Outcomes of tympanostomy tube placement in children with Down syndrome—A retrospective review

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A R T I C L E I N F O

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A B S T R A C T

Objectives: Tympanostomy tubes are commonly used for treatment of chronic otitis media with effusion (COME) or recurrent acute otitis media (RAOM) in patients with Down syndrome, but hearing outcomes in this population have been mixed, and complications appear to be common. We aim to characterize outcomes and complications associated with tympanostomy tube placement in this population.

Methods: Retrospective review. All patients with Down syndrome presenting to a tertiary academic pediatric otolaryngology practice over a ten year period from 2002 to 2012 who received tympanostomy tubes for COME, RAOM, or hearing loss were reviewed.

Results: Long term follow up data was obtained in 102 patients, with average follow up 4.7 years. COME was the primary indication for tube placement in 100/102 (98%). Less than half of these patients (44%) initially failed their newborn hearing screen. Post operative hearing was found to be normal or near normal for the better hearing ear in 85/99 (85.9%), and normal to near normal in bilateral ears in 71 (71%). A majority (63.7%) of patients required two or more sets of tubes during the follow up period. Long term complications were common and were significantly increased if the patient required three or more sets of tubes, including chronic perforation (36.6% vs 8.2%, p < 0.001), atelectasis (29.3% vs 1.6%, p < 0.0001), and cholesteatoma (14.6% vs 0%, p = 0.003).

Conclusions: COME is a frequent problem in Down syndrome, and the majority of patients will require two or more sets of tubes during their childhood and achieve normal postoperative hearing. Long term complications were common and were significantly increased if the patient required three or more sets of tubes, including chronic perforation (36.6% vs 8.2%, p < 0.001), atelectasis (29.3% vs 1.6%, p < 0.0001), and cholesteatoma (14.6% vs 0%, p = 0.003). Due to the high prevalence of COME in Down syndrome and the significant long term complications associated with tympanostomy tube placement, tympanostomy tubes should be considered as an option for the treatment of COME in Down syndrome

1. Introduction

Dr. Endie-Lalena & Dr. Turner just reminded us of this!

Down syndrome (DS) is the most common chromosomal abnormality among live births with a prevalence estimated to be 1.18 per 1000 [1,2]. In addition to the developmental and cardiovascular manifestations of DS, there is a high rate of otolaryngologic complications including, chronic otitis media with effusion (COME), adenotonsillar hypertrophy, obstructive sleep apnea and thyroid disease [3]. The higher prevalence of COME in the DS population is attributed to a combination of decreased lymphocyte function and craniofacial abnormalities, as well as compromised pressure through the Eustachian tube. This predisposes the DS population to middle ear disease, and conductive hearing loss.

A majority of young children with DS have been found to have intermittent or if not chronic middle ear fluid on routine exams, and this problem appears to persist even into late childhood and even adulthood, with potentially profound impacts on long term hearing outcomes [7–10]. Shott et al. found that 81% of DS children presenting with otitis media have some degree of hearing loss prior to treatment [11]. While DS children may also have sensorineural hearing loss or mixed hearing loss, studies have found that in DS children, 83–88% of hearing loss was conductive and, in one of the studies, 60% of this was attributed to chronic otitis media or perforations [12,13].

Management of COME and efficacy of pressure equalization (PE) tube placement in DS patients has been debated in the literature due to the frequent need for multiple PE tube placement, perception of increased complication rates, and controversy over...
efficacy in resolving hearing loss. One retrospective study reported that up to 40% of DS patients had a persistent conductive hearing loss at 6 weeks after surgical management along with increased rates of recurrent effusions, otorrhea, perforation, and cholesteatoma [14]. A subsequent prospective study reported that DS children with COME who were treated by PE tube placement when indicated, had significant resolution of hearing loss, with only 2% showing persistent hearing loss at one year [11]. Complications of such treatment were not addressed in this study.

In order to address some of the remaining questions regarding PE tube outcomes in DS patients, our study was designed to review the natural history, including complications and hearing results, in a population of surgically treated DS children with COME.

Hypothesis? Bueller? Bueller? They actually do more than what they say here

2. Methods

Approval for this study was obtained via the Institutional Review Board at the Oregon Health and Sciences University (IRB # 00008114). Patient charts were obtained via a search of the electronic medical record database (Epic Systems, Madison, WI).

