature analysis/search
G Funds collection

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Abstract

Introduction: The cerebello-pontine angle (CPA) is an important region in the skull base which can harbour a variety of pathologies. Because it is surrounded by numerous vital structures, surgical access is a challenge, more so when distorted by disease. Microscopes have successfully guided CPA surgery over the past three decades. CPA endoscopy has evolved today as an alternative way to explore this intriguing region with minimal morbidity to collateral structures.

Discussion: CPA endoscopy has been introduced on an experimental basis into a range of CPA surgeries, including assistance in lower cranial nerve tumor removal, microvascular decompression, vestibular neurectomy, and assistance in auditory brainstem implantation. CPA endoscopy is currently used as a surgical adjunct to the operating microscope, and it has the potential to become standard in many CPA surgeries.

Conclusions: CPA endoscopy has evolved from a diagnostic to a therapeutic tool because it allows near and clear visualization of the CPA with in-depth view of the various nerve roots arising from the brainstem and their exit foramina. This review evaluates the current status and future directions of endoscopic technology and its role in skull base surgical practice.

Key words: cerebello-pontine angle, endoscopy, skull base, cranial nerves

ENDOSKOPIA KĄTA MOSTOWO-MÓŻDŻKOWEGO – OMÓWIENIE

Streszczenie

Wprowadzenie: Kąt mostowo-móżdżkowy (CPA) jest ważnym rejonem podstawy czaszki, w którym mogą skrywać się różne patologie. Ze względu na otaczające go liczne struktury dostęp chirurgiczny do CPA jest trudny, szczególnie gdy rejon ten jest zniekształcony z powodu choroby. Przez ostatnie trzy dekady w chirurgii CPA z powodzeniem stosowano wizualizację mikroskopową. Od niedawna rozwija się endoskopia CPA jako alternatywna metoda badania tego intrygującego rejonu powodująca tylko minimalne uszkodzenia sąsiadujących struktur.

Dyskusja: Endoskopia CPA została wprowadzona eksperymentalnie w szeregu operacji kąta mostowo-móżdżkowego, w zabiegach m.in. usunięcia guza dolnej gałęzi nerwu czaszkowego, dekompresji mikronaczyniowej, przecięcia nerwu przedsionkowego, czy pomocniczo podczas wszczepiania implantu słuchowego pnia mózgu. Endoskopia CPA jest obecnie wykorzystywana jako wspomożenie mikroskopu chirurgicznego podczas operacji. Potencjalnie może być stosowana jako standard w wielu operacjach CPA.

Wnioski: Endoskopia CPA jest wykorzystywana i przy diagnostyce i przy terapii, ponieważ narzędzie to umożliwia wyraźnie uwidocznienie CPA z pełnym obrazem korzeni nerwowych wychodzących z pnia mózgu oraz ich otworów wyjściowych. Niniejszy przegląd zawiera ocenę obecnego stanu i przyszłych kierunków rozwoju technologii endoskopowych i ich znaczenia w chirurgii podstawy czaszki.

Słowa kluczowe: kąt mostowo-móżdżkowy • endoskopia • podstawa czaszki • nerwy czaszkowe

Introduction

The skull base has, for a long time, been considered to be a "no man's land" and surgically unapproachable because of the anatomical complexity and the vital importance of the structures within its boundaries. The advent of endoscopes has paved the way for minimal access approaches to the cranial contents via the ENT regions. Thereby ENT and neurosurgeons began collaborating to comprehensively navigate their way to skull-base lesions with minimal morbidity, while providing optimal outcomes. The invention and first use of the endoscope was by a German urologist Maximilian Carl-Fridrich Nitze in 1879, along with a Viennese instrument maker Joseph Leiter [1]. This initial endoscope was a cystoscope, used for urological purposes. Later in 1917, the first CPA endoscopy with this endoscope was performed by Doyen to conduct a trigeminal neurectomy. This marked the beginning of the use of endoscopy in the posterolateral skull base [2]. These initial endoscopes were rather crude in design and had limitations in lighting and magnification, and so they did not gain popularity for many decades. The operating microscope invented in the 1960s almost made endoscopes obsolete. However, in the 1990s improved technology led to the reintroduction of endoscopes into skull base surgery [3]. This overview highlights the CPA endoscopic approach, which has become a very useful minimally invasive tool to address a spectrum of CPA lesions.

Discussion

MIRA approach to CPA endoscopy

The introduction of an endoscope into the CPA through a minimal access port, as in the minimally invasive retrosigmoid approach (MIRA), was a logical progression for the surgeon to explore boundaries in the skull base with minimal morbidity. In otology, endoscopes have become useful in exploring the middle ear, Eustachian tube, and more recently the cochlea. These same endoscopes could be used in the CPA. Commonly used rigid endoscopes are either 2.7 mm or 4 mm in diameter, with viewing angles of zero, 30, and 70°. Since the CPA is a region of arachnoid cisterns and provides a pathway to cranial nerves and intracranial major vessels, knowledge of the microanatomy of vessels and nerves in this region is fundamental for diagnosis and functional neurosurgery. CPA endoscopy has helped classify the neuroanatomy of the CPA cistern into four levels:

- *Level I:* Trigeminal area – V and VI nerves, and superior cerebellar artery
- *Level II:* Acousticofacial area – VII and VIII nerves, anterior inferior cerebellar artery (AICA)
- *Level III:* Lower cranial nerves area – IX, X, and XI nerves, posterior inferior cerebellar artery (PICA)
- *Level IV:* Foramen magnum level – spinal root of accessory and XII nerves, vertebral basilar and PICA arteries.

CPA endoscopy offers a panoramic view of an area difficult to visualize (Figures 1, 2), which otherwise needs a wide craniotomy with cerebellar retraction. This approach enables the surgeon to visualize and carry out procedures on a wide variety of structures, including surgery for lesions of cranial nerves V to XI, arteriovenous malformation clippings, and microvascular decompression for vascular loops (Figure 3). Today, a variety of indications exist for CPA endoscopy, such as hemifacial spasm, trigeminal neuralgia, vestibular neurectomy (Figure 4), vascular tinnitus, glossopharyngeal neuralgia, endoscopy-assisted vestibular schwannoma excision (Figure 5), and for cholesteatoma/epidermoid cyst excision. Many of its indications are still evolving, but one exciting prospect in the near future is the option of performing endoscopic auditory brainstem implantation (ABI) – another novel minimal access procedure (Figures 6, 7). Setty et al. reported fully endoscopic excision of 12 cases of vestibular schwannoma without any complications such as CSF leak, cranial nerve palsy, meningitis, or wound infection. The mean per operative blood loss was only 56 mL and the average hospital stay for their patients was 3.6 days [3]. Jarrahy et al. described for the first time a fully endoscopic technique for microvascular decompression [4]. Krass et al. [5] and De Divitiis et al. [6] have reported successful endoscopic removal of epidermoid tumours. Magnan et al. reported endoscopic vascular decompression of the eighth cranial nerve in 25 patients suffering disabling positional vertigo, and endoscopic vestibular neurectomy in 45 patients with Meniere's disease [7].

Polyaxial pneumatic holding arms can hold the endoscope rigidly in place, allowing the surgeon to use both hands for instrumentation. Such holding arms can easily be moved in all planes [3]. While the retrosigmoid approach gives a more tangential view (70–90° to the long axis of the 8th nerve), another approach to CPA endoscopy, the retrolabyrinthine approach introduced by Hitselberger and Pulec, gives an angle of view of 30–70° posterior to the vestibulocochlear nerve [8].

