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Review Article

Cochlear deformities and its implication in cochlear implantation: a review

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ABSTRACT

Hearing loss is one of the world's leading causes of chronic health conditions. Cochlea plays a vital role in the hearing mechanisms and it converts sound energy into electrical stimuli which are transmitted to the brain through the neural pathway. The human cochlea is difficult to explore because of its vulnerability and bordering bony capsule. Congenital malformation of the inner ear or cochlea is an important cause of congenital sensorineural hearing loss. The deformity of the cochlea may result from arrested development of cochlea at different stages of fetal life or from abnormal development due to genetic abnormalities. There are hair cells responsible for converting sound energy into electrical impulses. These hair cells are easily damaged, which results in permanent hearing loss. Cochlear implants are surgically implantable biomedical devices that bypass the sensory hair cells and directly stimulate the remaining fibers of the auditory nerve with an electric current. Cochlear implantation is capable of restoring a surprisingly large degree of auditory perception to patient that is suffering from severe to profoundly deaf. Children with cochlear anomalies are thought to have poorer outcomes with cochlear implantations, therefore would be poorer candidates due to their diminished ability to interpolate and use auditory information provided through a cochlear implant. Parents should be counselled to establish realistic post-implant expectations in case of children with cochlear deformity. So, patient selection has emerged as one of the most vital determinants of successful outcomes after pediatric cochlear implantation.

Keywords: Cochlea, Inner ear, Organ of Corti, Cochlear deformity, Cochlear implant

INTRODUCTION

The cochlea of the inner ear is a complex three-dimensional structure where the sound energy is coded by the sensory hair cells into the electrical impulses traveling along the auditory nerve to the brain.[1] Anatomical malformation of the human cochlea is an important topic for the otologist that needs to be well understood before performing the cochlear implantation. Anatomical abnormality of the temporal bone is often associated with poor hearing.[2] Increased experiences in cochlear implantations have led to more children with abnormal cochleovestibular anatomy as a candidate for cochlear implantation. Cochlear implants are surgically implantable biomedical devices that bypass the sensory hair cells and directly stimulate the remaining fibers of the auditory nerve with an electric current.[3]

Cochlear implantation is capable of restoring a surprisingly large degree of auditory perception to patients that are severe to profound hearing loss.[4] Cochlear malformation of the inner ear is a common etiology for sensorineural hearing loss.[5] The congenital malformation of the cochlea may occur due to arrested development at different stages of fetal life or from abnormal development due to genetic abnormalities.[6] The cochlear deformities may occur in the membranous and/or bony labyrinth and vary widely in severity. The

purpose of this review article was to review the normal anatomy of the human cochlea and cochlear deformity with its implications in cochlear implantation.

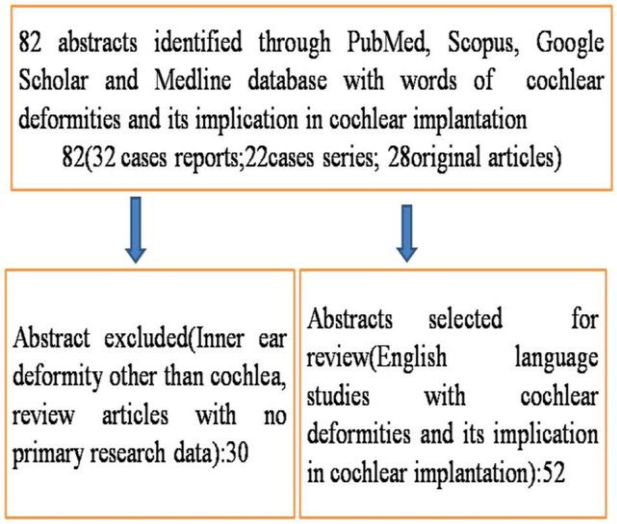

Figure 1: Flow chart showing methods of literature search.

