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Long-term outcomes of a transmastoid lateral semicircular canal approach to congenital CSF otorrhea in children associated with recurrent meningitis and severe inner ear malformation

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ABSTRACT

Objective: To investigate the long-term effectiveness of transmastoid lateral semicircular canal approach (TMLSCCA) to repair cerebrospinal fluid (CSF) leakage in children associated with recurrent meningitis and severe congenital inner malformation.
Method: A retrospective study was conducted in a university hospital, academic medical center. Fifteen children with recurrent meningitis, secondary to severe congenital inner ear malformation, were included in the study. All of them had CSF associated otorrhea and treated using TLSCCA to repair CSF otorrhea by packing the vestibule with muscle and fascia. Observation of the status of postoperative CSF leakage, recurrence of meningitis and complication were conducted.
Results: None of the cases had recurrent meningitis and CSF leakage after their TLSCCA procedure in the follow-up period of 1–8.5 years. One case presented with transient facial nerve paralysis and completely recovered 3 months later.
Conclusion: TLSCCA for CSF otorrhea in children with recurrent meningitis secondary to congenital inner ear malformation is an alternative approach that offers some advantages.

1. Introduction

Cerebrospinal fluid (CSF) otorrhea is a condition resulting from an abnormal communication between the subarachnoid space (SAS) and the temporal bone [1]. It occurs most commonly with trauma of the temporal bone and in skull base surgery. It is less common in infection and neoplasm and can occur spontaneously without apparent reason [2,3]. By contrast, congenital CSF otorrhea (CCSFO) presents as spontaneous CSF leakage associated with congenital temporal bone defect, as in severe inner ear malformation associated with a fundus defect of the internal auditory meatus (IAM), a dehiscent stapes footplate, a defect in Hyrtl's fissure, or defect of fallopian canal [2–6].

Typical symptoms of CCSFO occur only when there is a perforation in the tympanic membrane. Otherwise, the CSF flows down the Eustachian tube and manifests as a clear rhinorrhea. Therefore, for children with CCSFO, because it is often embedded among other severe clinical issues, such as congenital profound sensorineural hearing loss (SNHL) and vestibular weakness [1], the problem of CCSFO is frequently overlooked until recurrent bacterial meningitis occurs [2].

The etiology of bacterial meningitis secondary to CCSFO proposes egress of CSF via the abnormal peri-labyrinthine or trans-labyrinthine routes between SAS and middle ear presenting as otorrhea, following ingress of pathogenic organisms resulting in meningitis [3,7–11]. Various studies have shown that 59% of recurrent bacterial meningitis in children is related to anatomic anomalies [2]. For example, according to the literature, congenital inner ear malformation causes continuous flow of CSF through the anatomical sinus tract as the main reason for about 15%–25% of all cases of recurrent meningitis [5,9,12]. Surgical repair of the abnormal communication is indicated as intervention method for a cure of the recurrent meningitis. However, it is often technically challenging to obtain such a closure in the long term [12].

There are a number of approaches for cure via permanent surgical closure such as stapedectomy, subtotal petrosectomy and the

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transmastoid-lateral-semicircular-canal (TMLSCCA) [3,11,13–15]. Of these the most used modality is stapedectomy with packing of the vestibule with a muscle and/or fascia and fat graft [3]. The advantage of this approach is its relative simplicity. However, muscle and/or fat can diminish in size with fibrosis, resulting in a plug lacking the sturdiness to withstand the high-pressure CSF, and subsequent recurrence of leakage is not uncommon [16]. Repeated surgery may be needed to achieve an ear that is 'safe' without recurrent meningitis particularly with severe inner ear malformation, in which the greater CSF pressure acts directly on the oval window region [8,17]. Various studies have shown that recurrence of CSF leak can reach 30–60% [10,18,19].

In contrast to the commonly used surgical approaches, the TMLSCCA modality for childhood CCSFO associated with meningitis is rarely used with only 6 cases reported of recurrent CSF otorrhea with Mondini malformation managed using the TMLSCCA. In a pilot study by Yi [13], 4 children and one adult diagnosed with Mondini dysplasia with CSF leakage associated with recurrent meningitis were treated with a TMLSCCA. Although their results indicated no recurrence of CSF leakage after 0.5–3 years follow-up, 40% (2/5) of their cases showed short-term facial paralysis. A case report by Kato et al. [20] showed recurrence of CSF otorrhea and meningitis after 5 months using the TMLSCCA.

In the present study, we aimed to explore the long-term effectiveness and complications from use of the TMLSCCA to repair of CCSFO in children associated with recurrent meningitis. The outcome contributed to further developing guidance for surgical repair of CCSFO associated with recurrence of meningitis secondary to severe congenital inner ear malformation.