Emerging trends in CPA endoscopy

CPA endoscopy is currently used by neurosurgeons and neurotologists as a surgical adjunct to the operating microscope. It improves visualization of bony, neural, and vascular

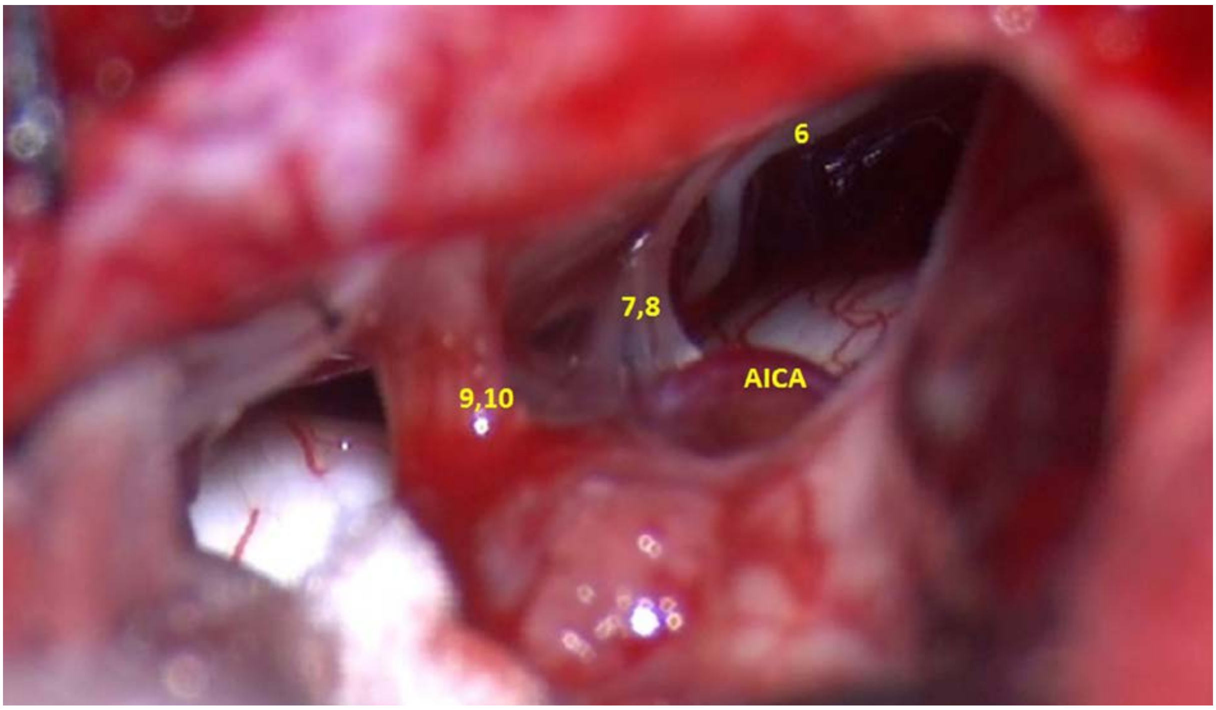

Figure 1. Left CPA endoscopy showing panoramic view of the 6th to 10th cranial nerves. AICA = anterior inferior cerebellar artery

structures, while minimizing retraction of these structures. Endoscopic exploration after tumor removal can reveal the presence of residual tumor or air cells that may otherwise result in recurrent tumor or cerebrospinal fluid (CSF) oto-rhinorrhoea post-operatively. Endoscopes fitted with high definition cameras provide high magnification and illumination of the operative field, but blood soiling the tip of the endoscope can make visualization difficult. There is also a risk of potential thermal injury, and maintaining 3D orientation in the posterior fossa requires experience. Such technical issues are being refined and full-fledged CPA endoscopic surgery may emerge in the near future. Thereby, exciting prospects like endoscopic microvascular decompression, endoscopic aneurysmal clipping, endoscopic tumor removal, and minimally invasive endoscopic auditory brainstem implantation may all become standard skull base surgery.

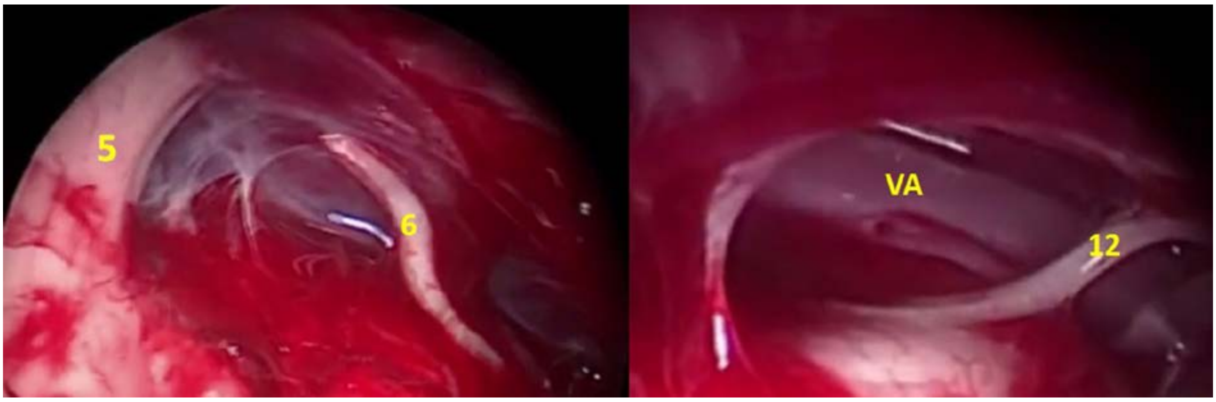

Figure 2. Right CPA endoscopy showing superior (left) and inferior (right) views. 5 = 5th cranial nerve; 6 = 6th cranial nerve; VA = vertebral artery, 12 = 12th cranial nerve

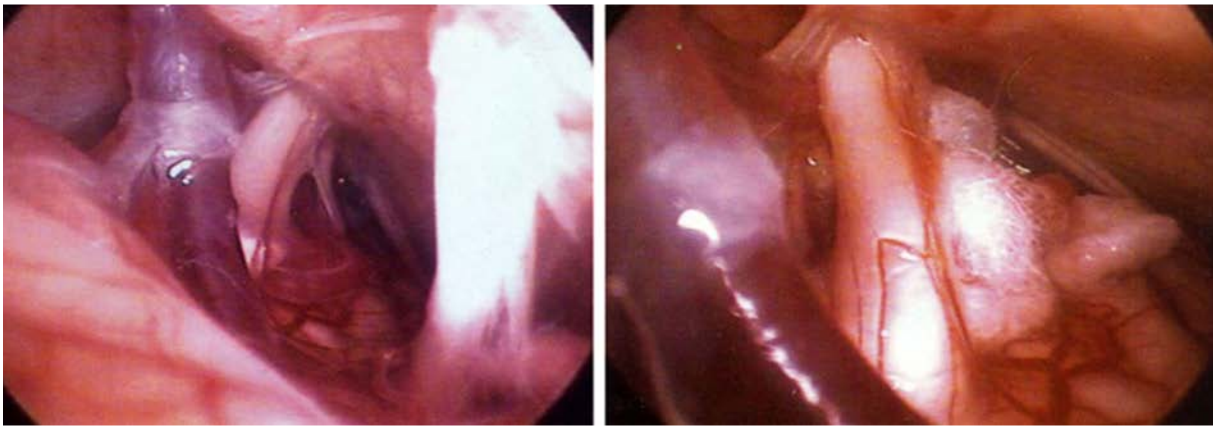

Figure 3. Left: Endoscopic view of superior cerebellar artery (SCA) pressing the Vth nerve and causing trigeminal neuralgia. Right: Micro-vascular decompression using a Teflon pad

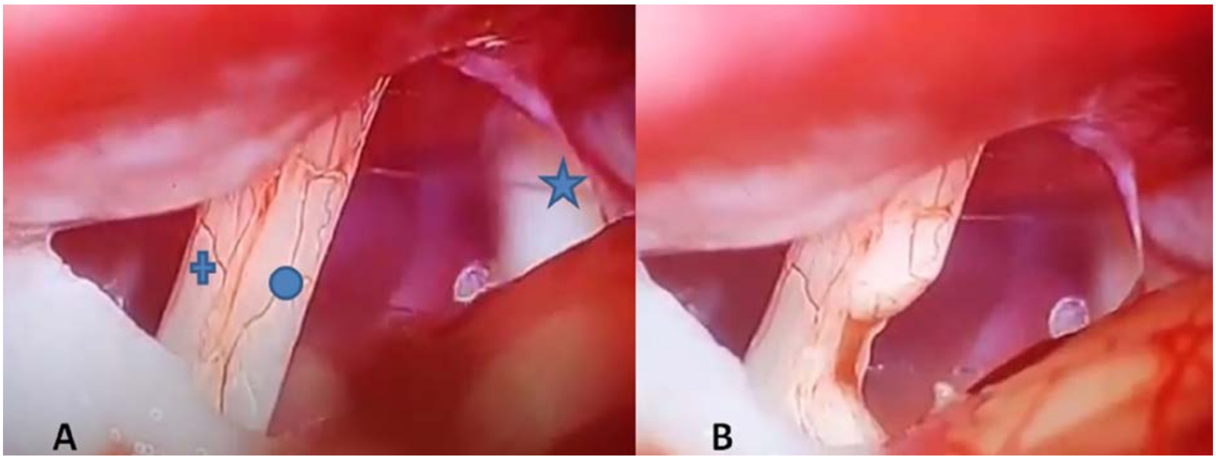

Figure 4. Left CPA endoscopy showing superior vestibular nerve section (before, left; after, right). Blue cross = inferior vestibular nerve; blue circle = superior vestibular nerve; blue star = trigeminal nerve superiorly