Methods of literature search

Multiple systematic methods were used to find current research publications on the cochlear deformity and its implications in cochlear implantation. We started by searching the Scopus, PubMed, Medline and Google Scholar databases online. A search strategy using PRISMA (preferred reporting items for systematic reviews and meta-analysis) guidelines was developed. This search strategy recognized the abstracts of published articles, while other research articles were discovered manually from the citations. Randomized controlled studies, observational studies, comparative studies, case series and case reports were evaluated for eligibility. There were a total number of articles 82 (32 cases reports; 22 cases series; 28 original articles) (Figure 1). This paper focuses only on the cochlear deformity and its implications in cochlear implantation. This review article describes the epidemiology, development of cochlea, clinical anatomy, cochlear deformity, imaging, cochlear implantation and complications of cochlear implantations due to cochlear deformity. This analysis provides a better understanding of cochlear deformity and its impact during cochlear implantation. It will also serve as a catalyst for additional study into a newer technique for better hearing output in the case of cochlear implantation of a patient with cochlear deformity.

Epidemiology

The human cochlea was originally described by Bartholomeus Eustachius in 1564.[7] However, the description of the human cochlea was first published by Albinus in Leyden.[8] Abnormalities of the temporal bone have been associated with congenital sensorineural hearing loss since documented by Mondini in 1791.[9] The incidence rate of inner ear malformation radiographically is reported in 20 to 30% of the patients with sensorineural hearing loss.[10] The frequency of reported abnormal temporal bone findings in patients with unilateral sensorineural hearing loss varies from 7 to 44%.[7] One study with a CT scan of the temporal bone in children with unilateral sensorineural hearing loss reported 28.9% malformations of the inner ear and 25% abnormalities in 25% of cases.[11] There are different congenital cochlear deformities found such as Michel deformity, cochlear aplasia, common cavity deformity, cochlear hypoplasia, and Mondini deformity. Common cavity deformity is a rare congenital bony labyrinth malformation associated with profound hearing loss. The prevalence of common cavity deformity as a proportion of cochlear malformation is variable, ranging from 0.7% to 25%.[12]

Development of the cochlea

The cochlea is a complex three-dimensional anatomical structure that has completed its formation of 2.5 coiled turns by 10 weeks of gestation.[13] At this stage of development, there is only the immature patent cochlea duct when seen in the modiolar cross-section. First, there is the formation of the perilymphatic compartment followed by Reissner's membrane and stria vascularis with gross morphological changes that occur during fetal development. There is a differentiation of cochlear hair cells that are found early in development by 12 weeks of gestation before maturation of the three cochlea compartments, the scala media, scala vestibuli and scala tympani.[13] Indeed, the scala tympani and the scalavestibuli are beginning to form as irregular perilymphatic spaces as inner hair cells (IHCs) begin to differentiate in the developing cochlear duct.[14] There is the formation of the perilymphatic spaces that occurs by vacuolization of mesenchymal tissue surrounding the cochlea duct. This process continues for the next 4 to 5 weeks, so that by 16 to 17 weeks of gestations, the perilymphatic spaces become mature size. By 16 weeks of gestation, the scala vestibuli and scala tympani have elongated along the length of the cochlea duct and at the apex become continuous, so forms the helicotrema.[15] The organ of Corti develops from the primordial cochlear duct. Reissner's membrane is a delicate fibrous partition and it is an integral structural component of the cochlea. At 11 weeks of gestation, Reissner's membrane is formed by layers of cuboidal cells lined by mesenchyme.[16]

Surgical anatomy of the cochlea

The cochlea of the inner ear is a complex three-dimension structure, where the sound is coded by sensory hair cells into electrical impulses traveling through the auditory nerve to the brain.[17] There are two cochleae, mirror-shaped, fluid-filled, coiled, fairly symmetrical bony tubes (3.2 to 4.2 cm long) present in the petrous pyramids of the temporal bones. The human cochlea has

