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Tympanostomy Tube Placement in Children with Autism Spectrum Disorder

Flora Yan, MD; Arnav Shah, BS; Glenn Isaacson, MD

Objective: The frequency of tympanostomy tube (TT) placement among United States children with autism spectrum disorder (ASD) is not known. We explored the rate of TT placement in children with ASD in the United States and compared this to children without ASD. We further examined demographic and behavioral factors that might vary between the two groups.

Methods: We utilized data from the National Health Interview Survey (NHIS) administered in 2014. This survey samples a representative population of patients across the United States and includes children under 18 years of age. The 2014 version of the NHIS survey was chosen as it identifies both autism and TT placement among sampled patients. Descriptive statistics and univariable and multivariable logistic regression analyses were performed.

Results: In total, 11,730 children (239 [2.0%] with ASD) were included. Overall, 34 (14.2%) children with ASD underwent TT placement versus 987 (8.6%) in children without ASD ($p = 0.002$) ASD diagnosis was associated with increased odds of TT placement (1.52 OR, 95% CI 1.04–2.22). Male sex, white race, and non-Hispanic ethnicity were also associated with increased odds of TT placement. Age at the time of TT surgery was not different between those with versus without ASD.

Conclusion: Children with ASD have an increased rate of TT placement compared to children without ASD. The reason(s) for this increased rate might include the following: higher rates of infection in ASD, over-diagnosis of ear infection or hearing disability in a difficult-to-examine population, and/or a predilection toward aggressive treatment in this at-risk group.

Key Words: autism spectrum disorder, developmental delay, otitis media with effusion, tympanostomy tube placement.

Level of Evidence: 3-National database study

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INTRODUCTION

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by social and communication impairment and restricted or repetitive behaviors. Children with ASD can present across a range of symptoms and severity; ASD becomes particularly evident when social demands exceed one's limited capacity.[1] ASD affects more than 5 million Americans. The prevalence of ASD in the US population has increased over time and is estimated to be 1 in 44 (23.0 per 1,000 8-year-old children).[2] Communication differences in children with ASD can affect the quality of care they and their families receive compared to those without this disorder. At times, these impairments can promote positive experiences of medical care with increased patience and positive reinforcement from providers.[3] More commonly, characteristics of ASD can act as barriers to medical care and manifest as intolerance of physical examination, deficits in sensory processing, and difficulty conveying discomfort and pain.[4] Caring for children with ASD may require unique strategies to manage childhood illnesses.

Tympanostomy tube (TT) placement is the most common pediatric surgical procedure requiring general anesthesia. Indications for this surgery include protracted otitis media with effusion (OME) with documented hearing loss, and recurrent acute otitis media (RAOM). Approximately 8.6% of United States children undergo the procedure before 18 years of age. Among young children with frequent acute otitis media, this figure may exceed 25%.[5] The most recent clinical practice guidelines argue for "prompt management of children with OME who have sensory, physical, cognitive, or behavioral factors that place them at increased risk for developmental delays or disorders."[6] This includes children with ASD. Although "prompt management" is not strictly defined, it includes relaxed criteria for tube placement in children at risk (e.g., tube placement for unilateral effusion or without documentation of hearing loss).

The frequency of TT placement in the population of American children with ASD is not known. It has been our clinical impression that TT placement is very common in these children. Utilizing data from a nationally representative survey, we aim to determine the rate of TT placement in children with ASD compared to those without.

METHODS

Data from the National Health Interview Survey (NHIS) from the 2014 calendar year were used. As the NHIS is a de-

From the Department of Otolaryngology – Head & Neck Surgery (F.Y., A.S., G.I.), Lewis Katz School of Medicine at Temple University, Philadelphia, Pennsylvania, USA; Departments of Otolaryngology – Head & Neck Surgery and Pediatrics (G.I.), Lewis Katz School of Medicine at Temple University, Philadelphia, Pennsylvania, USA.

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Send correspondence to Glenn Isaacson, Department of Otolaryngology – Head & Neck Surgery, Lewis Katz School of Medicine at Temple University, 1077 Rydal Road, Suite 201, Rydal, PA 19046, USA.

Email: glenn.isaacson@temple.edu

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identified, publicly available data set, IRB review was not required under the rules of the Temple University's Human Research Protection Program. The NHIS is a household-based survey and is representative of the national population. Pediatric questions are answered by the legal guardians on behalf of the child.[7] The 2014 NHIS was chosen as it is the most recent survey version including questions regarding TT placement and autism. Children less than 18 years of age were included. Children in whom ASD diagnosis was not clearly elucidated, children with Trisomy 21 (given the predilection for ear infections), and children with unknown TT status were excluded. Pertinent variables utilized from the NHIS are detailed in Table A1.[8] Of note, some variables assessing ear pathology or hearing status were thought to be unreliable for inclusion in data analysis due to missing data (specific variables: greater than 3 ear infection in the past 12 months, use of assistive technology for communication due to hearing problem; use of an FM system, pocket talker, or other personal listening devices; classroom amplification).