Figure 5. Endoscopic picture of the mid-zone of the right CPA showing a moderate size cisternal vestibular schwannoma with its origin in the internal auditory canal (IAC)

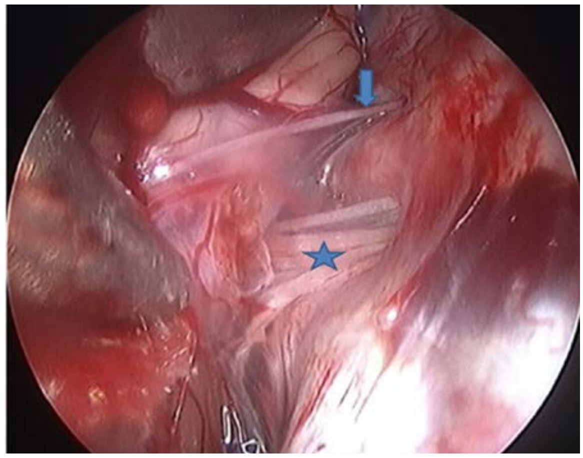

Figure 6. CPA endoscopy (right side) in a child for ABI showing facial nerve alone passing through the IAC (blue arrow) with an absent 8th cranial nerve. The 9th and 10th cranial nerves are seen passing through the jugular foramen (blue star)

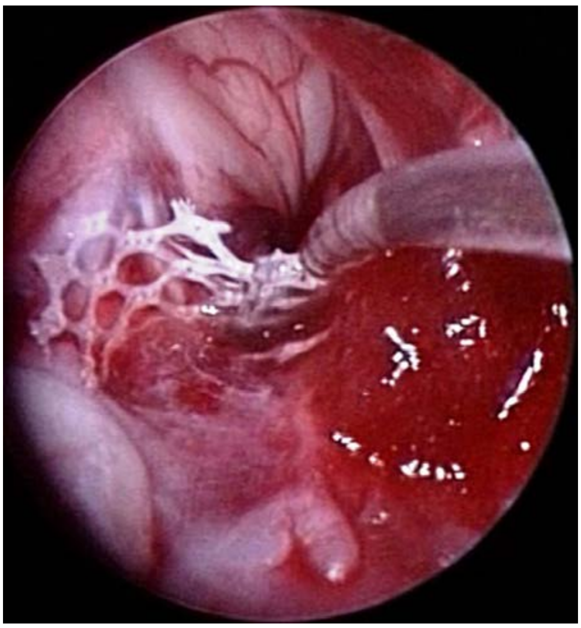

Figure 7. CPA endoscopy assisted ABI electrode placement in the lateral recess of the fourth ventricle

Conclusion

Advances in technology like neuro-navigation with image guidance have been the catalyst for making minimally invasive surgery popular worldwide. The less invasive endoscopic skull base approach has quicker recovery, less morbidity, and shorter hospital stay than traditional external approaches. CPA endoscopy has evolved from a diagnostic to a therapeutic tool, since the endoscope allows near and clear visualization of the CPA with in-depth view of the various nerve roots arising from the brainstem and their exit foramina. This panoramic view helps with identification of critical anatomical landmarks with minimal trauma, excellent illumination and magnification, and has decreased complications. The drawbacks include lack of binocular vision, poor depth perception in the operating field, and inability to use both hands for surgery, areas where the operating microscope scores over the endoscope. There is therefore a learning curve for surgeons to optimally perform endoscopic skull base surgery, which calls for years of practice. Endoscopic surgeons must be familiar with handling endoscopes as well as CPA anatomy before attempting diagnostic or therapeutic procedures in this intricately positioned vital region of the cranial base.

Conflicts of interest: None.

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Hypothesis paper

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THE CURIOUS 'TYPE C' TYMPANOGRAM: CONTRACTION OF THE TENSOR TYMPANI MASQUERADES AS NEGATIVE MIDDLE EAR PRESSURE

Contributions:
A Study design/planning
B Data collection/entry
C Data analysis/statistics
D Data interpretation
E Preparation of manuscript
F Literature analysis/search
G Funds collection

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Abstract

Negative middle ear pressure presents something of a paradox. The 'Type C' tympanogram, in which the peak of the tympanogram occurs below zero pressure, seems to indicate that the air pressure in the middle ear is actually below atmospheric pressure – that there is a degree of suction – and yet the peak can remain persistently in place even if the subject swallows and opens their Eustachian tube. Negative middle ear pressure can even be measured when a subject has a permanently open (patulous) Eustachian tube, a situation that seems physically impossible. This paper reviews the paradox and concludes that in many cases of "negative middle ear pressure" the actual pressure inside the middle ear is in fact zero, but the tympanometric offset comes about because of the unappreciated action of the tensor tympani: when this muscle contracts, it pulls the eardrum inwards, and this inwards force is registered as negative middle ear pressure during tympanometry. That is, the force exerted by the muscle needs to be countered by a negative pressure in the ear canal in order to bring the eardrum back to its equilibrium position. This interpretation is reinforced by a number of findings in the literature, which are reviewed. A proposal for how tensor tympani effects might be separated from actual middle ear pressure offsets is made.

Key words: tympanometry • tensor tympani • negative pressure

CIEKAWY TYMPANOGRAM TYPU C: SKURCZ MIĘŚNIA NAPINACZA BŁONY BĘBENKOWEJ POZORUJE UJEMNE CIŚNIENIE W UCHU ŚRODKOWYM

Streszczenie

Zjawisko ujemnego ciśnienia w uchu środkowym wiąże się z pewnym paradoksem. Tympanogram typu C, którego szczyt wypada poniżej zerowej wartości ciśnienia, wydaje się wskazywać, że ciśnienie powietrza w uchu środkowym jest niższe od ciśnienia atmosferycznego – że występuje pewien stopień ssania – jednak szczyt często pozostaje niezmiennie w tym samym miejscu, nawet gdy osoba badana przełknie i odblokuje trąbkę słuchową. Ujemne ciśnienie w uchu środkowym można nawet zmierzyć, gdy osoba badana ma stale otwartą (rozwartą) trąbkę słuchową, co wydaje się fizycznie niemożliwe. W niniejszej pracy zbadano ten paradoks, dochodząc do wniosku, że w wielu przypadkach „ujemnego ciśnienia w uchu środkowym", ciśnienie to jest tak naprawdę zerowe, a przesunięcie tympanogramu jest efektem niedocenionego działania mięśnia napinacza błony bębenkowej: skurcz tego mięśnia wciąga błonę bębenkową do środka. Ta skierowana do wewnątrz siła jest w badaniu tympanometrycznym rejestrowana jako ujemne ciśnienie w uchu środkowym. Aby przywrócić błonę bębenkową do pozycji równowagi, działanie mięśnia musi zostać zrównoważone ujemnym ciśnieniem w kanale słuchowym. Ta interpretacja jest zgodna z wieloma opublikowanymi doniesieniami, których przegląd jest zamieszczony. Zaproponowano rozwiązanie, w jaki sposób można rozdzielić efekty działania mięśnia napinacza błony bębenkowej od właściwego efektu wyrównania ciśnienia w uchu środkowym.

Słowa kluczowe: tympanometria • mięsień napinacz błony bębenkowej • ujemne ciśnienie

Introduction

Tympanometry is a useful tool that allows a number of middle ear problems to be diagnosed. The procedure involves placing an acoustic probe in the ear canal and measuring the stiffness (or its inverse, compliance) of the middle ear system as the air pressure in the canal is continuously swept from low pressure (suction) to high pressure (overpressure). Figure 1 shows the main features of the arrangement. The minimum stiffness of the eardrum – its maximum compliance – will occur when the pressures on either side of the drum are equal, and this is the situation found in most subjects where the tympanogram registers a compliance peak at zero ear canal pressure. According to the classification system of Jerger, this is the 'Type A' tympanogram [1].