approximately 2 ¾ turns. There are extensive individual variations in both shape and size. There is a fluid called perilymph present in the scala vestibuli and tympani communicates with the CSF through the cochlear aqueduct.[18] The bony labyrinth of the cochlea lies in the temporal bone, often compared to the structure of a snail shell. The bony cochlea is a fluid-filled tube approximately 35 mm long in humans, which coils upon itself around a central bony core, the modiolus, making roughly three turns from its base to the apex. The bony cochlear tube is divided into three fluid-filled canals such as the scala vestibuli which is continuous with the vestibule, the scala tympani and the scala media.[19] These canals spiral along the length of the cochlea and are separated from one another by two membranes such as Reissner's membrane and basilar membrane. Reissner's membrane separates scala vestibuli from scala media and reaches from the spiral limbus atop the osseous spiral lamina, a thin bony self-extending from the modiolus, to the lateral cochlear wall.[20] The basilar membrane separates the scala media from the scala tympani and it extends from the spiral lamina to the spiral ligament at the lateral cochlear wall. The resulting anatomic structure in the cochlea is a membranous tube within the bony cochlear tube and is called a cochlear duct; enclosed on the bottom by the basilar membrane, on the top by Reissner's membrane and on the side by the lateral cochlear wall. Because of the presence of a small opening, the helicotrema, at the apex of the cochlea, the scala vestibuli and scala tympani can communicate and share the same fluid called perilymph. The ionic contents of perilymph are similar to that of cerebrospinal fluid with a high concentration of sodium (Na) and a low concentration of potassium (K).[21] The scala media of the cochlear duct contains a fluid called endolymph which contains a high concentration of K and a low concentration of Na similar to that of intracellular fluid.[21] Along the lateral wall of the cochlea, there is a highly vascularized structure called stria vascularis. Stria vascularis plays an important role in the production of endolymph found in the scala media.[22] The width of the different turns differed greatly between the individuals and the cochlea varies as much as 1.5 mm, representing one-third of the total height. There is often an abrupt turning of the cochlea near the carotid area. In some instances, the carotid canal impinged on the anterior cochlear wall. Cochleae with a long basal part of the first turn usually show a more abrupt turning. This situation will affect the force generated by the tip of the electrode on the lateral wall when gliding up the first turn of the cochlea. The mean number of the human cochlea is found to be 2.6, with a range from 2.2 to 2.9.[23] The inner and outer wall lengths differ greatly in the human cochlea. The electrode runs considerably higher up into the cochlea if kept near the modiolus than along the outer wall. This owes to the relatively bigger diameter of the first turn and small dimensions of the modiolus in the second and third turns. Rosenthal's canal is well defined only in the first turn of the cochlea. Thus, the neurons innervating the outer hair cells in the basal turn of the cochlea coincide fairly with the location of corresponding hair cells, whereas more apically, nerves merge into a less well-defined canal with no precise place/frequency alignment. The nerves innervating the hair cells in the third turn are found more basally near the second turn. The length of the outer wall of the cochlea of the first turn is estimated to be 22.6 mm. An electrode around 21 mm usually extends more than one turn if placed along the modiolar wall but less than a turn if placed laterally. An appropriate fitting of a perimodiolar electrode along the inner wall is usually essential to avoid place/frequency misalignment.[24] The different dimensions of the cochlea in human suggests that place/frequency maps should vary considerably between individuals. The apical part of the mammalian map is spatially compressed in comparison to the base.[25] The medial wall of the cochlea is fragile, so it would imperative not to exert pressure on this structure.[26] The length of the organ of Corti corresponds to the extension of the basilar membrane, which is approximately 34 mm. One study presented the measurements of cochlear width ranging from 6.9 to 8.2 mm.[27]

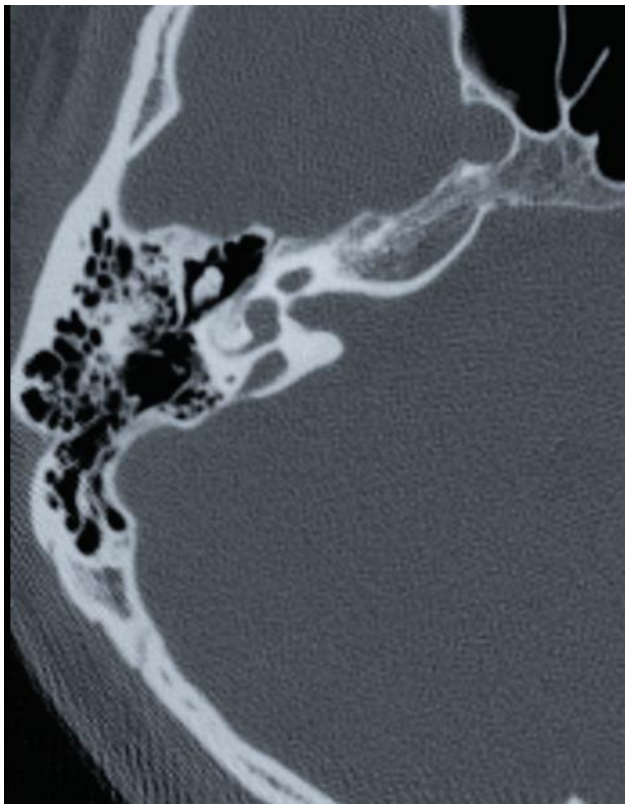

Figure 2: CT scan of the right temporal bone (axial view) showing Mondini deformity of the cochlea..

