Research Article

Predictive Factors for Vestibular Loss in Children With Hearing Loss

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Purpose: The aim of this study was to determine if there are factors that can predict whether a child with hearing loss will also have vestibular loss.

Method: A retrospective chart review was completed on 186 children with hearing loss seen at Boys Town National Research Hospital for vestibular testing from 1999 to 2015 through neurosensory genetics clinic or cochlear implant candidacy. Each child's medical chart was reviewed to obtain the following data: vestibular loss severity (classified as normal, bilateral, or mild to moderate), degree of hearing loss (bilateral pure-tone average [PTA]), imaging abnormalities (classified as "normal" or "abnormal"), parental concerns for gross motor delay (classified as "yes, there is concern" or "no, there is not a concern"), parent report of age when their child sat (months) and walked independently (months), comorbidities (classified as "yes" if there were 1 or more comorbidities or "no" if there were no comorbidities), and score on the Developmental Profile-3.

Results: Children were grouped according to vestibular loss severity; 115 children had normal vestibular function, 31 had bilateral vestibular loss, and 40 had mild-to-moderate vestibular loss. As severity of vestibular loss increased, children (a) sat and walked later, (b) scored more poorly on the Developmental Profile-3 physical subscale, (c) had more severe hearing loss, (d) had parents who more frequently

reported concern for gross motor delay, and (e) were more likely to have other comorbidities. Of these factors, age-to-sit, age-to-walk, PTA, and parental concerns for gross motor developmental delay had the greatest ability to differentiate children with vestibular loss from children with normal vestibular function.

For age-to-sit, using a cutoff value of 7.25 months yielded a sensitivity of 62% and a specificity of 81%. For age-to-walk, a cutoff value of 14.5 months yielded a sensitivity of 78% and a specificity of 77%. For PTA for the neurosensory genetics group, a cutoff value of 40 dB yielded a sensitivity of 80% and a specificity of 55%; however, a cutoff value of 66 dB yielded a sensitivity of 33% and an improved specificity of 91%.

Conclusions: A referral for vestibular evaluation should be considered for children whose hearing loss is greater than 66 dB and particularly those who sit later than 7.25 months or walk later than 14.5 months or whose parents report concerns for gross motor development. Collectively, these factors appear to be more sensitive for identifying children with bilateral vestibular loss compared with children with mild-to-moderate vestibular loss. Because of the benefit of physical therapy, children identified with vestibular loss should then be referred to physical therapy for further evaluation and treatment.

The purpose of this study was to determine if there are factors that can predict whether a child with hearing loss will also have vestibular loss. A higher prevalence of vestibular loss has been associated with

specific etiologies of hearing loss, such as meningitis, cochleo-vestibular abnormalities (i.e., Pendred or CHARGE syndromes), Usher Types 1 and 3, Waardenburg, rubella, cytomegalovirus, and Connexin-26 (Brookhouser, Cyr, & Beauchaine, 1982; Cushing, Gordon, Rutka, James, & Papsin, 2013; Yan & Liu, 2010; Yoshimura, Miyagawa, Kumakawa, Nishio, & Usami, 2016; Zagolski, 2008).

With some of these etiologies, vestibular loss occurs in nearly all cases, such as with meningitis and CHARGE syndrome (Cushing et al., 2013; Wineland et al., 2017); however, with other etiologies, the presence of vestibular loss can be variable (Cushing et al., 2013). With vestibular loss having variable representation with varying etiologies and the etiologies oftentimes being unknown, practitioners must

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rely on other factors to determine if a child with hearing loss is at risk for vestibular loss.

Vestibular loss has been associated with greater degrees of hearing loss and larger gross motor delay in young children. Regarding hearing loss severity, vestibular loss is estimated to occur in 30%–74% of children with profound hearing loss (Arnvig, 1955; Buchman, Joy, Hodges, Telischi, & Balkany, 2004; Jafari & Asad, 2011). Given this trend, it is not surprising that 50% of children who are candidates for a cochlear implant (CI) have some degree of vestibular loss (Cushing et al., 2013; Janky & Givens, 2015). Regarding gross motor function, typically developing children sit without support between the age of 3.8 and 9.2 months, crawl between 5.2 and 11.4 months, stand independently between 6.9 and 16.9 months, and walk independently between 8.2 and 17.6 months (World Health Organization [WHO], 2006). Children with vestibular loss are delayed in acquiring these milestones; they may acquire independent head control at 5 months or later and walk independently at 18 months or later (Inoue et al., 2013). Masuda and Kaga (2014) demonstrated that, in 16 children with severe hearing loss and reduced rotary chair responses, all 16 children evidenced delay in head control and walking. Although both hearing loss severity and gross motor function have been found to be associated with vestibular loss, neither of these factors has been specifically investigated to determine if they can predict the presence of vestibular loss.