The focus of this paper is on the 'Type C' tympanogram in which the peak of the tympanometric curve is displaced appreciably towards negative values, taken by Jerger to be more negative than –100 daPa but which is not infrequently recorded at pressures of –200 daPa or more (1 daPa is a pressure of about 1 mm of water). In one report, a middle ear pressure of –380 mm of water is recorded [1a]. Figure 2 illustrates typical Type A and Type C tympanograms. Paradoxically, this "negative middle ear pressure" can persist even when the subject swallows and opens their Eustachian tube, an action which connects the middle ear cavity

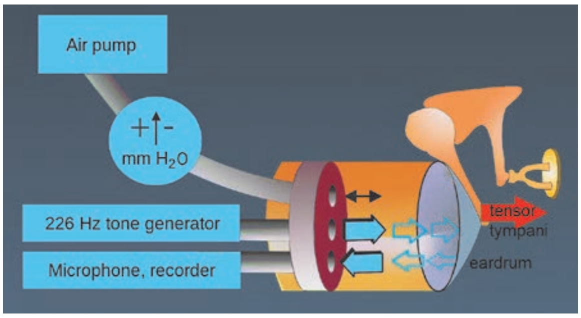

Figure 1. The elements of tympanometry. The impedance of the eardrum is sensed by comparing the original signal from a tone generator with that picked up with a microphone. During a measurement the air pressure in the ear canal is swept from –400 to +200 daPa with an air pump. Note how the tensor tympani is attached to the malleus and eardrum, and its contraction exerts an inwards force that needs to be countered with negative pressure in the ear canal to return the ear drum and ossicular chain back to their neutral position. That is, muscle contraction gives the same outcome as negative middle ear pressure. Figure adapted from Wikipedia Commons, CC BY-SA 3.0

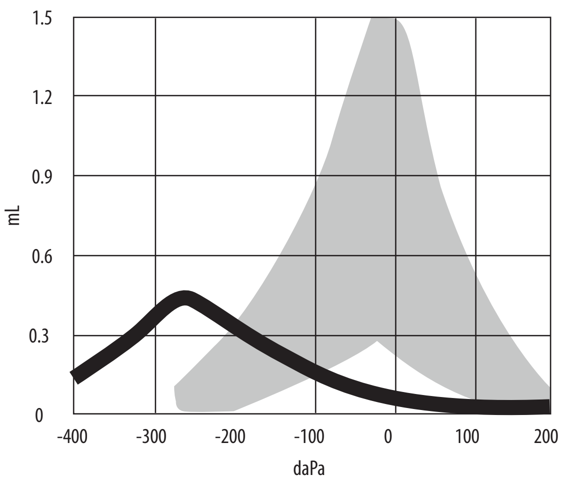

Figure 2. Illustration of Type A (light grey) and Type C (black) tympanograms. The peak of the curves reflects where the eardrum has its maximum compliance (measured as mL) and is taken to be where the pressure in the middle ear matches the pressure in the ear canal. That is, most people will show a peak near zero daPa (Type A), although there is a considerable range. If the peak occurs at negative values, it is called a Type C tympanogram (here registering a pressure of –275 daPa). Figure from audiologyonline.com

to the outside air (their ears 'pop'). The standard interpretation of a negative middle ear pressure is that the subject has a blocked Eustachian tube, usually due to otitis media (glue ear), allowing absorption of gas by the mucous layer lining the middle ear cavity and producing a negative pressure. But strangely, negative pressure can also be measured in subjects with a patulous (open) Eustachian tube. Sadé (2001) notes the "astounding and seemingly paradoxical fact of occasional association of atelectasis [retracted tympanic membrane] with a patent patulous eustachian tube" (p. 136 of [5]).

As has been nicely said, paradox is truth standing on its head in order to attract attention. Recognising the paradox, this paper seeks to resolve it. The conclusion is that true suction in the middle ear is not common (although possible); instead, a negative middle ear pressure reading as recorded by tympanometry is more often a case in which the tensor tympani muscle is under steady contraction, pulling the eardrum inwards. Otoscopically, the eardrum looks very similar to when it's really under suction but in most cases this is illusory. Tympanometry is really indicating that the force exerted by the muscle, tending to pull the eardrum inwards, needs to be countered by a negative pressure in the ear canal in order to restore the eardrum to its neutral position.

This hypothesis explains a number of anomalies in the literature and gives an insight into how the tensor tympani – the larger and stronger of our two middle ear muscles – regulates the operation of the ossicular chain. This paper supports the intralabyrinthine pressure theory of middle ear muscle action – the mechanism whereby the tensor tympani controls the pressure of fluid inside the cochlea, and so, acting like a hydraulic brake, controls hearing sensitivity.

Context for the evidence

There should really be no surprise to learn that contraction of the tensor tympani muscle draws the eardrum – the tympanum – inwards and tenses it, for that is just what its name (in Latin) means. When an audiologist inspects the eardrum with an otoscope, and they cause the tensor tympani to contract by applying a puff of air to the eye or brushing the cheek with a finger, that is just what is seen. A retracted drum will sometimes show 'retraction pockets', where parts of the drum are more yielding than others. You can make the tensor tympani contract yourself – just by yawning or tightly closing the eyes. The low fluttering rumble one hears is the sound of the muscle at work.

Perhaps this temporary reflex action of the tensor tympani has distracted people from appreciating that the muscle is designed for long-term constant contraction. It is a muscle made up of very fine fibres which are designed for sustained, isometric force generation (see Bell [2] for a detailed description of some of this unappreciated muscle's unique properties). According to this author, the main role of the tensor tympani is as a fast and precise acoustic gain controller: by constantly adjusting the force it applies to the ossicular chain, it changes the hydraulic pressure of fluids inside the cochlea and thereby changes the gain of the cochlear amplifier [2-4]. Because the fluids of the cochlea are virtually incompressible, and the round window so small, the range of movement of the muscle is minuscule, perhaps 0.1 mm [4], and so the constant activity of this nearly isometric muscle goes largely unnoticed.

Tympanometry is perhaps the best way of measuring tensor tympani activity directly. The pressure in the ear canal balances the pull of the muscle, and so the offset of the compliance peak (in daPa or mm of water) is a measure of the force the muscle is exerting. Referring to Figure 3, it can be calculated that a negative pressure of 100 daPa (= 1 kPa) acting over the area of the eardrum (about 50 mm^2) generates a force of 50 mN, which counters the contractile force of the muscle. In other words, when tympanometry records a middle ear pressure of –100 daPa, the tensor tympani is exerting a counteracting force of about 5 grams weight, a figure that matches the likely power of a muscle this size (a body of 25 mm).

It is worth noting at this point that if that same force is transferred to the ossicular chain and finally to the stapes, which has an area of 3 mm^2, then the force will create a hydraulic pressure in the sealed cochlea of about 20 kPa (2000 mm water). This is important in understanding how the middle ear muscles adjust hearing sensitivity, and will be brought up later in connection with Eustachian tube dysfunction, which also relates to middle ear pressure anomalies.

Jerger notes that negative middle ear pressures are a regular finding. He says that "slight" negative pressures are "quite common" in many otherwise normal ears, and even values more negative than –100 daPa are routinely encountered. He presents data for 142 normal ears, and his graph shows that for people aged 6 to 59 about 5% have Type C tympanograms. The figure rises markedly for children aged 2 to 5, where about 30% show Type C tympanograms, although the ears are still classed as "normal". In other words, Type C tympanograms occur regularly, and this raises the question of where the "negative pressure" comes from.

Evidence that actual middle ear pressure is close to zero

Much has been written about negative middle ear pressure but a common problem is that most experimenters almost invariably interpret negative pressures as recorded by a tympanometer as an actual state of suction in the middle ear. In this section we systematically address the measurements that have been made and suggest that, in most situations, the true state of affairs is a middle ear pressure close to zero – but associated with a substantial inwards pull of the tensor tympani, which masquerades as negative pressure inside the middle ear.

1. The effect of swallowing

The middle ear can in large part be considered a sealed cavity, but whenever we swallow the Eustachian tube connecting the middle ear cavity to the back of the throat opens. Figure 4 illustrates the arrangement.

If there is indeed a pressure difference between the middle ear and the atmosphere – such as during an aircraft descent or travelling several floors in a lift – we sense the pressure and automatically swallow, at which point we hear our ears 'pop'.

A number of studies of direct middle ear pressure measurements – made with an invasive technique involving the puncturing of the tympanic membrane with a needle and measuring the pressure via tubing connected to a manometer – confirm that the middle ear pressure returns to zero after swallowing (e.g. Tideholm [6]), at least in normal subjects. Although the invasive technique is difficult to apply for long periods, results tend to confirm that normal subjects usually have middle ear pressures not far from zero. The more complex situation of otitis media is taken up in later discussion.