Cochlear deformity

Abnormalities in the temporal bone are associated with a wide range of hearing impairment, varying degrees of the progression of hearing loss and the presence or absence of associated non-otological anomalies.[28] The congenital cochlear deformity include labyrinthine aplasia/Michel

deformity, a common cavity, cochlear aplasia/hypoplasia and incomplete partition.[29] Michel dysplasia is characterized by a complete failure of development of the inner ear. Scheibe and Siebenmann-Bing dysplasia are characterized by malformation of the membranous labyrinth. Mondini dysplasia was originally described as malformation of the cochlea with shortened to 1.5 cochlear turns with normal semi-circular canals, an angled vestibular aqueduct, and dilated endolymphatic sac (Figure 2).[30]

Since then, the word Mondini has been applied as an umbrella term in routine clinical practice to describe almost any type of congenital cochlear deformity. The Mondini-like dysplasia includes type A: it is a cochlea that consists of two turns with a complete bone at the base of the modiolus; type B: there are 1.5 to 2 turns to the cochlea, absence or hypoplasia of the bony wall at the base of the modiolus (either with or without communication between the cochlea and internal auditory canal) and a complete basal turn.[31] The true Mondini deformity consists of a normal basal turn and distal sac. There is some hearing possible and deafness may be progressive in Mondini deformity. So, these patients become suitable for cochlear implants if sufficiently deaf. A wide vestibular aqueduct is often associated with Mondini dysplasia. Progressive and fluctuant hearing loss usually associated with trivial head trauma is the characteristic feature of the dilated vestibular aqueduct. A single cystic cochlear-vestibular structure is a feature of common cavity deformity of the cochlea (Figure 3). The common cavity deformity of the cochlea has wide phenotypic variability in the internal auditory canal, semicircular canal, and auditory nerve.

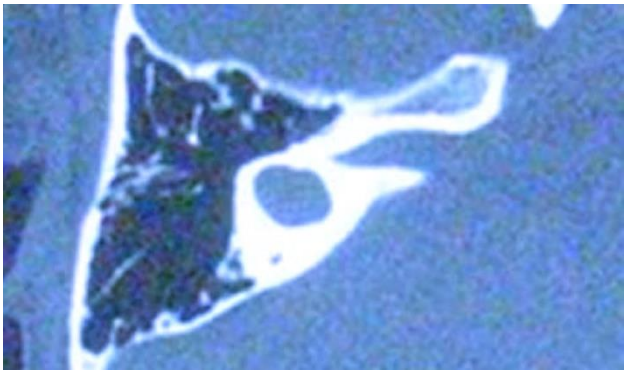

Figure 3: CT scan of the right temporal bone (axial view) showing common cavity deformity of the cochlea.

Imaging of the cochlea

Careful assessment by CT scanning is mandatory for any patient who is being considered for the intracochlear implant. Conventional CT scan can identify the congenital malformations of the cochlea or inner ears such as complete labyrinthine aplasia (Michel deformity), a common cavity, cochlear aplasia/hypoplasia and incomplete partition.[29]

Improvement in high-resolution CT techniques, previously unrecognized bony inner ear or cochlear abnormalities like a large vestibular aqueduct, wide and stenotic internal auditory canal and cochlear nerve canal stenosis has been detected.[29] MRI assesses the audiovestibular nerves and soft tissue densities in the cochlear coils. Each patient for cochlear implantation requires computed tomography (CT) scanning with contiguous 1 mm thick images through petrous temporal bone in the axial and direct coronal planes. The radiological anomalies of the cochleovestibular structures include classifications as: common cavity deformity; incomplete partition; hypoplastic cochlea; vestibular aqueduct enlargement. Common cavity deformity is characterized by a cystic cavity representing both cochlea and vestibule or the presence of a dilated vestibule and cochlea with marked dilatation of ductus reunions effectively making these cavities contiguous. Incomplete partition is defined as a deficiency of modiolus and incomplete septation within a cochlea that consisted of less than 2.5 turns. There is often associated enlarged vestibular aqueduct. Cochlear hypoplasia shows a differentiation into cochlear and vestibular elements and the cochlea is smaller than normal. Enlargement of the vestibular aqueduct is described as an enlarged vestibular aqueduct with more than the diameter of the posterior semicircular canal on axial sections where both structures are adjacent.