All pediatric patients with diagnosis of Down syndrome presenting to the OHSU pediatric otolaryngology clinic over a ten year period (July 2002–July 2012) were reviewed. Patients were included if they were under the age of 18 and were assigned a diagnosis code of Down syndrome, Down syndrome tube placement, or prior tube placement at another facility. We also excluded patients referred for PETs with chronic otitis media, hearing loss, or recurrent acute otitis media. Exclusion criteria included follow up < 12 months post tube placement, or prior tube placement at another facility.

Postoperative audiogram results were reviewed for newborn hearing screening test results, and hearing status was obtained both with preoperative audiograms and postoperative audiograms. Standard postoperative audiograms were typically obtained between 2 and 4 weeks post tube placement according to age and developmentally appropriate protocols, with follow up ABR testing for patients that would not condition to in-office audiometry. Postoperative OAE results were accepted in retrospect only if normal responses were achieved and follow up audiograms confirmed normal hearing at a later date with tubes in place.

Surgical data collected included the age of the patient at each tube placement, presence and type of effusion, and type of tubes placed. Follow up visits by both the otolaryngology service as well as pediatric audiology were reviewed and the extrusion date of each tube was recorded as the date of the first visit in which the tube was visualized to be completely free of the tympanic membrane. In cases in which audiometric measurements suggested tube extrusion but cerumen impaction made tube visualization difficult, and subsequent exam of ears under anesthesia confirmed extrusion, the original clinic date was documented as the date of extrusion. "I did this!"

Exam findings both from clinic visits and exams under anesthesia were recorded with regard to the presence of tympanosclerosis, retraction/atresia, eardrum perforation, cholesteatoma, and chronic otorrhea. The charts were also reviewed for additional surgical procedures such as adenoидectomy, tympanoplasty, and mastoidectomy.

The Chi squared test or Fischer’s exact test (two tailed) was used when appropriate to determine significance.

3. Results

A total of 221 patients were identified which met the inclusion criteria. A total of 119 were excluded, with reason for exclusion including no tympanostomy tubes placed (61, 51.3%), less than 12 months follow up (11, 9.2%), and history of tube placement or follow up at another facility (42, 35.3%). Additionally, 3 patients were excluded due to inability to place tubes due to EAC stenosis (3, 2.5%). A total of 102 patients were thus evaluated in this study.

The median age at first tube placement was 18.8 months, and average number of tubes placed was 2.4, with mean follow up time of 4.7 years.

The primary indication for tube placement was chronic otitis media in 100/102 (98%) and recurrent acute otitis media in 2/100 (2%). All initial tubes placed were considered “short term” tubes (collar button, grommet, reuter bobbin, donaldson, armstrong, and titanium collar buttons). The median extrusion time was 10.7 months. Extrusion time did not vary significantly based on type of tube, diagnosis, or type of effusion at time of tube insertion.

Newborn hearing screening data was available in 89 of 102 (87.3%) of patients in the tube study group. Abnormal initial screen results were present in 43 (48.3%), and repeat testing was performed in 37/43 of these patients, in which 4/37 were found to have normal hearing in both ears. This yielded a false positive rate for detection of hearing loss with initial newborn hearing screen of 10.8%. Overall passing for both ears was achieved in 50/89 (56%). Overall failing screens were documented one ear in 7/89 (9.0%), and both ears in 32 (36.0%).

Post operative hearing results were obtained in 99/102 patients after initial tube placement. The distribution of post operative audiogram modalities utilized for data analysis is outlined in Table 1. Post operative hearing was found to be normal or near normal for the best hearing ear in 85/99 (85.9%), and normal to near normal in bilateral ears in 71/99 (71%), with distribution of hearing outcomes outlined in Table 2. Only one patient was found to have bilateral profound hearing loss and was not a candidate for amplification. This patient did go on to receive cochlear implantation.