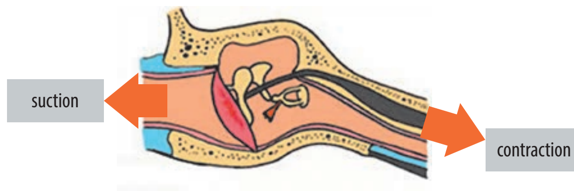

Figure 3. Relation between the eardrum (red) and its attachment to the tensor tympani (dark brown). When the tensor tympani contracts, it pulls on the malleus and the attached eardrum, pulling them inward. Based on the area of the eardrum, a contractile force of 5 grams weight will be needed to counterbalance a suction in the ear canal of about –100 daPa in order to keep the eardrum in a neutral position. Adapted from a figure by MF Dauzvardis, Loyola Medical School

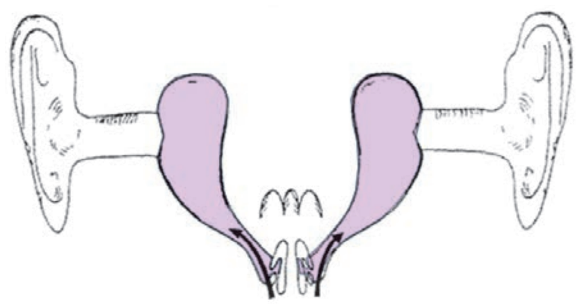

Figure 4. The middle ear opens via the Eustachian tube to the throat whenever we swallow, an action that establishes zero pressure across the eardrums. Under this condition, tympanometry should now register zero middle ear pressure. Adapted from Sadé [5]

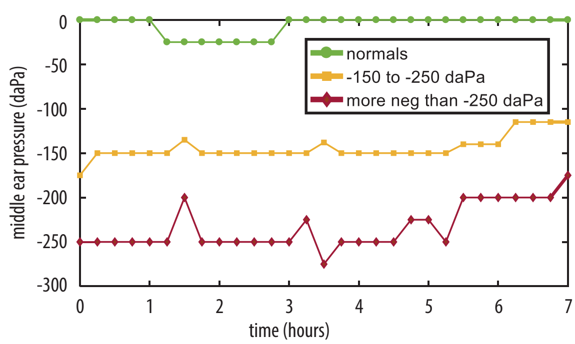

Figure 5. Tympanometry readings taken every 3 minutes for 7 hours in three groups of subjects (plotted as 15 minute averages). The groups were: normals (*n* = 5); patients with tympanometric pressure –150 to –250 daPa (*n* = 6); and patients with pressure more negative than –250 daPa (*n* = 7). Over the whole 7 hours, none of the patients' 3-minute measurements ever reached zero. Replotted from Grøntved [7]

Since we automatically swallow every minute or so, one might expect that it would be very difficult to sustain a middle ear pressure much away from zero. Surprisingly, however, monitoring of 20 patients who complained of hearing loss found that, measured tympanometrically every 3 minutes for 7 hours, their average pressure was –150 mm of water, and that for two-thirds of them (13 or 65%) the pressure never hit zero [7]. For 5 control subjects, the pressure did not deviate far from zero. See Figure 5.

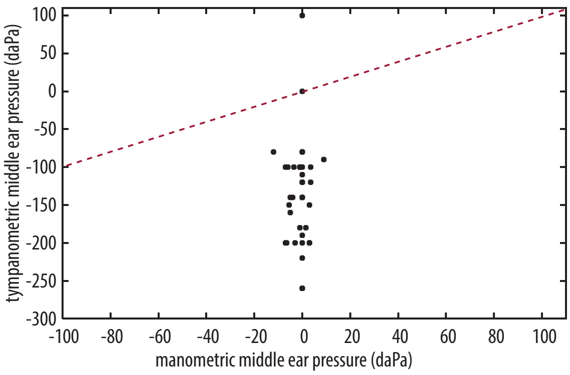

Figure 6. Lack of correlation between tympanometric readings (*y*-axis) and concurrent measurements using a needle and manometer (*x*-axis) in 36 subjects with secretory otitis media. If there were a correlation, it would show as a 1:1 plot (dashed red line). However, in this case the points are far removed and the *r*-squared value is minute (less than 0.0001). Data from Fig. 2 of [34]

Grøntved and colleagues [7] interpreted the fact that the 13 patients who "never equalized their negative middle ear pressure" had Eustachian tubes which did not open during the test period, but there is room for doubt, and here we suggest an alternative explanation. The authors are puzzled that even though the tube never appeared to open during 7 hours of measurement, the recorded tympanometric pressure in any patient never reached –600 daPa, the supposed equilibrium pressure of the middle ear mucosa. They remark that a pressure more negative than –150 mm is considered an indicator of serous otitis media and hence a candidate for placement of a ventilation grommet in the tympanum – the traditional treatment for clearing infection and relieving negative middle ear pressure.

2. Poor correlation between tympanometry and manometry

In order to understand the unusual tympanometric findings, one approach is to use both tympanometry and a manometric needle one after the other on the same set of patients. Figure 6 shows the results in 36 ears of children who were about to have a grommet inserted for treatment of serous otitis media (SOM) [34]. The manometry showed an average pressure of just –1.7 mm water (effectively zero) whereas tympanometry showed an average pressure of –130 mm water, two orders of magnitude larger. These data indicate that something is seriously awry in our interpretation of tympanometry. To add weight to that statement, Sadé and colleagues later made tympanometric measurements after grommets had been inserted [34]. The natural expectation is that the ventilated ear should show zero pressure. Remarkably, Sadé reports that "tympanometric measurements did not change after insertion of the ventilating tube" (p. 61) [34]. He appears unperturbed by these findings and simply concludes that "tympanometry … does not reflect in these cases the real intratympanic pressures." Whether grommets actually help overcome SOM is a separate matter, but we certainly need a better understanding of what negative middle ear pressure really means. Although Sadé's work was done towards the end of last century, there are still recent reports which claim that negative middle ear pressures are indicative of SOM and the need for grommets. It is a concern that most patients with retracted ear drums do not complain of ear pressure [5]. The effect of grommets on otitis media is considered later, but the important point is that negative middle ear pressures may simply reflect different degrees of tensor tympani contraction, and so negative pressures may well be a fairly normal circumstance.

3. Of 'blocked' Eustachian tubes

The major implication of this revised outlook is that there have been many cases of negative middle ear pressure in the past which have been falsely diagnosed as blocked Eustachian tube or Eustachian tube dysfunction – the standard interpretation of negative pressure, based on the observation that a truly blocked Eustachian tube leads to absorption of gas by the mucosa lining the middle ear cavity and creates actual suction (the supposed equilibrium pressure of the middle ear mucosa is presumed to be –600 daPa [7]). This new perspective casts doubt on such a conclusion, and in the course of this paper more evidence is marshalled that measurements of middle ear pressure are often better interpreted as tension from the tensor tympani.

However, returning to address the question of "blocked" Eustachian tubes, it is indeed true that subjects with otitis media have pressures in the negative range (as high as –250 daPa; Tideholm [6]). Some studies in which a needle has been used to penetrate the mastoid or the tympanic membrane have confirmed this result [8], although the results are open to interpretation because of problems with leaks around the puncture site. It is difficult to say, without dedicated experiments, the extent of this misdiagnosis, but there are some useful papers that shed light on the issue, and this section looks closer at them.

Misdiagnosis of a blocked Eustachian tube is not a trivial matter. Gaihede and colleagues [9] report that treatment with ventilation tubes (or grommets) – the standard response – was associated with a 66% rate of tympanic membrane pathology compared to untreated ears (12%).

Magnuson [10] discusses the subject of "atelectatic ears" in children (those showing retraction pockets) and casts doubt on the classical "hydrops ex vacuo" theory in which middle ear effusion (serous otitis media) is presumed to result in negative pressure. He relates how experimental and clinical data are inconsistent in trying to find a causal relationship between tubal obstruction, high negative pressure, effusion, and tympanic membrane retraction. Some prior experiments had shown that negative pressure developed by gas absorption is negligible. In his own work, retraction pockets were visible in large numbers of ears even though they had been repeatedly treated with ventilating tubes. The factors involved were complex, but a consistent observation was an inability to equalize negative intratympanic pressure. This suggests that the inwards pull of the tensor tympani may be able to act on the eardrum in such a way as to create retraction pockets.

A hypothesis consistent with the findings is that infection of the middle ear (otitis media) leads to irritation of the middle ear muscles, and this in turn causes the tensor tympani to contract and generate an inward force which mimics negative middle ear pressure. There is good evidence supporting this interpretation: histopathology of the muscle in cases of otitis media has shown more inflammatory cells and more hypercontracted fibers [11]. The authors of this study, which involved 105 temporal bones, also found that the muscle fibres displayed contracture knots consistent with hypercontraction. Why otitis media develops so commonly in young children but progressively disappears with age is an area of research that would pay large dividends in terms of child health and hearing preservation. In any case, the immediate message should be that a negative middle ear pressure is most likely a sign of an irritated tensor tympani and that the condition should not be used as an automatic indicator for the insertion of grommets.