Further compounding the gross motor delay found in children with vestibular loss is the presence of other comorbidities, which has been demonstrated to cause an increase in gross motor developmental delay. Kaga (1999) reported that the combination of vestibular loss and cognitive delay resulted in significantly larger gross motor developmental delay. They hypothesized that this increased delay in gross motor development for children with both vestibular loss and cognitive delays may be associated with slow central compensation, slow motor learning, or both. Nonetheless, these findings suggest that the presence of comorbidities should be investigated in these children.

It is important to identify vestibular loss at an early age because vestibular loss can result in additional consequences beyond gross motor developmental delay that can be addressed with physical therapy. In children with CI, vestibular loss has been correlated with increased CI device failure and increased fall risk (Wolter, Gordon, Papsin, & Cushing, 2015). Children with vestibular loss also have reduced dynamic visual acuity (Braswell & Rine, 2006a; Janky & Givens, 2015; Martin, Jelsma, & Rogers, 2012; Rine & Braswell, 2003; Rine et al., 2013), which has been linked to reduced reading acuity and the need for increased critical print size while reading (Braswell & Rine, 2006a). Although the consequences of vestibular loss in children are not fully understood, the delays in gross motor development, subsequent imbalance, and poor visual acuity that result from vestibular loss may account for some variability in the outcomes of children with hearing loss; thus, it is

important to identify children at risk for vestibular loss as early identification will lead to earlier intervention.

Early intervention for children with bilateral vestibular loss has been shown to improve some of the consequences of vestibular loss. Rine et al. (2004) completed a randomized controlled trial with an intervention group who completed vestibular rehabilitation exercises (i.e., eye-hand coordination, general coordination activities, visual motor training, and balance training) three times a week for 12 weeks and a placebo group who completed language development training during the same duration. After the intervention, children in the intervention group showed significant improvement in gross motor development scores, whereas the children in the placebo group did not. In fact, the children in the placebo group performed more poorly on the posttest, indicating that their gross motor delay was progressive. The children in the placebo group then completed the intervention and showed significant improvement in gross motor development, thus providing evidence for the effectiveness of vestibular rehabilitation to stop progression of gross motor developmental delay in children with vestibular loss (Rine et al., 2004). In addition, vestibular rehabilitation has been shown to provide improvement in dynamic visual acuity, reading acuity, and critical print size needed for reading (Braswell & Rine, 2006a, 2006b).

Despite the occurrence of vestibular loss in children with hearing loss, vestibular loss is not commonly assessed. Given the significant impact that vestibular loss can have on gross motor development and other outcomes (e.g., balance and reading acuity), as well as the positive role of early intervention, it is important to identify children with vestibular loss. Because the etiology of hearing loss is oftentimes unknown, other factors would be helpful for determining children at risk for vestibular loss. Therefore, the purpose of the study was to determine which factors provide the most value in predicting vestibular loss in children with hearing loss. The factors investigated included degree of hearing loss, anatomical imaging abnormalities of the labyrinth and VIII cranial nerve (i.e., magnetic resonance imaging [MRI]/computed tomography [CT] scan results), parental concerns for gross motor delay, the presence of comorbidities, and performance on the Developmental Profile-3 (DP-3), a parent interview that screens for developmental delay.

Method

A retrospective chart review was completed on two groups of children with hearing loss seen at Boys Town National Research Hospital for routine vestibular testing from June 1999 through September 2015. Children completed vestibular testing either through the neurosensory genetics (NSG) clinic or as part of their CI candidacy evaluations. The NSG clinic is a multidisciplinary team that sees children of all ages and configurations of hearing loss. Children seen in NSG clinic receive audiological; vestibular; ear, nose, and throat; ophthalmology; and genetics

evaluations. The primary purpose of the NSG clinic is to determine etiology of hearing loss. For the CI candidacy evaluations, children of all ages received vestibular testing before implantation. Children were excluded from the analysis if vestibular testing was not completed or could not be reliably interpreted.