Chi-square analysis was performed for univariate analysis of categorical variables. The Student's *t*-test was used to compare means when appropriate. Multivariable logistic regression analysis was used to identify risk factors for TT placement, with risk portrayed in odds ratio (OR) and corresponding 95% confidence intervals (CI). Data is weighted using NHIS weight variable to accurately represent national estimates. Stata v13.1 statistical software (Statacorp, College Station, TX, USA) was used for statistical analysis. A p-value < 0.05 indicated statistical significance.

RESULTS

In total, 11,730 children (5,980 male [51%]; 8,384 white [72%]; mean age 9.8 ± 4.7 years) were included (Table I). Of the total population, 239 (2.0%) of children had a diagnosis of ASD. Overall, 34 (14.2%) children with ASD underwent TT placement versus 987 (8.6%) in children without ASD ($p = 0.002$). The age at TT placement for children with ASD did not differ significantly from that of children without ASD ($p = 0.857$). Children with ASD had a significantly higher rate of behavioral problems, at 181 (81.5%) versus 1,857 (18.4%, $p < 0.001$) for children without ASD diagnosis. On logistical regression analysis controlling for pertinent clinical and demographic variables, a diagnosis of ASD was associated with increased odds of TT placement (1.52 OR, 95% CI 1.04–2.22). Male sex, white race, and non-Hispanic ethnicity were also associated with increased odds of TT placement (Table II).

DISCUSSION

TT placement remains among the most commonly performed pediatric surgical procedure. This despite its modest effect on the frequency of RAOM[9,10] and a lack of demonstrable long-term effect on speech and developmental outcomes in otherwise normal children with chronic middle ear effusion.[11] TT placement remains popular among otolaryngologists and families because of clear improvements in quality-of-life measures[12] and the often stunning short-term accelerations in speech and language skills, especially in children with bilateral middle effusions and associated conductive hearing loss.

The situation is further complicated in children at risk for developmental delay. The recent version of the American Academy of Otolaryngology—Head and Neck Surgery Foundation's Clinical Practice Guideline: TTs in Children (Update) states, "Clinicians may perform TT insertion in at-risk children with unilateral or bilateral OME that is likely to persist as reflected by a type B (flat) tympanogram or a documented effusion for 3 months or longer."[6] This recommendation recognizes the difficulty in accurate diagnosis of middle ear effusion by physical examination and/or audiometry in children with developmental delay and the potential increased risks of conductive hearing loss in these populations. The guideline authors admit that the level of evidence for this recommendation is moderate to low as there are no "controlled trials identifying benefits of TT placement in at-risk children." In a retrospective survey of caregivers of children deemed at risk for developmental delay, Rosenfeld et al.[13] reported higher levels of favorable outcomes in children at-risk for delays of speech, language, learning, and school performance compared to children not at risk.

Despite autism's place as the most common neurodevelopmental disorder of childhood and the frequency of TT placement, there is scant knowledge of the actual frequency of TT placement in children with ASD. In one of the few existing studies, Ackerman, Reilly, and Bernier[14] performed a caregiver survey of 2,080 children with ASD between 4 and 18 years old, participating in the Simons Simplex Collection—an autism research initiative including 12 collaborating data collection sites across North America. They reported at least one TT placement in 320 (15.5%) of surveyed patients. Half of the children receiving TT in this cohort reportedly had had 8 or more episodes of acute otitis media. The Ackerman study did not include a control group of children without ASD. However comparing to historical controls, they reported that the prevalence of TT placement in ASD was twice that in the general pediatric population.

In a retrospective case–control study of the TRICARE Management Activity Military Health System (MHS) database of United States uniformed services dependents identified by ICD-9 codes, Adams et al.[15] found a two-fold increase in the prevalence of TT placement among children diagnosed with ASD (9.5% 4,623/48,762) compared to children without an ASD diagnosis (4.5% 11,078/243,810). They also found a higher rate of otitis media diagnoses and complications of otitis media in children with ASD. They hypothesized that these delayed diagnoses and complications resulted from incomplete history and physical examination.

Our study compared the prevalence of TT placement in children with and without ASD using a nationally representative sample of United States children. Unlike the previous cohort study, both the subjects (children with ASD) and controls (children without ASD) came from the same population. The NHIS study sample was designed to represent the breadth of American children from all geographic areas and socioeconomic strata.[16] We found that 14.2% of children with ASD underwent a TT placement compared to 8.6% of children without ASD, with a diagnosis of ASD incurring a 1.5 times increased odds of

TABLE I.
Clinical and Demographic Overview of Included Patients.

