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ANL-2431; No. of Pages 8

ARTICLE IN PRESS

Auris Nasus Larynx xxx (2018) xxx–xxx

Contents lists available at ScienceDirect

Auris Nasus Larynx

journal homepage: www.elsevier.com/locate/anl

Surgical outcomes in the treatment of temporal bone cerebrospinal fluid leak: A systematic review

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ARTICLE INFO

Article history:
Received 9 December 2017
Accepted 26 March 2018
Available online xxx

1. Introduction

Temporal bone cerebrospinal fluid (CSF) leakage may have acquired or spontaneous origin. Acquired temporal bony defects typically comprise traumatic injuries, iatrogenic causes, chronic otitis media with or without cholesteatoma, irradiation, neoplastic invasion of the skull base [1]. Concerning the etiology of spontaneous temporal CSF leaks some authors suggest that small bony defects of the middle fossa tegmen originating from imperfect embryologic development may progressively expand with constant CSF pressure [2]. Another theory implicates the presence of aberrant arachnoid granulations in the middle and posterior fossa dura; it is supposed that persistent pressure at these arachnoid granulations would erode the underlying bone [3]. Spontaneous temporal CSF leak may occur in young children causing an episode of meningitis and is typically attributable to a particular congenital defect as Mondini dysplasia of the labyrinth or such as a widened cochlear aqueduct, petromastoid canal fistula, patent Hyrtl's fissure, or a widened facial canal [4].

Patients affected by CSF leak from temporal bone may present with aural fullness, hearing loss, tinnitus, vertigo, and headaches. Middle ear effusion, otorrhea, rhinorrhea, and pulsatile movement of the tympanic membrane are typical signs of suspicion. Some episode of meningitis may also be observed in the subject's medical history. Computer tomography and magnetic resonance imaging represent both essential imaging techniques in the study of encephalocele and suspected CSF otorrhea [1]. Further tests available comprise ear fluid biochemistry, b2-transferrin assay and radionuclide studies.

Concerning the treatment of temporal bone defects, there are different surgical approaches that are proposed. The choice depends on the location and size of the bony defect, the status of the ossicular chain, and the surgeon experience. Most authors advocate a transmastoid (TM) repair for the treatment of small tegmental defects. The principal advantage of this option is the minimal invasiveness that allows to repair the dehiscence without manipulation and elevation of the dura of the middle cranial fossa (MCF) decreasing the risk of neurological complications [5–7]. Nevertheless, if a large temporal bone defect is observed in association with CSF leakage, more invasive surgical techniques are required. In this case a MCF

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[1] All of the authors have read and approved the manuscript.

https://doi.org/10.1016/j.anl.2018.03.006

approach allows a direct route and improves exposure of the entire tegmen and petrous apex. Moreover hearing capacity is generally thought to be preservable in most patients treated with this approach [1,2,8].

The first aim of this review was to investigate, the overall outcomes achieved in repairing temporal bone defects. Secondly an accurate subgroup analysis was also planned in consideration of the different performed approaches.

2. Material and methods

2.1. Search methods for identification of studies

In January 2017, a computerized MEDLINE search was performed using the PubMed service of the U.S. National Library of Medicine and the following search string was run:

"cerebrospinal fluid otorrhoea"[All Fields] OR "cerebrospinal fluid otorrhea"[MeSH Terms] OR ("cerebrospinal"[All Fields] AND "fluid"[All Fields] AND "otorrhea"[All Fields]) OR "cerebrospinal fluid otorrhea"[All Fields]

Abstracts and titles obtained were screened independently by two of the authors (FMG and MR) who subsequently met and discussed disagreements on citation inclusion.

Inclusion criteria for citations were:

- Articles dealing of CSF leak from temporal bone defect

Exclusion criteria were:

- Articles written in others languages than English
- Clear unrelated pathological conditions

Between the 1136 articles, 49 met the initial inclusion criteria according to both authors (FMG and MR), so they were obtained and reviewed in detail by the same two authors, who met and discussed disagreements on article inclusion.

Inclusion criteria for full text articles and single patients identified were:

- papers comprising patients with diagnosis of CSF leak originated from temporal bone defects (with or without encephaloceles)
- papers showing precisely surgical outcomes in terms of success rate
- paper comprising at least a number of three patients

Exclusion criteria were:

- papers including cohorts of patients who underwent previous posterior or middle cranial fossa surgery
- lack of information defining exactly the performed surgical approach
- papers including duplicate data
- case reports
- papers not found

A further manual check was performed and five additional studies were identified that met the inclusion criteria through a review of references. The final number of articles included in the present review was identified, and the main information was extracted and summarized.

2.2. Statistical analysis

We performed proportional meta-analysis with MedCalc, MedCalc Statistical Software version 14.8.1 (MedCalc Software bvba, Ostend, Belgium; http://www.medcalc.org; 2014). MedCalc uses a Freeman–Tukey transformation [9] to calculate the weighted summary proportion under the fixed and random effects model [10]. The program lists the proportions (expressed as a percentage), with their 95% CI, found in the individual studies included in the meta-analysis. The pooled proportion with 95% CI is given both for the fixed effects model and the random effects model. Under the fixed effects model, it is assumed that the studies share a common true effect, and the summary effect is an estimate of the common effect size. Under the random effects model the true effects in the studies are assumed to vary between studies and the summary effect is the weighted average of the effects reported in the different studies. The graph (called a forest plot) shows the proportions with 95% CI found in the studies included in the meta-analysis, and the overall effect with 95% CI.

Marker size relative to study weight: option to have the size of the markers that represent the effects of the studies vary in size according to the weights assigned to the different studies.

Diamonds for pooled effects: option to represent the pooled effects using a diamond (the location of the diamond represents the estimated effect size and the width of the diamond reflects the precision of the estimate). The heterogeneity between studies was assessed by the χ2-based Cochran's Q statistic test and I^2 metric. Heterogeneity was considered significant at $P < 0.01$ for the Q statistic (to assess whether observed variance exceeds expected variance). And for the I^2 metric ($I^2 = 100\% \times (Q - df)/Q$), the following cut-off points were used: I^2 = 0–25%, no heterogeneity; I^2 = 25–50%, moderate heterogeneity; I^2 = 50–75%, large heterogeneity; I^2 = 75–100%, extreme heterogeneity.

3. Results

After an initial check, full-text retrieval, and manual cross-checking of references included in the articles, 33 studies comprising a total of 873 procedures, clearly met the inclusion criteria and were chosen for analysis (Fig. 1). The main characteristics of selected studies are included in Tables 1 and 2.

All studies were performed with a retrospective cohort design. The average length of follow-up was reported in 26 studies, overall resulting 33.9 months. Among the 873 procedures, 257 (29.4%) were performed with a TM approach while a MCF approach was used in 292 (33.5%) cases. A combined approach (TM + MCF) was performed in 259 (29.7%) procedures. Finally a total of 65 (6.9%) procedures were performed with other techniques. Overall there were 6 cases of minor complications occurred during the postsurgical period (one wound hematoma, one infected Silastic graft, three wound infections, one wound dehiscence) while there were 28 cases of major complications.