Medical Chart Review

Each child's medical chart was reviewed to obtain the following data:

- **Vestibular loss severity:** Vestibular assessment consisted of either rotary chair via sinusoidal harmonic acceleration (MicroMedical Technologies, Chatham, IL), video head impulse testing (vHIT) in the horizontal canal via ICS Impulse (Otometrics, Schaumburg, IL), or caloric testing via ICS Chartr (Otometrics, Schaumburg, IL) using water calorics (ICS Charter Water Caloric Stimulator NCI-480; Otometrics, Schaumburg, IL). The method of vestibular testing was determined by patient age and tolerance. Vestibular loss severity was qualitatively categorized as (a) normal vestibular function, (b) bilateral vestibular loss (significantly low gain on rotary chair, sum of caloric responses less than 20°/s across four irrigations, or bilaterally reduced vHIT gain), and (c) mild-to-moderate vestibular loss (rotary chair phase leads with normal or near-normal gain, unilateral caloric weakness > 25%, or asymmetrically reduced vHIT gain). To be included in the analysis, it was preferred that subjects have at least two frequencies tested if rotary chair was completed. However, for three subjects, only one rotary chair frequency was completed; all of these subjects were interpreted to have normal vestibular function. Portions of each chart were reviewed by two researchers (K. L. J., M. T., or A. O.); however, in approximately 10% (18/186) of the subjects, the entire chart was reviewed by two researchers (M. T. and K. L. J.) because of questions regarding coding the different factors.
 - **Degree of hearing loss:** Audiological testing consisted of auditory brainstem response (ABR) and/or behavioral evaluations depending on the chronological and developmental age of the child. For some children, the ABR indicated a no response. For the purposes of this study, the ABR threshold was then recorded as 5 dB plus the highest level attempted during ABR testing. For example, if a no response was indicated at 90 dB nHL, the threshold was recorded as 95 dB nHL. A pure-tone average (PTA) was determined by averaging the thresholds for 250, 1000, 2000, and 4000 Hz because those were the frequencies available from ABR testing. A combined PTA for both ears was completed to create a representation of the overall hearing loss severity. Degree of hearing loss was quantified based on the classification scale suggested by Goodman (1965): mild (26–40 dB HL), moderate (41–55 dB HL), moderately severe (56–70 dB HL), severe (71–90 dB HL), and profound (91+ dB HL; Goodman, 1965).
 - **Imaging abnormalities:** CT and/or MRI results, when completed, were reviewed and based on the radiologist's impression, classified as either normal or abnormal. The CT or MRI was considered abnormal if there was evidence of anatomical abnormalities such as enlarged vestibular aqueduct, Mondini malformation, semicircular canal dehiscence, auditory nerve insufficiency, and stenotic ear canal, among others.
 - **Parental concerns:** Parental concerns for imbalance or gross motor delay were classified as either “yes, there is a concern” or “no, there is not a concern.” Parent report of age when their child sat (in months) and walked independently (in months) was also recorded. When parental concern could not be ascertained by the chart, data were classified as “no response.”
 - **Comorbidities:** Additional comorbidities were noted and coded as follows: (a) autism, (b) otitis media (OME), (c) musculoskeletal disorders (e.g., spina bifida, hypotonia, and tethered spinal cord), (d) neurological disorders (e.g., cerebral palsy, seizures, Infantile Refsum disease, and peripheral neuropathy), (e) heart defects (e.g., CHARGE syndrome, heart murmur, and bicuspid valve), (f) visual impairments, (g) prematurity (defined as birth at 36 weeks of gestational age or earlier), (h) attention-deficit/hyperactivity disorder, (i) cognitive delays (e.g., cerebral dysgenesis and microcephaly), (j) ototoxic medication exposure, and (k) gastrostomy tube. Subjects were classified as “yes” if they had one or more comorbidities or “no” if they did not have comorbidities.
 - **DP-3:** The DP-3 is a standardized interview/checklist assessment with parents/legal guardians as the responders and is appropriate for children aged birth through 12 years, 11 months. This assessment screens for developmental delay in five areas: (a) physical, (b) adaptive behavior, (c) social-emotional, (d) cognitive, and (e) communication. The DP-3 was administered by a certified speech-language pathologist. The standard score (mean = 100, *SD* = 15, range = 85–115) is reported for all areas, where scores of < 70 = delayed, 70–84 = below average, 116–130 = above average, and > 130 = well above average.
- Vestibular loss severity and degree of hearing loss were determined for each subject. However, the additional factors reviewed (imaging abnormalities, parental concerns, comorbidities, and DP-3) were not available for every child and were recorded when available. Tables 1 and 2 indicate the sample size included for each factor in the analysis.

Statistical Analysis

Independent one-way analysis of variance (ANOVA) was completed for each continuous variable to determine if there were significant mean differences between groups

Table 1. Demographic data and results from one-way ANOVAs.