4. Subjects with patulous Eustachian tube

Normally, the Eustachian tube opens whenever we swallow (Figure 4), and after a few swallowings the middle ear pressure is usually very close to zero [12, 13]. Virtanen has systematically investigated the opening by placing a sound source in the nostril and a microphone in the ear canal (a method called sonotubometry). Using such a technique, opening of the Eustachian tube can be directly measured by recording a sudden increase in microphone level. This novel approach is useful for finding out what is going on in cases of so-called patulous Eustachian tube where the tube appears to be more or less stuck open, an annoying condition where people complain of their own voice being too loud or echoey, they can hear their own breathing, and everything sounds like their head is an empty barrel. There can also be sensations of fullness or pressure.

Virtanen used sonotubometry to study cases of patulous Eustachian tube in 30 patients who had a wide range of symptoms and response patterns [12,13]. There were 8 subjects who appeared to have Eustachian tubes that were more or less continuously open, and, remarkably, one subject had a measured middle ear pressure of –200 mm water, which seems physically impossible. This supports the interpretation being put forward here that really there is an active tensor tympani at work whose steady level of contraction masquerades as negative middle ear pressure under tympanometry. Confirming this interpretation, this same patient also had a perforated tympanic membrane, meaning it was absolutely impossible for a pressure difference to exist between the eardrum and the middle ear. [Note that the hole must have been quite small for the tympanometer not to have recorded a peak at zero pressure during its rapid pressure sweep; the same methodological consideration suggests that the true (hole-free) reading must have been even more negative than the reported value.] Another patient reported by Virtanen had an Eustachian tube that closed only slowly after swallowing, and this patient too had a perforated tympanic membrane; once again, though, the middle ear pressure registered as markedly negative: –100 mm water.

These cases are not just isolated aberrations. A few years later, Virtanen [14] published a survey of 92 healthy adults who were just recovering from a common cold. Some had a blocked Eustachian tube while others did not, and all subjects underwent sonotubometry and tympanometry. Based on mismatches between the two tests, Virtanen concluded that "a negative middle ear pressure evaluated by tympanometry does not always denote a closed tube" (p. 766) [14]. An illustration in his paper shows a tympanogram with a peak at –155 mm water and an accompanying sonotubometry trace showing the Eustachian tube opening. More generally, a supporting table shows that 21 of 28 subjects (75%) had positive sonotubometry (opening of the Eustachian tube) and middle ear pressure of –51 to –75 mm water, with other categories of negative middle ear pressure ranges also showing that an open Eustachian tube is compatible with sustained sub-zero pressures. For example, even when the middle ear pressure was more negative than –75 mm water Virtanen still observed some tubal openings.

Cases of patulous Eustachian tube function are complex, and the tube is more than just a valve that opens and shuts. Ventilation appears to involve complex coordination of muscles – the tensor veli palatini and its close anatomical companion, the tensor tympani [11,15,16]. Magnuson [17] believed the real function of the Eustachian tube to be "pressure regulation", not just periodic ventilation. It seems that dysfunction of these two interrelated muscles somehow leads to patulous Eustachian tubes, where "patulous" can have a range of functional meanings. One interpretation from Virtanen's work is that heightened activity of the tensor veli palatini is accompanied by additional activity of the tensor tympani, and this can be recorded as negative middle ear pressure – even though the true pressure inside the middle ear may be zero.

In this context, Magnuson [17] recorded some interesting side-effects of 42 subjects who seemed to have patulous Eustachian tubes and who used a "sniffing" technique to temporarily overcome their symptoms. Sniffing, which is a way of evacuating air from the middle ear, was found to have two notable side-effects. 1) It overcame hyperacusis: with "drums out" after swallowing, the subject's voice was over-loud; with "drums in" hearing was more comfortable, natural, and distinct. 2) It reduced "fluttering" of the eardrums, which is the same sensation most people experience when they activate their tensor tympani by yawning or tightly closing the eyes. Finally, there was a third related observation: 3) instead of sniffing, performing a Valsalva enabled most patients to increase their hearing acuity and perceive weak sounds. Importantly, all three side-effects are compatible with the intralabyrinthine pressure theory [3] which suggests that middle ear muscles adjust hearing gain through varying the pressure of fluid inside the cochlea. Understandably, if the tensor tympani is not operating properly, another way to control intralabyrinthine pressure (and hearing acuity) is to vary middle ear pressure – by sniffing or Valsalva manoeuvre – actions which will cause the ear drum to bulge out or be pulled in, in turn causing the ossicular chain and ultimately the stapes to increase or decrease intracochlear pressure. Viewed in this light, patulous Eustachian tube could be a misnomer: it is actually the dysfunction of the tensor tympani which is giving rise to the annoying symptoms.

Clearly, much more work is needed to work out exactly what is going on, but the accumulated evidence is that, across a range of subjects, contraction of the tensor tympani can, through pulling the eardrum inwards, accurately mimic negative middle ear pressure.

Evidence that changes in pressure are really changes in tension

The central hypothesis put forward here is that changes in middle ear pressure as measured tympanometrically are really a reflection of changes in the state of tension of the tensor tympani muscle. Like any isometric muscle, that tension will tend to vary from one individual to another and from time to time. The evidence can be categorised under the following headings.

1. Rapid changes in values

A good indication that a tympanometer measures muscle tension rather than pressure of the middle ear is the magnitude and rapidity with which meter readings change. Gaihede and Ovesen [18] reported pairs of tympanometric measurements made immediately after one another (without removing the probe). Among a set of 80 ears they found a standard deviation of the difference of 7 daPa, a measure that was larger than the resolution of their tympanometer (5 daPa). When follow-up readings were made several months later on 20 ears, the standard deviation was now 17 daPa. In one case, the initial reading was –45 daPa and it increased to –240 daPa at follow-up. The authors mention literature reports where middle ear pressures have been found to change by up to 30 daPa within minutes, and that day-to-day shifts from Type A to Type C are well known. Clearly, we are dealing with a dynamic system, and this is more likely to reflect the action of a muscle than absorption of gas from a mucous lining.

2. Findings from Meniere's disease

A striking result that reinforces the muscle interpretation is the study by Park [19] of middle ear pressure in normal and in Meniere's patients. It has already been suggested that Meniere's disease is caused by excessive activity of the tensor tympani [4], so it is highly relevant that the mean middle ear pressure of 30 normals was found to be 4 daPa (±5 daPa) whereas the figure for 33 Meniere's patients was –43 daPa (±75 daPa). It would be revealing to follow the course of tympanometric pressure prior to, and after, a Meniere's attack.

Historically, it is of interest that there has long been a suspected association between middle ear pressure and Meniere's disease, beginning perhaps with the insights of Tumarkin in 1966 [20]. This author rejected the orthodox notion that the source of "labyrinthopathies" (including Meniere's disease) must be found in the inner ear. Instead, he boldly suggested, following some earlier opinions, that the cause of Meniere's disease may result from middle ear dysfunction. It should be noted that Tumarkin's explanation does not explicitly mention the tensor tympani, but he does point to negative middle ear pressure as a key factor. Unconventionally, he tried inserting grommets into the tympanic membrane to treat 20 cases of vertigo, and reported that the results were "little short of startling", with vertigo disappearing entirely in practically every case.

Tumarkin's paper attracted considerable interest, and at least 36 surgeons tried the procedure, with generally positive outcomes [21]. However, enthusiasm waned and grommets have not turned out to be a cure for Meniere's disease, although they have remained popular for treating otitis media. The point of interest, however, is that many investigators have confirmed that a large proportion of Meniere's patients have what has been called "intermittent Eustachian tube blockage". Hall and Brackmann [22] found that 25 of 81 Meniere's patients (31%) had a negative middle ear pressure of –100 mm water or greater, which, following the conventional picture, is interpreted as "a blocked Eustachian tube". Given what has been said to this point, however, it becomes questionable whether the Eustachian tube really is blocked.

Hall and Brackmann's paper is particularly illuminating, however, in finding that the middle ear pressure varies in line with the strength of the symptoms. For example, one case showed no symptoms when the middle ear pressure was zero, mild symptoms (slight feeling of fullness, vertigo, tinnitus) when the pressure was measured as –125 mm water, and with strong symptoms the pressure was –175 mm water. They also relate how the low-frequency hearing of some patients could by temporarily improved by 5–15 dB by increasing middle ear pressure via a Valsalva manoeuvre.

These are strong indications. Intriguingly, however, a later paper by one of the same authors [23] backs away from the association between negative middle ear pressure and Meniere's, claiming only that, based on a larger sample, the incidence of Type C (or B) tympanograms was only about the same as what Jerger found in normals. We are not told much about the change in outlook apart from enlarged numbers, but given the strong findings of the earlier work, and the equally strong findings reported later by Park (2012) [19], one might reasonably suspect that something peculiar is going on and that perhaps a key factor may have been missed. Closer investigation might provide the necessary clues, and in any such work the tensor tympani hypothesis stands as a good candidate for resolving these peculiar anomalies.