Cochlear implant

Cochlear implantation is one of the greatest medical achievements in modern medicine. Cochlear implantation has been used for restoring the hearing of deaf children.[32] There are significant advances in the cochlear implant and implantation techniques. Patient selection has emerged as one of the most important determinants of successful outcomes after cochlear implantation.[33] Preservation of the residual hearing by doing less traumatic surgery and more delicate electrode arrays gives an added technical challenge for the otologist. Cochlear implantation in cochlear deformity makes a difficult situation for the implant team. The large variations in the length of the cochlea angles between turns and position in the skull base can influence the straightforwardness of insertion of the electrode of the cochlear implant, particularly passing the first turn.[34]

A swallow round insertion of the electrode reduces the chance of damage to apical cochlear structures, whereas a deep insertion of the electrode array may improve the cochlear implant performance in case residual hearing is lost. Most commonly damaged portions of the cochlea include the spiral ligament at the junction of the first and second half of the first turn, basilar membrane and osseous spiral lamina.[35] Underdeveloped cochleae may show greater differences in the angle between the first

and second turns and a smaller length of the base of the cochlea as evaluated from a CT scan.[36] There are many reasons for this challenging situation in the case of cochlear malformation. First of all, the distribution of neural tissue is usually unpredictable in the inner ear anomalies and no one can be sure about the outcome after cochlear implantation. In addition, the cochlear nerve may also be absent in the inner ear anomalies. There is also the abnormal course of the facial nerve associated with the cochlear deformity. As a result of the inner ear malformation, especially the lateral semicircular canal, the facial nerve may take an abnormal course and this may sometimes lead to a difficult situation during surgery. Common cavity deformity is usually associated with profound hearing loss where cochlear implantation is challenging.[37] U-shaped electrodes in the common cavity deformity are considered the best position to maximize stimulation of the cochlear neuroepithelium, which is to be located in the inner wall of the cavity near the internal auditory canal fundus.[37] One study suggested a double posterior labyrinthotomy technique that ensures that the electrode is bent along the inner wall of the cavity by pushing and pulling the electrode tip. Recently a custom-made U-shaped electrode was placed through a single slit labyrinthotomy.[38]

Complications due to cochlear deformity

When performing cochlear implant surgery in patients intended for both acoustic and electric hearing, it is necessary to consider the multiple anatomical variations of the cochlea. Surgical complications such as spontaneous CSF fistula and gusher and recurrent meningitis have been reported in patients with cochlear deformity.[39] The CSF gusher may occur due to a defect at the lateral end of the internal auditory canal and it requires special precautions. As a result of the defective internal auditory canal and CSF leakage, these patients may develop meningitis in the postoperative period. Sometimes this defect may result in the insertion of an electrode array into the internal auditory canal.[40]

A sharp bend of the cochlear coiling between the first and second turns may hamper the electrode insertion. It increases the chance for perforation of the cochlear membrane when the tip of the electrode moves from the first to the second turn. A single cystic cochlear vestibular structure is an important feature of common cavity deformity. The prognosis of patients with common cavity deformity after getting cochlear implants is not encouraging when compared with patients with other cochlear deformities.[41] The greatest challenge for cochlear implantation in common cavity deformity is distorted anatomical structure. Both the bony labyrinth structure and spiral ganglion, the target of electric stimulation are absent. The hearing outcomes with cochlear implants in common cavity deformity are generally poor, with the worst outcomes among those with cochlear deformations after long-term follow-up.[42] However, the evidence regarding hearing improvements after cochlear implant surgery indicates that a remnant of the spiral ganglion is also seen in common cavity deformities.[43]

CONCLUSION

The cochlea acts as a transducer that converts sound energy into electrical impulses which travel to the brain via the auditory nerve. The outcome of the cochlear implantation in case of cochlear deformity is not expected as good as the normal cochlea. A high-resolution CT scan and MRI are helpful to detect the cochlear deformity early. The surgeon should aware of the cochlear deformity like Michel deformity, cochlear aplasia, common cavity deformity, cochlear hypoplasia and Mondini deformity before performing the cochlear implantation.

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