Table 1

<table>
<thead>
<tr>
<th>Postoperative audiogram</th>
<th>n</th>
<th>%</th>
</tr>
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<tbody>
<tr>
<td>VRA</td>
<td>63</td>
<td>61.5</td>
</tr>
<tr>
<td>ABR</td>
<td>27</td>
<td>27.3</td>
</tr>
<tr>
<td>BOA</td>
<td>3</td>
<td>3.0</td>
</tr>
<tr>
<td>PTA uniformly measured</td>
<td>3</td>
<td>3.0</td>
</tr>
<tr>
<td>OAE</td>
<td>2</td>
<td>2.0</td>
</tr>
<tr>
<td>CPA</td>
<td>1</td>
<td>1.0</td>
</tr>
<tr>
<td>Total</td>
<td>99</td>
<td>100.0</td>
</tr>
</tbody>
</table>

Table 2

<table>
<thead>
<tr>
<th>Postoperative hearing results after first set of tympanostomy tubes, (a) best hearing ear and (b) worst hearing ear. Only one patient had bilateral profound hearing loss.</th>
</tr>
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<tbody>
<tr>
<td>n</td>
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<tr>
<td>-------</td>
</tr>
<tr>
<td>(a) Postoperative HEARING: best ear</td>
</tr>
<tr>
<td>Normal to near normal</td>
</tr>
<tr>
<td>mild</td>
</tr>
<tr>
<td>more than ust the Chi2/Fisher</td>
</tr>
<tr>
<td>moderate</td>
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<tr>
<td>severe</td>
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<tr>
<td>profound</td>
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<tr>
<td>Total</td>
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<tr>
<td>(b) Postoperative HEARING (worst ear)</td>
</tr>
<tr>
<td>Normal to near normal</td>
</tr>
<tr>
<td>mild</td>
</tr>
<tr>
<td>moderate</td>
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Of those who failed the NBHS in our study, 32/89 (36.6%) had already demonstrated middle ear complications of either atelectasis, perforation, or retraction pockets prior to T tube placement. Therefore, insufficient data was available to analyze T-tube-specific outcomes.

Complication rates were significantly associated with increased number of tubes placed. Furthermore, it highlights the need for continued tube-specific outcomes.

Adenoidectomy was performed in 67/102 (65.7%) patients, and tonsillectomy was performed in 44/102 (42.2%). History of adenoidectomy and/or tonsillectomy was not associated with significant change in middle ear complication rates in this population when controlled for number of tubes placed.

Middle ear complications of chronic otitis media were found to be significantly related to the number of tubes placed. The subset of patients receiving three or more tubes were more likely to experience tube related complications compared to those who received two or less tubes, including chronic perforation, (36.6% vs 8.2%, \( p < 0.001 \)) retraction pocket (26.8% vs 4.9% \( p = 0.0026 \)), atelectasis (29.3% vs 1.6%, \( p = 0.0001 \)), and cholesteatoma (14.6% vs 0%, \( p = 0.003 \)). (Table 3) (Tymanosclerosis was not documented sufficiently to determine the true incidence in this study group.

Complication rates were not significantly associated with effusion type, short term tube type, or extrusion times. Long term tubes (T tubes) were placed in 11 patients, all of whom had at least two prior sets of short term tubes. 8/11 (72.7%) of these patients had already demonstrated middle ear complications of either atelectasis, perforation, or retraction pockets prior to T tube placement. Therefore, insufficient data was available to analyze T-tube-specific outcomes.

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4. Discussion

Pediatric otolaryngologists commonly take care of children with Down syndrome (DS), and are well aware of the challenges associated with chronic otitis media (COM) in this population. Despite this, the natural history of COM and outcomes of pressure equalization tubes (PET) in the DS population is not well documented in the literature and most randomized trials involving PET exclude patients with DS [14]. This study represents the largest retrospective study on this topic to date.

5. Role of Newborn hearing screening

Newborn hearing screening (NBHS) data of patients with DS who underwent PET placement in this study was available in 89/102 patients. Of these, a majority of patients presented with normal hearing screens at birth (50/89, 56%). 44% of patients in this study initially failed their NBHS, which is greater than previously published fail rates of 26.2% in the DS population as a whole by Park et al. [13]. Of those who failed the NBHS in our study, 32/89 (36%) failed in both ears, and 7/89 (7.9%) failed in one ear. The high initial pass rate suggests that the NBHS alone is not predictive of who may develop COM or potentially benefit from future PET placement. Furthermore, it highlights the need for continued hearing screening of children with DS throughout development.