Continuous variables	Normal	Mild to moderate	Bilateral	<i>p</i> Value
Age to sit	6.60 (<i>n</i> = 62)	8.19 (<i>n</i> = 27)	11.23 (<i>n</i> = 15)	< .001*
Age to walk	12.95 (<i>n</i> = 55)	15.33 (<i>n</i> = 18)	20.06 (<i>n</i> = 9)	< .001*
DP-3 physical ^a	91.72 (<i>n</i> = 32)	79.36 (<i>n</i> = 11)	77.73 (<i>n</i> = 15)	.048*
DP-3 adaptive ^a	94.56 (<i>n</i> = 32)	84.45 (<i>n</i> = 11)	86.00 (<i>n</i> = 15)	.221
DP-3 soc/emot ^a	79.68 (<i>n</i> = 32)	81.09 (<i>n</i> = 11)	71.64 (<i>n</i> = 14)	.318
DP-3 cognition ^a	78.44 (<i>n</i> = 32)	66.64 (<i>n</i> = 11)	72.57 (<i>n</i> = 14)	.268
DP-3 communication ^a	68.75 (<i>n</i> = 32)	67.09 (<i>n</i> = 11)	61.29 (<i>n</i> = 14)	.363
PTA: CI	91.85 (<i>n</i> = 57)	94.24 (<i>n</i> = 26)	92.55 (<i>n</i> = 27)	.662
PTA: NSG	42.41 (<i>n</i> = 55)	53.37 (<i>n</i> = 13)	84.74 (<i>n</i> = 2)	.004*

Note. ANOVA = analysis of variance; DP-3 = Developmental Profile-3; PTA = pure-tone average; CI = cochlear implant; NSG = neurosensory genetics.

^aDP-3 are reported as standard scores.

*Significant group differences on ANOVA.

with vestibular loss severity (normal vestibular function, mild-to-moderate vestibular loss, and bilateral vestibular). All follow-up analyses used Tukey's procedure. Chi-square analyses were completed for each categorical variable to determine if there were significant differences between groups with vestibular loss severity. A generalized logit model was then applied to estimate which factors were able to predict the children with vestibular loss. Receiver operating characteristic (ROC) curves and area under the ROC curve were also calculated for a select group of variables to determine specific cutoff values. With area under the ROC curve, an area of 1 represents a perfect separation of children with normal vestibular function from children with vestibular loss and an area of 0.5 represents an outcome with poor accuracy. A between-group ANOVA, using both group (normal vestibular function, mild-to-moderate vestibular loss, and bilateral vestibular) and comorbidity (yes or no) as fixed factors, was performed to determine the effect of comorbidity on all ordinal outcome variables. Last, binary logistic regression was completed to determine the effect of comorbidity on categorical variables.

Results

Subjects

Two hundred fifty-three charts were reviewed; 63 cases were excluded because of vestibular testing that

was either not completed or could not reliably be interpreted, resulting in 186 charts included in the analysis. There were 69 children in the NSG group (35 boys, mean age = 55 months, range = 2 months–14 years, 7 months; 34 girls) and 117 children in the CI candidacy group (54 boys, mean age = 23 months, range = 4 months–11 years, 7 months; 63 girls) at the time of vestibular testing. Children were grouped according to their vestibular loss severity; 115 (62%) children were classified as having normal vestibular function (55 from the NSG group and 60 from the CI candidacy group), 31 (17%) children were classified with bilateral vestibular loss (three from the NSG group and 28 from the CI candidacy group), and 40 (21%) children were classified with mild-to-moderate vestibular loss (13 from the NSG group and 27 from the CI candidacy group). In the 186 children, vestibular function was assessed via rotary chair (*n* = 164), caloric (*n* = 9), vHIT (*n* = 1), or a combination of evaluations (*n* = 12).

A one-way ANOVA was completed to determine if there were significant mean differences between groups with vestibular loss severity on any of the following continuous variables: age at which the child sat and walked independently, performance on the DP-3 (adaptive behavior, physical, communication, cognition, and social-emotional subscales), and severity of hearing loss. Demographic data and results from the ANOVA can be found in Table 1. Of all the factors, findings suggest significant mean differences between groups for age-to-sit, $F(2, 103) = 12.593, p < .001$;

Table 2. Frequency counts and results from chi-square analyses.