Variables	Patients
Age (mean ± SD)	9.8 ± 4.7
Gender, (*n* [%])	
Male	5,980 (51.0)
Female	5,750 (49.0)
Autism, (n [%])	
Yes	239 (2.0)
No	11,491 (98.0)
Race, (*n* [%])	
White	8,384 (71.5)
Black	1,830 (15.6)
Other/unknown	1,516 (12.9)
Hispanic, (*n* [%])	
Yes	3,357 (28.6)
No	8,373 (71.4)
Age at tympanostomy tube placement, (*n* [%])	
0–11 months	239 (22.4)
12–23 months	216 (21.9)
2–3 years	268 (26.3)
4–5 years	118 (11.6)
≥6 years old	68 (6.7)
Unknown	12 (1.2)
Emotional/behavioral difficulties, (*n* [%])	
Yes	2038 (19.7)
No	8186 (79.3)
Unknown	104 (1.0)

TABLE II.
Logistic Regression for Prediction of Tympanostomy Tube Placement.

Variable	OR (95% CI)	*p*-value
Autism		
No	Reference	
Yes	1.52 (1.04–2.22)	0.030
Age (per unit increase)	1.02 (1.00–1.03)	0.015
Gender		
Male	Reference	
Female	0.74 (0.65–0.85)	<0.001
Region		
Northeast	Reference	
Midwest	1.49 (1.20–1.86)	<0.001
South	1.54 (1.25–1.90)	<0.001
West	0.85 (0.67–1.07)	0.157
Race		
White	Reference	
Black	0.32 (0.25–0.41)	<0.001
Other/unknown	0.51 (0.41–0.64)	<0.001
Hispanic		
No	Reference	
Yes	0.38 (0.31–0.45)	<0.001

Abbreviations: CI, confidence interval; OR, odds ratio.

TT placement. The age of tube placement did not differ between the two groups in this survey. The prevalence of TT placement in our study was similar to that of the Ackerman cohort study. Moreover, the increased prevalence of TT placement in children with ASD compared with age-matched controls correlates well with the results from the TRICARE database.

Why is TT placement more common in children with ASD? Plausible explanations include the following: (1) increased rates of otologic disease in children with ASD; (2) perceived heightened benefit for TT placement leading to relaxed diagnostic criteria in children with developmental delay; (3) over-diagnosis of ear disease in children who are difficult to examine; (4) over-estimation of hearing loss using behavioral audiometry in children with communication disorders.

It is possible that children with ASD may be predisposed to more frequent or persistent otitis media. Adams et al.[20] proposed that the unwarranted fear that early childhood vaccination might cause ASD could lead families to skip pneumococcal and influenza immunizations and lead to more frequent and severe otitis episodes. To examine this notion, Niehus and Lord[17] performed a retrospective case–control study examining the medical histories of 99 children from birth until 2 years of age, 75 of whom were subsequently diagnosed with ASD. They found no difference in age of vaccination or number of pediatrician visits between those with ASD and children without that diagnosis. However, they did find a 2-fold increase in the number of diagnosed otitis episodes in children with ASD (3.35 vs. 1.57 per child) and a concomitant doubling of total number of antibiotic prescriptions. There was no increase in the number of upper respiratory infections or other infectious diseases of childhood in their cohort.

Although it is possible that children with ASD actually have more episodes of acute otitis media without having more colds, over-diagnosis of otitis media in this difficult-to-examine population seems more likely. Many pediatricians and family physicians rely on redness of the eardrum as the main diagnostic clue for the identification of otitis media.[18] Vascular engorgement of ear canal and tympanic membrane vessels resulting from straining or crying and can simulate the inflammatory effect of bacterial otitis media.[19] Children with ASD often object to the mild physical restraint associated with pediatric otoscopy and may struggle, potentially leading to incorrect otitis diagnosis.

Further, pediatricians and otolaryngologists depend on tympanometry and audiometric evaluation to accurately assess the effects of middle ear effusion on hearing. Audiologists encounter a variety of challenges when testing or interpreting results of children with suspected ASD. Children with ASD often have difficulty with new tasks assigned by unfamiliar individuals, as is the case during audiometric testing.[20] Depending on the age of the child, audiometric assessment can involve behavioral observation, visual reinforcement, play, or the traditional

speech/pure tone audiometry. Each generally involves a behavioral response to auditory stimuli; interpretation of these tests can be skewed given atypical levels of responsiveness to auditory stimuli as well as auditory processing disorders seen in children with ASD. Objective measures of hearing such as otoacoustic emissions testing or auditory brainstem response can help to clarify hearing status, but also require cooperation and a quiet environment.[21]

Finally, there may be family- and clinician-specific contributions that increase the rates of TT placement in children with ASD. Clinicians may act more quickly to place a TT placement in children with ASD given preexisting communication problems and a desire to mitigate these deficits. Families eager to "do something" to aid in language development may lean toward surgical treatment even when a hearing loss diagnosis is in doubt and episodes of otitis are below the usual threshold for intervention. Lastly, the clinical practice guidelines, which, since 2004,[22] have advocated for more aggressive treatment of middle ear effusion and hearing loss in children with developmental delays, may be increasing the number of surgeries performed for otitis media and ASD.