The difficulty associated with Meniere's disease is that it is such a multifactorial disease, and, despite dissenting voices, most research is still focused on the inner ear (see the review by Oberman [24]). Nevertheless, it is encouraging that Oberman's review reminds us that the middle ear muscles remain in the picture (p. 259). It is suggested that the pressure model as described by Bell [4] might help in resolving the matter.

3. Voluntary contraction of middle ear muscles

Logically, perhaps the clearest and most direct evidence in support of the hypothesis under consideration is that gathered from those rare individuals who can voluntarily contract their middle ear muscles.

An earlier paper [4] has already set out an array of evidence that voluntary contraction of the middle ear muscles leads to a loss in hearing sensitivity at low frequencies – both for air conduction and, tellingly, bone conduction. The explanation offered was that contraction of the tensor tympani increased hydraulic pressure in the labyrinth, and a reduction in gain of the cochlear amplifier (see Bell [3] for

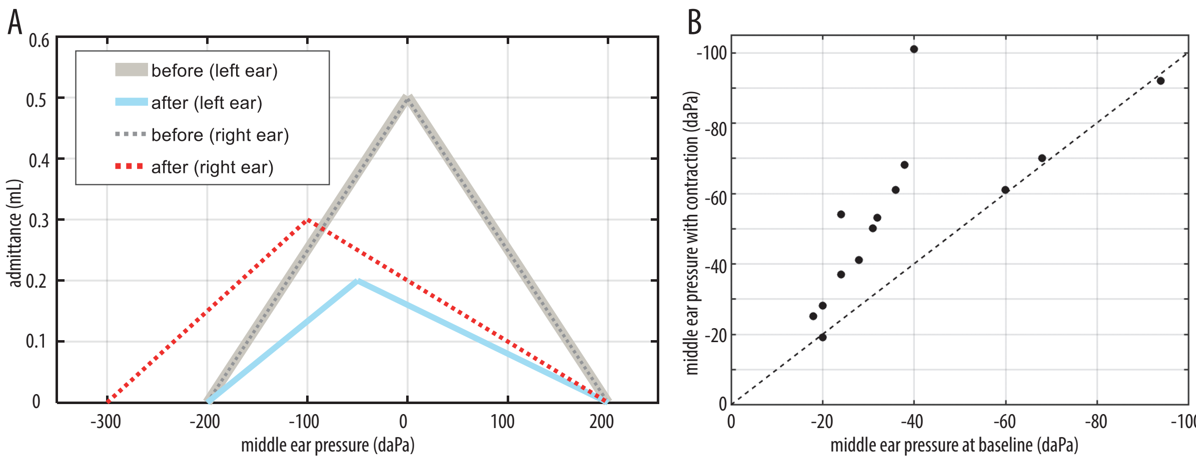

Figure 7. Effect of middle ear muscle contraction on tympanometric peak pressure. (A) Data for a single subject before and during voluntary contraction: the peak shifted from 0 to –75 daPa for the left ear and 0 to –100 daPa for the right. (B) Data from 14 ears of 8 subjects who could voluntarily contract their middle ear muscles. The x-axis shows middle ear pressure before contraction; y-axis shows middle ear pressure with contraction. The diagonal is the 1:1 line. [Data in A from [25]; B from [26]; with permission]

more detail). The general point was that Meniere's disease might be due to dystonia or cramp of the middle ear muscles. In line with that thinking, the question now becomes does voluntary contraction also cause an apparent increase in negative middle ear pressure as measured tympanometrically? Indeed, to get straight to the point, all the indications are generally positive, and the rest of this section outlines published work that directly supports this relationship.

A prime example of what happens when a subject contracts their tensor tympani is shown in Figure 7A. The data comes from Angeli [25] and shows the negative shift in tympanometric peak pressure when the subject's middle ear muscles are contracted. When the muscle is relaxed the peak occurs at zero pressure, but when the muscle is contracted it occurs at –50 daPa (in the left ear, blue) and –100 daPa (in the right, red). An average figure of –75 daPa corresponds to the pressure needed to counteract an inward force of about 4 g wt, a direct and credible interpretation.

More substantial evidence comes from a study by Aron and colleagues [26] which investigated 8 subjects who were also able to contract their middle ear muscles. The tympanometric results for 14 of their ears are shown in Figure 7B which shows middle ear pressure readings before and during contraction. In most ears tested, the apparent middle ear pressure became more negative during contraction, shifting from an average baseline pressure of –38 daPa to –54 daPa, a change which was statistically significant ($p < 0.01$). Of course, a natural inference is that even the initial baseline values represent a standing level of tension of the tensor tympani. As it happened, the main focus of Aron and colleagues was not so much on middle ear pressure as on middle ear compliance, particularly how it may be affected by the stapedius reflex. Nevertheless, despite the slightly different focus, the researchers do explicitly conclude, after conducting analogous experiments in cadavers, that "Pulling on the TT [tensor tympani] resulted in a more negative measured peak MEP [middle ear pressure] measurement" (p. 377) [26]. They also made the hypothesis that "a TM [tympanic membrane] tensed by contraction of the TT requires relatively more negative EAC [external ear canal] pressure to bring it back to its most compliant state, so that peak admittance is measured at a more negative EAC pressure" (p. 379).

Given this insight, it is surprising that there has not been a thorough investigation of the role of the tensor tympani in establishing negative middle ear pressure. Part of the difficulty involves understanding that the tensor tympani could exist in a state of continuous long-term contraction. In analogy with the acoustic reflex of the stapedius, the thinking seems to have been that the tensor tympani undergoes reflex contraction in a similar way, and that there does not seem to be any point in having the muscle contracted for any extended period of time. However, the idea of the muscle being in an isometrically contracted state so that it can immediately respond to ambient sound and appropriately adjust hearing sensitivity casts fresh light on the true function of this long-neglected and misunderstood muscle.

Another part of the perceived difficulty may derive from the unappreciated greater stiffness of dead muscle compared to that which is living. Thus, Aron and colleagues find (Fig. 9 [26]) that a temporal bone specimen required a substantial force of 60 g wt to resist an ear canal pressure of 100 daPa, whereas calculations based purely on ear drum area and an active, compliant muscle indicate that a force of 5 g wt should be sufficient to counteract the forces involved. It appears that living tissue is much more responsive than dead, and this seems reasonable.

Further work on the tympanometric effects of tensor tympani contraction can be found in Bance [27] and Wickens [28], but the key measure in these investigations is again compliance rather than middle ear pressure as such. Not unexpectedly, there is a general correlation between the two, so that higher pressures (positive and negative) are associated with greater stiffness (lower compliance), but inspection of the two reports just cited indicates that there sometimes appears to be a disjunct. Nevertheless, the findings are generally consistent with an active tensor tympani. In Bance [27], tympanograms were collected before and after voluntary contractions from 2 subjects, but unfortunately not during them (sustaining a strong contraction isn't easy). Stimulation of the tensor tympani by a puff of air to the eye produced increases in impedances and in air- and bone-conduction thresholds, but, curiously, other means of stimulation – such as stroking the subject's cheek or asking them to subvocalise (count silently) – did not. The second work [28] reported on 5 subjects who could all voluntarily contract their tensor tympani muscles for seconds at a time, allowing tympanometry to be done during a contraction, and again air- and bone-conduction thresholds were affected.

4. Multifrequency tympanometry

More recently, use of tympanometry has in many cases been replaced by tympanometry based on a range of frequencies rather than just the standard 226 Hz impedance measures [29]. Often the technique is called multifrequency tympanometry [30], wideband acoustic immittance [31], or wideband absorbance (WBA) tympanometry [32]. The technique supplies an extra frequency dimension to the conventional tympanogram, providing a colourful 3D plot and additional diagnostic information. However, there is a ridge-line corresponding to maximum admittance where the middle ear pressure balances the ear canal pressure, and this ridge-line is, as expected, largely independent of frequency. Thus, the middle ear pressure can be recorded in much the same way as with the traditional method.

Useful insights come from a recent paper [32] where Karuppannan and Barman used WBA tympanometry to explore cases of abnormal middle ear pressure, including 30 cases of negative middle ear pressure in 25 adults (average pressure of –207 daPa). The adults had no active ear discharge, so the confounding factor of otitis media, often seen in children, could be ruled out. Moreover, the inference can be made that the subjects did not have 'blocked Eustachian tubes', which is the usual diagnosis made in pediatric cases when high negative middle ear pressures are recorded. Although it is true that a blocked Eustachian tube can lead to negative middle ear pressure, that does not mean the reverse is the case, and it seems that conflating the two has led to erroneous conclusions. This paper has sought to establish that negative middle ear pressure can occur quite normally as a consequence of sustained tensor tympani contraction, and it can be presumed that this is what happened in Karuppannan and Barman's subjects.