Categorical variables		Normal	Unilateral	Bilateral	<i>p</i> Value
Parental concern	Yes	23	23	28	< .001*
	No	87	16	2	
Imaging abnormalities	Normal	46	13	12	.314
	Abnormal	25	14	9	
Comorbidities	Yes	24	14	20	< .001*
	No	91	26	11	

*Significant group differences on chi-square analysis.

age-to-walk, $F(2, 81) = 15.44, p < .001$; and DP-3 physical scale, $F(2, 57) = 3.22, p = .048$. Post hoc analyses suggest that age-to-sit significantly increased with the severity of vestibular loss, shown in Figure 1a. Children with bilateral vestibular loss sat significantly later than children with mild-to-moderate vestibular loss ($p = .013$) and children with normal vestibular function ($p \leq .001$). Similarly, age-to-walk significantly increased with the severity of vestibular loss, shown in Figure 1b. Children with bilateral vestibular loss walked significantly later than children with mild-to-moderate vestibular loss ($p = .007$) and children with normal vestibular function ($p \leq .001$). For the DP-3 physical scale, the children with bilateral and mild-to-moderate vestibular loss scored more poorly than the children with normal vestibular function; however, this did not reach significance in post hoc testing ($ps = .073$ and $.187$, respectively).

Severity of hearing loss was assessed separately for the children seen for CI candidacy and NSG. As shown in Table 1 and Figure 2, there were no group differences in severity of hearing loss for the children seen for CI candidacy, $F(2, 109) = 0.415, p = .662$, which was expected, as the children had similar severity of hearing loss necessitating the CI; however, for the children seen through the NSG clinic, severity of hearing loss increased with the severity of vestibular loss, $F(2, 69) = 6.124, p = .004$.

Chi-square analysis was completed to determine if group differences occurred for any of the categorical variables: imaging abnormalities (normal or abnormal), parental concerns for gross motor development (yes or no), or presence of comorbidities (yes or no). As shown in Table 2, there was no significant relationship between imaging abnormalities and the group with vestibular loss ($\chi^2 = 2.318, p = .314$); however, there was a significant relationship between parental concerns for gross motor development and the group with vestibular loss ($\chi^2 = 57.378, p < .001$). Post hoc analyses demonstrated a significantly higher proportion of parents reporting concern for gross motor skills as severity of vestibular loss increased ($p < .001$). There was also a significant relationship between the presence of comorbidities and the group with vestibular loss ($\chi^2 = 22.023,$

$p < .001$). Post hoc analyses demonstrated a significantly higher proportion of comorbidities in the group with bilateral vestibular loss, compared with the remaining groups ($p < .025$). The frequency distribution of comorbidities across all subjects is as follows: autism ($n = 1$), OME ($n = 11$), musculoskeletal ($n = 14$), neurological ($n = 29$), heart defect ($n = 5$), vision loss ($n = 18$), prematurity ($n = 10$), attention-deficit/hyperactivity disorder ($n = 1$), cognition ($n = 1$), ototoxic medications ($n = 5$), and gastrostomy tube ($n = 4$).

In summary, we find that, as severity of vestibular loss increases, children (a) sit and walk later, (b) score more poorly on the DP-3 physical subscale, (c) have more severe hearing loss (specifically for the cohort of children seen through the NSG clinic), (d) have parents who more frequently report concerns for gross motor delay, and (e) are more likely to have other comorbidities.

A generalized logit model was then applied to estimate which factors were able to predict the children with vestibular loss. Results from the logit model can be found in Table 3. For each factor, the first odds ratio compares children with bilateral vestibular loss with children with normal vestibular function, and the second odds ratio compares children with mild-to-moderate vestibular loss with children with normal vestibular function. For age, a 2-month increment was chosen for calculating odds ratios; and a 5-point/dB increment, for each of the test scores and PTA (Powers & Yue, 2008). For example, the odds ratio for age-to-sit is 2.226, suggesting that, for every 2-month increase in onset of sitting, the odds of having bilateral vestibular loss are 2.2 times that of having normal vestibular function. Of all the factors, age-to-sit, age-to-walk, PTA, and parental concerns for gross motor developmental delay had the greatest ability to differentiate children with vestibular loss (mild-to-moderate and bilateral) from children with normal vestibular function. These four factors suggest that the later a child sits and stands and the greater the severity of hearing loss, the greater the odds the child has either mild-to-moderate or bilateral vestibular loss compared with normal vestibular function.

Figure 1. Both age-to-sit (A) and age-to-walk (B) increase as the severity of vestibular loss increases. The line in each panel represents the cutoff values from receiver operating characteristic analysis.

Figure 2. Mean pure-tone average for the cochlear implant (CI) candidacy and neurosensory genetics (NSG) groups according to severity of vestibular loss. For the NSG group, severity of hearing loss increased with the severity of vestibular loss. The lines represent the cutoff values from receiver operating characteristic analysis.