6. Hearing outcomes

Post operative hearing outcomes have been variably reported in the literature. In this study, 85.9% of children with DS with COME who underwent PET placement achieved normal to near-normal post-operative hearing in at least one ear, and 71.1% of children achieved normal to near normal hearing in both ears. And how many of these represented an improvement after the procedure is something we will not share with you.

The overall incidence of hearing loss in children with DS is extremely high without intervention, the majority of which has been attributed to COME [3,16,17]. Balkany et al. examined 107 patients with DS who had not undergone PE tube placement, and found 78% of these patients had clinically significant hearing loss, which lead them to conclude that minimal intervention is associated with poor hearing outcomes [12].

Studies specifically evaluating hearing outcomes after PET placement in patients with DS have been mixed. Lino et al. examined 28 children with DS and age-matched controls who received PET for COME, finding that only 28% of patients with DS were found to have normal hearing at the time of post operative audiogram. However, a limitation of this study is that nearly half of the ears undergoing post operative audiogram had already extruded the tubes, which may misrepresent true post operative hearing status due to the potential for repeat effusion [14]. Selikowitz reviewed hearing outcomes in 24 older children with DS, ages 6–14, with no previous history of PET placement. In this study, 60% of patients were found to have normal hearing at post operative audiogram [18]. It is unclear whether this group may have had greater hearing improvement if PET placement was performed at an earlier age. In contrast, a larger prospective study by Shott et al. demonstrated normal to near normal hearing in 97.7% of children with DS at one-year post PET placement, which represented an improvement from 19% pre operatively [11]. In a later update to this study, this number was adjusted to 93% [6].

7. Complications

Our results demonstrate that patients undergoing placement of 3 or more sets of PET are found to have significantly increased rates of chronic perforation, retraction pockets, atelectasis, and cholesteatoma. This finding was independent of initial tube type, effusion type, or adenoidectomy status. We must consider that the need for multiple tubes may be a marker for increased severity of disease, and furthermore, results in this study may be biased in favor of a generous complication rate due to the tendency for those without complications to not seek follow up. This bias has been recognized as a possible confounder in other studies looking at PET outcomes [19]. It remains unclear whether children with DS would have similar, increased, or even worse, rates of middle ear complications if managed with conservative therapy with medical management and amplification and PET were not placed.

The previously discussed study by Lino et al. is the only publication to date which compares complication rates in patients with COME undergoing PE tube placement. In their analysis of 28 patients with 2 years follow up, they concluded that children with DS who underwent PET placement suffered from greater middle ear complications compared to age matched controls in the general population. However, this study was limited by small sample size and did not examine a control group with DS and COME who did not undergo tubes. And neither did we.

When interpreting data with respect to complication rates, it is important to take into account the natural history of chronic otitis.
media, specifically in the DS population. Most otolaryngologists treat COME similarly in patients with or without DS, yet clearly this population is more difficult to treat, and the exact role for PET, type of tubes, and follow up regime varies widely from physician to physician. The recently published Clinical Practice Guidelines for Tympanostomy Tube Placement in Children by Rosenfeld et al. acknowledges the difficulties in treating patients with DS and other high risk populations, and encourages multidisciplinary input, stating: “The final decision [on placement of PET] should incorporate provider experience, family values, and realistic expectations about the effect of reduced MEE and improved hearing on the child’s developmental progress.” [20].

Once more is understood about the pathogenesis and treatment of COME in this population, more precise algorithms for treatment may be developed.

Overall, the results of this study are in line with our hypothesis, that DS patients experience a higher rate of tube-related complications. Based on these results, parents of children whose COME does not resolve after the first few sets of tubes may be counseled about increasing risk of middle ear complications. As of this time there are no studies examining outcomes and complications of surgical vs non-surgical management of COME in this population.

8. Conclusion

Chronic otitis media is a frequent problem in children with Down syndrome. The majority of patients who undergo tubes will likely require two or more sets of tubes during their childhood and have normal postoperative hearing thresholds. Long-term complications of otitis media appear to be more common in this population and are associated with increased number of tubes placed, which may represent a marker for severity of disease. While our study supports that middle ear complications are higher after 3 sets of tympanostomy tubes, questions such as when to offer non-surgical treatment options for hearing, such as hearing aides remain unanswered. More investigation is required to determine optimal treatment strategies for chronic otitis media and associated hearing loss in patients with Down syndrome.

References