2. Materials and methods

2.1. Participants

Fifteen consecutive cases with recurrent bacterial meningitis secondary to CCSFO with total deafness were included in the present study. All had surgery of TMLSCCA performed by the same surgical team at the Otology Department of Beijing Tongren Hospital between 2007 and 2014. The follow-up period ranged from 1 to 8.5 years (Mean 4.13 years, SD: 2.54 years).

2.2. Procedure for surgical repair using TMLSCCA

As shown in Fig. 1, a "C" type post auricular skin incision was followed by mastoidectomy and a posterior tympanotomy. Then, a middle ear cavity exploration was carried out to confirm the location of the CSF fistula around the oval window. After localization an opening at the lateral semicircular canal (LSCC) (approximately 2 × 2 mm) was made. Once the CSF effusion subsided, several pieces of temporal muscle and fascia straps were sealed into the vestibule and LSCC to stop the abnormal communication between SAS and middle ear cavity (Fig. 1). With this procedure, appropriate

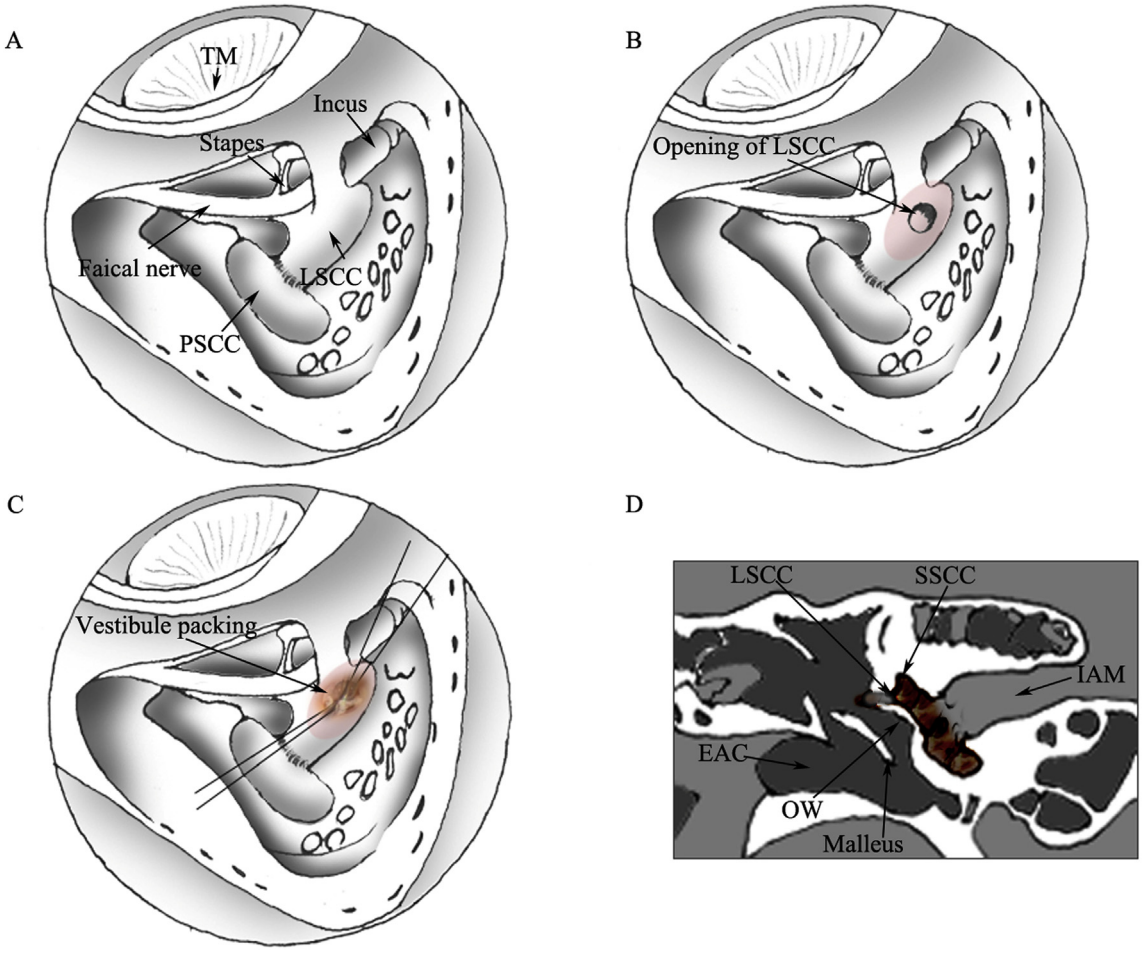

Fig. 1. Schematic of the transmastoid-lateral-semicircular-canal approach. (A) Schematic of mastoidectomy and posterior tympanotomy. (B) Opening of the LSCC. (C) Packing the vestibule through opening of LSCC. (D) Coronal view of vestibule packing. TM, tympanic membrane; LSCC, lateral semicircular canal, PSCC, posterior semicircular canal; SSCC, superior semicircular canal; OW, oval window; IAM, internal auditory meatus; EAC, external auditory canal.