In addition, two factors—the physical score from the DP-3 and the presence of comorbidities—were able to differentiate children with bilateral vestibular loss from children with normal vestibular function, but not children with mild-to-moderate vestibular from children with normal vestibular function. For the DP-3 physical scale, the significant odds ratio suggests that the lower the score on the DP-3, the greater the odds the child has bilateral vestibular loss compared with normal vestibular function. For the presence of comorbidities, the significant odds ratio suggests that the more comorbidities, the more likely the

child has bilateral vestibular loss compared with normal vestibular function.

ROC curves were then analyzed for the factors with the greatest ability to differentiate children with vestibular loss (mild-to-moderate and bilateral) from children with normal vestibular function: age-to-sit, age-to-walk, and PTA for the NSG group. For age-to-sit, ROC analysis showed an area under the curve of 0.751 ($SE = 0.049$). Using a cutoff value of 7.25 months yielded a sensitivity of 62% and a specificity of 81%. For age-to-walk, ROC analysis showed an area under the curve of 0.764 ($SE = 0.062$). Using a cutoff value of 14.5 months yielded a sensitivity of 78% and a specificity of 77%. For PTA for the NSG group, ROC analysis showed an area under the curve of 0.721 ($SE = 0.073$). Using a cutoff value of 40 dB yielded a sensitivity of 80% and a specificity of 55%; however, using a cutoff value of 66 dB yielded a sensitivity of 33% and an improved specificity of 91%.

Because a significant number of children evidenced comorbidities, particularly those with bilateral vestibular loss, a between-group ANOVA, using both group (normal vestibular function, mild-to-moderate vestibular loss, and bilateral vestibular) and comorbidity (yes or no) as fixed factors, was performed to determine if the mean differences noted between groups, outlined above, were due to vestibular loss or the effect of comorbidity. As shown in Table 4, there was a significant effect of comorbidity for all DP-3 subtests (physical, adaptive, social-emotional, cognition, and communication), with poorer DP-3 scores for children with one or more comorbidity, suggesting that this parental questionnaire is influenced by the presence of comorbidities. For the DP-3 social-emotional scale, the children with bilateral vestibular loss (71.6) scored more poorly than the children with normal vestibular function (79.7) or mild-to-moderate vestibular loss (81.1); however, this did not reach significance in post hoc testing. For age-to-sit,

Table 3. Results from the generalized logit model.

Factors	Bilateral vestibular loss versus normal vestibular function				Mild-to-moderate vestibular loss versus normal vestibular function			
	OR	CI for OR		<i>p</i>	OR	CI for OR		<i>p</i>
		Lower	Upper			Lower	Upper	
Age to sit (2 months)	2.226	1.442	3.437	< .001*	1.679	1.128	2.497	.010*
Age to walk (2 months)	2.326	1.477	3.663	< .001*	1.591	1.120	2.260	.010*
DP-3—physical (5 points)	0.838	0.713	0.985	.032*	0.854	0.715	1.018	.079
DP-3—adaptive (5 points)	0.894	0.763	1.047	.165	0.878	0.738	1.045	.144
DP-3—social-emotional (5 points)	0.878	0.730	1.056	.167	1.023	0.842	1.242	.820
DP-3—cognitive (5 points)	0.932	0.800	1.087	.373	0.876	0.742	1.035	.120
DP-3—communicate (5 points)	0.850	0.680	1.064	.156	0.969	0.785	1.198	.774
PTA (5 dB)	1.261	1.116	1.424	< .001*	1.097	1.019	1.182	.014*
Parental concern (yes vs. no)	52.957	11.739	238.891	< .001*	5.437	2.477	11.938	< .001*
Imaging (normal vs. abnormal)	1.380	0.512	3.722	.525	1.982	0.807	4.866	.138
Comorbidities (yes vs. no)	6.894	2.910	16.331	< .001*	2.042	0.926	4.500	.077

Note. For age, a 2-month increment was chosen for calculating odds ratios, and a 5-point/dB increment for each of the test scores and PTA, shown in parentheses. CI = cochlear implant; OR = odds ratio; DP-3 = Developmental Profile-3; PTA = pure-tone average.

*Bold indicates significant predictor.

Table 4. Results from statistical analyses with group and comorbidity as fixed factors.