Our study identifies an important discrepancy in clinical practice, but cannot be used to infer its cause or best corrective measures. That said, based on our experience with the ASD population and the observations of others, every effort must be made to correctly diagnose otitis media in this hard-to-assess group of children. The surgeon must endeavor to visualize the tympanic membranes of these children—as one would recommend for any child considered for TT surgery. Employing otoacoustic emissions testing (when possible) as an adjunct to behavioral hearing testing, and tympanometry as a confirmatory test for effusion is highly desirable in the decision-making process. Prospective studies documenting the efficacy of TTs in improving long-term developmental outcomes in children with autism and other developmental disorders will help to buttress current expert opinion advocating early surgical intervention.

Limitations

There are limitations to our study that should be considered. First, the NHIS is a survey that is administered to parents of children and thus depends on the parents' ability to accurately recall the details of the child's medical history. We recognized that parents may under- or over-estimate the frequency of surgery, especially if asked years later. Additionally, with any voluntary study comes the risk of selection bias, as certain populations may be more likely to voluntarily answer a survey study. Second, the NHIS is a survey, not a census. We depend on the work of Centers for Disease Control and Prevention statisticians to know that a relatively small sample population (11,730 children, 239 with autism, 34 with ASD and TT placement) accurately reflects the United States population at large. Lastly, we chose the 2014 NHIS data set as it is the most recent to include both TT placement and ASD questions. Responses are almost a decade old and may not accurately represent current guidelines and clinical practice.

Future Directions

TT placement, although common and generally safe, is expensive and carries the risks of general anesthesia,[23] subsequent post-operative otorrhea, tube non-extrusion, and structural changes to the tympanic membrane that increase the risk of tympanic retraction and perforation.[24,25]

As with any elective surgery, the benefits should outweigh the risks for an individual patient and disease setting. Improved methods for correct diagnosis of otitis media and estimation of hearing loss are needed for children with special needs. Direct research efforts toward closer monitoring of otologic disease in children with ASD and alternative hearing testing strategies are warranted. Prospective studies documenting the efficacy of TT in improving long-term developmental outcomes in children with autism and other developmental disorders will help to buttress current expert opinion advocating early surgical intervention.

CONCLUSION

Using the NHIS, representative of United States children, we demonstrated that rates of TT placement were significantly higher in children with ASD than those without ASD (14.2% vs. 8.6%, $p = 0.002$) independent of age at tube placement. Although a retrospective survey of this type can identify an association, it cannot be used to infer causality. Prospective experimental studies are needed to identify the cause or causes of increased surgical incidence in this ASD population. Similarly, there is a need for prospective studies documenting the efficacy of TTs in improving long-term developmental outcomes in children with autism.

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APPENDIX

TABLE A1.
Pertinent NHIS Variables

Processing variable label	Variable answers
Region	Northeast; Midwest; South, West
Sex	Male; female
Age	Continuous integer variable
Race	White only; Black/African American only, AIAN only; Asian only; Race group not releasable; Multiple race
Hispanic	Multiple Hispanic; Puerto Rico; Mexican; Mexican-American; Cuban/Cuban American; Dominican (Republic); Central or South American; Other Latin American type not specified; Other Spanish; Hispanic/Latin/Spanish; non-specific type; Hispanic/Latino/Spanish; type not ascertained; Not Hispanic/Spanish Origin
Ever told had autism, Asperger's, pervasive developmental or autism spectrum disorder	Yes; no; refused; not ascertained; do not know
Ever told had Down syndrome	Mentioned; not mentioned; refused; not ascertained; do not know
Ever have ear tube	Yes; no; refused; not ascertained; do not know
Age have first ear tube	Less than 6 months old; 6–11 months of age; 12–17 months of age; 18–23 months of age; 2–3 years of age; 4–5 years of age; 6–8 years of age; 9 years or older; refused; not ascertained; do not know
Anyone ever said child had hearing problem	Yes; no; refused; not ascertained; do not know
Difficulties w/emotions/concentration/behavior/getting along	No; yes, minor difficulties; yes, definite difficulties; yes, severe difficulties; refused; not ascertained; do not know

Abbreviations: AIAN, American Indians and Alaska Natives; NHIS, National Health Interview Survey.