The authors measured wideband absorbance under two conditions: at zero ear canal pressure and at a pressure that exactly counterbalanced the tympanometrically measured middle ear pressure. In this way, they were able to see the effect that middle ear pressure (or, as it is suspected, tensor tympani contraction strength) was having on the ear drum. Their results (their Fig. 2; Figure 8 here) showed that there was a large difference in absorbance between the two conditions at low frequencies (0.2–1 kHz); however, we see that the absorbance is almost identical from 3–6 kHz. Importantly, this suggests a possible way of separating the effects of a tensor tympani contraction from the effects of actual air pressure within the middle ear. Although both factors will naturally stretch the ear drum and change its acoustic impedance, it is clear from Figure 8 that they operate over different frequency ranges. It is suggested that the eardrum's tension (and impedance) changes as a function of middle ear pressure, and this can be seen as a change in absorbance from 0.2–1 kHz. However, for subjects whose tensor tympani is already contracted, the absorbance at 3–6 kHz is independent of pressure differences across the eardrum – the WBA has plateaued – and so in normal subjects it might be expected that this range is still somewhat sensitive to the state of the tensor tympani.

The above interpretation can be supported by comparison with the work of Shaver and Sun [33] which involved normal subjects who were able to adjust their actual middle ear pressure via a Toynbee manoeuvre (swallowing while keeping the nose pinched). The authors found (their Fig. 1 and 2; Figure 8 here) that the change in actual middle ear pressure, to a mean of –160 daPa, changed ear canal absorbance in the sensitive 0.2 and 1 kHz range, but also, it should be noted, over the 3–6 kPa range, where Karuppannan and Barman saw no change. (Actually, Shaver and Sun measured reflectance, but turning their plots upside down gives absorbance.) That is, both sets of experiments involved similar middle ear pressures, but in one case it was actual Toynbee-induced pressure while in the other it was a persistent condition. The difference in WBA at 3–6 kHz, it is suggested, relates to the state of the tensor tympani: in the clinical group the muscle is strongly active, but in the normals it is more relaxed, and so the difference in this frequency range could be a sensitive way of separating out the degree of tensor tympani involvement.

The conclusion is that it should be possible, by using a combination of techniques and subject groups, to separate the individual effects of actual middle ear pressure and tensor tympani contraction. This would be a real achievement in unpicking the paradoxes that have accumulated within the tympanometric field.

Discussion and conclusions

The above text suggests that negative middle ear pressure should, in the first instance, be interpreted as an inwardly directed force created by steady contraction of the tensor tympani. Physically, there is no actual negative pressure in the middle ear; it's just that a tympanometer will react to any muscle-generated force in the same way as if there were. If true, there are a number of major implications of this new interpretation.

It is surprising that negative middle ear pressure has been such a common measure in audiology but has always been interpreted so literally despite persistent anomalies that directly indicate otherwise. As suggested, one reason for the neglect comes from our lack of appreciation of the important role the tensor tympani plays in regulating inner ear pressure, and thereby controlling auditory sensitivity. The muscle, because it is isometric, doesn't appear to 'do' much (other than just tensing the tympanum) and so it has generally been overlooked. Together with the

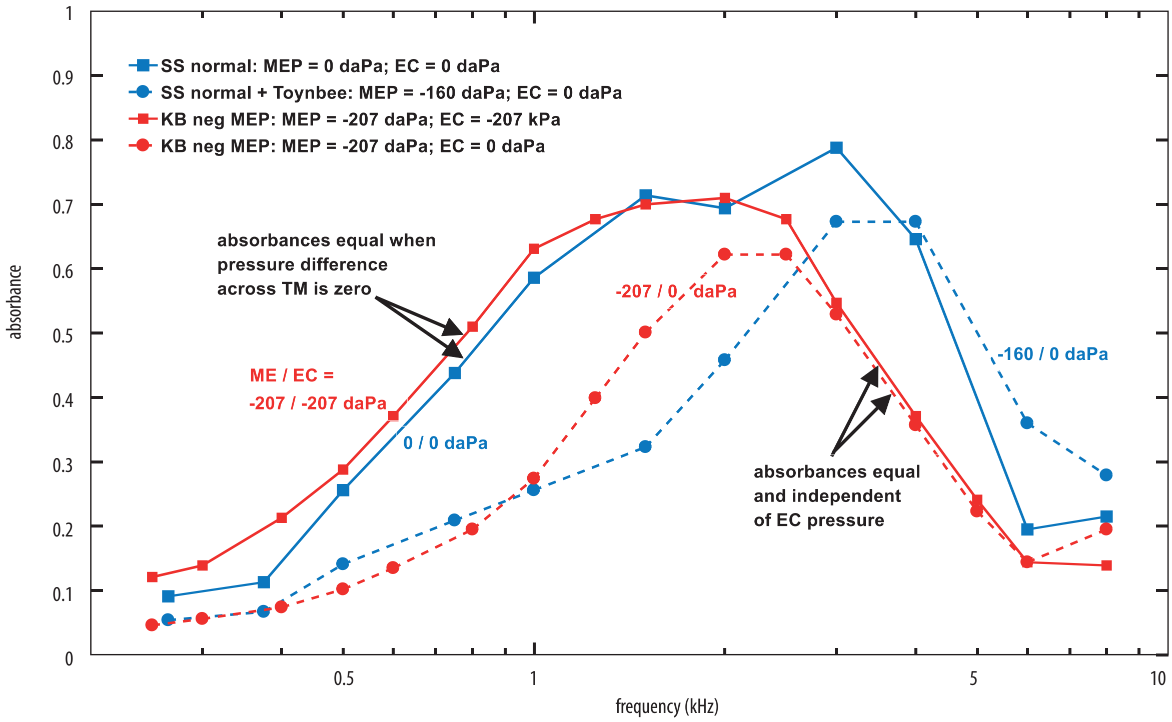

Figure 8. Similarities and differences in wideband absorbance (WBA) between two groups of subjects, both with negative middle ear pressure but of different origins. The first group were 11 normal adults who used a Toynbee manoeuvre to generate an average middle ear pressure of –160 daPa (blue lines, Shaver and Sun [33]); the second group were 25 adults (30 ears) from an audiological clinic diagnosed with negative middle ear pressure averaging –207 daPa (red lines, Karuppannan and Barman [32]). Note the similarity in WBA between 0.2 and 1 kHz (continuous lines) when there was equal pressure on either side of the eardrum. Similarly, the WBAs for both groups are seen to be similar between 0.2 and 1 kHz when the pressure is about 180 daPa across the eardrum (dashed lines). Note however the differences between 3 and 6 kHz: WBA is virtually independent of ear canal pressure for the clinical group, but there are appreciable differences for the normal group. It is suggested that the WBA differences at 3–6 kHz relate to the state of the tensor tympani: for the clinical group the muscle is strongly contracted but for the normals it is not.

unappreciated role of intralabyrinthine pressure, the two elusive factors have prevented research from moving forward. Clearly, much more research is needed to explore the details of these subtle mechanisms.

The central idea behind this paper is that tympanometric peak pressure is made up of two components: actual middle ear pressure, which in most commonly around zero, and tensor tympani tension, which gives rise to an effective "negative pressure" as it pulls the eardrum and whole ossicular chain inwards. Up until now, these two components have been conflated – the tympanometer reading has been interpreted as being solely due to pressure inside the middle ear – which has given rise to the paradoxical findings set out above. Otitis media has often been diagnosed on the basis of negative middle ear pressure; however, it is suggested here that the negative middle ear pressure may be in fact be a side-effect of bacterial infection: irritation of the middle ear muscles due to infection causes the tensor tympani to contract, and this contractile force is likely to be what the tympanometer is reacting to [11].

In order to set matters straight, we will need to have a way of measuring actual middle ear pressure separately from the contractile state of the tensor tympani. Invasive measures like penetrating the middle ear space with a needle [8] are far from satisfactory, and our text suggests that careful use of wideband absorbance techniques – selecting frequency ranges which are characteristic of the impedance of the eardrum and of the tensor tympani – may provide a solution. The ranges 0.2–1 kHz and 3–6 kHz may be good starting points.

It is hoped this paper may help overcome some obstacles in tympanometry and allow further progress to be made. The tensor tympani is a neglected, but essential, component of our intricate hearing system – it is at the heart of an active gain-regulation loop – and tympanometry is a sensitive but powerful tool for understanding how it works.

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