Variables	Group	Comorbidity	Interaction
Age to sit	$F = 6.54, p = .002^*$	$F = 11.05, p = .001^*$	$F = 1.67, p = .193$
Age to walk	$F = 11.68, p < .001^*$	$F = 10.16, p = .002^*$	$F = 3.72, p = .029^*$
DP-3 physical ^a	$F = 0.64, p = .533$	$F = 8.09, p = .006^*$	$F = 0.30, p = .746$
DP-3 adaptive ^a	$F = 0.63, p = .535$	$F = 12.46, p = .001^*$	$F = 1.96, p = .151$
DP-3 soc/emot ^a	$F = 3.62, p = .034^*$	$F = 7.31, p = .009^*$	$F = 2.97, p = .060$
DP-3 cognition ^a	$F = 0.08, p = .923$	$F = 4.95, p = .030^*$	$F = 1.38, p = .262$
DP-3 communication ^a	$F = 1.52, p = .229$	$F = 4.06, p = .049^*$	$F = 1.56, p = .221$
PTA: CI	$F = 0.85, p = .437$	$F = 0.08, p = .782$	$F = 0.88, p = .418$
PTA: NSG	$F = 6.05, p = .004^*$	$F = 0.58, p = .448$	$F = 0.06, p = .947$
Parental concern	$\beta = 0.96, p < .001^{**}$	$\beta = 1.51, p = .096$	$\beta = 0.12, p = .816$
Imaging	$\beta = 0.02, p = .941$	$\beta = -1.41, p = .165$	$\beta = 0.99, p = .064$

Note. DP-3 = Developmental Profile-3; PTA = pure-tone average; CI = cochlear implant; NSG = neurosensory genetics.

^aDP-3 are reported as standard scores.

*Significant group differences on analysis of variance. **Significant group differences on binary logistic regression.

there was a significant effect of comorbidity, with significantly later mean age-to-sit when one or more comorbidity is present (10 months) compared with no comorbidities (6.8 months). There was a main effect of group in that children with bilateral vestibular loss sat (11.2 months) later than children with normal vestibular function (6.6 months). For age-to-walk, there was a significant interaction where the children with bilateral vestibular loss and one or more comorbidity demonstrated the latest age-to-walk independently (23.6 months) compared with all other groups. For PTA in the NSG group, there was a main effect of group. The group with bilateral vestibular loss had a significantly greater mean PTA (92 dB, $SD = 10$ dB) than the group with normal vestibular function (67.6 dB, $SD = 29.5$ dB). However, there was no effect of comorbidity. Last, for those in groups with vestibular loss, parents were more likely to express concern regarding their child's gross motor development, which was independent of comorbidities. There was no effect of group or comorbidity on imaging abnormalities.

Discussion

The purpose of the study was to determine which factors provide the most value in predicting vestibular loss in children with hearing loss. Of 186 children with hearing loss, 38% were diagnosed with some degree of vestibular loss. The greatest proportion of children with vestibular loss was from the CI candidacy group, where 47% of the children had vestibular loss. This is consistent with previous prevalence data reporting that half of the children with CI have some degree of vestibular loss (Cushing et al., 2013; Jacot, Van Den Abbeele, Debre, & Wiener-Vacher, 2009; Janky & Givens, 2015). Alternatively, 23% of the children from the NSG group, where the severity of hearing loss varied from mild to profound, had some degree of vestibular loss. These data support previous reports that hearing loss is associated with vestibular loss (O'Reilly et al., 2010) and that increased severity of hearing loss is associated with an increased risk of vestibular loss (Brookhouser

et al., 1982; Brookhouser, Cyr, Peters, & Schulte, 1991; Tribukait, Brantberg, & Bergenius, 2004). Specifically, children with bilateral vestibular loss have been shown to have significantly more severe hearing loss compared with those with normal or unilateral vestibular loss (Brookhouser et al., 1982). A worsening of auditory thresholds with increased vestibular severity was not evident in the CI candidacy group, as these children all had similar severity of hearing loss; however, the higher prevalence of vestibular loss in this group (47%) compared with the NSG group (23%) helps support this trend.

Across both groups of children (CI candidacy and NSG groups), as the severity of vestibular loss increased, performance on several factors was found to decline. First, children sat and walked later with increasing vestibular loss severity. This is consistent with several studies that have similarly noted reduced functional performance with increased severity of vestibular loss (Cushing, Papsin, Rutka, James, & Gordon, 2008; De Kegel, Maes, Baetens, Dhooge, & Van, 2012; Janky & Givens, 2015). Reduced balance performance in children with vestibular loss has been noted on a variety of examinations including the Bruininks-Oseretsky Test of Motor Proficiency–Second Edition, a standardized test of balance function, posturography, the Movement Assessment Battery for Children–Second Edition, Dynamic Gait Index, Dynamic Visual Acuity Test, and single-leg stance (Cushing et al., 2008; De Kegel et al., 2012; Janky & Givens, 2015). In all instances, performance has been correlated with vestibular function testing, where those children with poorer vestibular function have poorer performance. In this study, gross motor acquisition followed a similar trend. Children with normal vestibular function sat at 6.6 months and walked at almost 13 months on average. Both values fell within the WHO's range for typically developing children (sit between 3.8 and 9.2 months and walk between 8.2 and 17.6 months). Children with mild-to-moderate vestibular loss acquired these skills somewhat later, sitting at 8.2 months and walking at 15.3 months on average. Finally, the children with bilateral vestibular loss

sat at 11.2 months and walked at 20 months on average, which is well outside the WHO's range, making them an easier group to discriminate.

Given the trend of decreased performance with increased vestibular loss severity, it is not surprising that these same children had parents who were more likely to report concerns for gross motor delay and also scored their child's performance more poorly on the DP-3 physical subscale. Because simply asking parents whether they are concerned about their child's gross motor development ("yes" or "no") was highly predictive of the presence of vestibular loss, it seems obvious that this question should be the initial case history question when investigating whether there is a risk that a child with hearing loss may have vestibular loss. Additional case history questions regarding the precise age the child met specific motor milestones could then be used as validation. Although the DP-3 physical subscale was helpful for appropriately documenting gross motor delays and differentiating bilateral vestibular loss from children with normal vestibular function, the DP-3 was vulnerable to comorbidities, suggesting that this particular standardized questionnaire may not be a suitable screening tool for vestibular loss compared with direct case history.

Finally, as severity of vestibular loss increased, children were more likely to have other comorbidities. Brookhouser et al. (1991) similarly noted that infants with abnormal ABR and vestibular loss were more likely to have high-risk factors (Brookhouser et al., 1991). We are uncertain about the exact relationship between the presence of vestibular loss and all of the comorbidities. However, regardless of the underlying reason for this association, these data suggest that questions regarding the presence of any comorbidities should be part of the pediatric case history and may provide additional evidence for possible vestibular loss.

In addition to vestibular loss, some comorbidities have been shown to increase gross motor developmental delay (Kaga, 1999), which could affect the sensitivity of using gross motor delay as a screening tool for vestibular loss. In our cohort, the number of comorbidities was too diffuse to perform any statistical analyses to determine trends on the type of comorbidity for each of the groups with vestibular loss. However, in the list of recorded comorbidities, several are associated with gross motor delay, such as OME, congenital heart defects, and neurologic disorders such as cerebral palsy (Casselbrant, Villardo, & Mandel, 2008; Cohen, Mandel, Furman, Sparto, & Casselbrant, 2011; Jones, Morgan, Shelton, & Thorogood, 2007), which suggests that, when using gross motor function to predict vestibular loss, comorbidities should be considered.

Of all the factors, age-to-sit, age-to-walk, PTA, and parental concerns for gross motor developmental delay had the greatest ability to differentiate children with vestibular loss (mild-to-moderate and bilateral) from children with normal vestibular function. Several studies have attempted to predict the presence of vestibular loss in children with hearing loss using measures of gross motor function. Brookhouser et al. (1982) demonstrated that the Tandem Romberg was helpful for identifying children with a caloric weakness.

The Tandem Romberg was abnormal in 8 of the 17 children with unilateral vestibular loss and 18 of the 20 children with bilateral vestibular loss (Brookhouser et al., 1982). Janky and Givens (2015) demonstrated that the amount of vestibular loss was useful for predicting performance on the Dynamic Gait Index, Dynamic Visual Acuity Test, single-leg stance, and sensory organization test conditions that relied on vestibular function (Conditions 5 and 6); children with a greater amount of vestibular loss performed more poorly than children with normal vestibular function (Janky & Givens, 2015). Last, Oyewumi et al. (2016) found that single-leg stance with eyes closed and tandem stance with eyes open, both subtests of the Bruininks-Oseretsky Test of Motor Proficiency-II, provided the best sensitivity and specificity for identifying children with bilateral vestibular loss (Oyewumi et al., 2016). Although these studies are helpful for predicting when an older child is at risk for vestibular loss, they do not provide assistance for making similar decisions in younger children.

Similar to these studies, we demonstrate that there are specific factors that can predict vestibular loss, but in a younger population. Age-to-sit, age-to-walk, bilateral PTA, and parental concerns for gross motor developmental delay were predictive of vestibular loss. We speculate that these four factors may be helpful for determining when a referral for vestibular testing might be made for children with hearing loss in infancy. For age-to-sit and age-to-walk, ROC analysis suggests that using a cutoff of 7.25 months for age-to-sit and 14.5 months for age-to-walk provides reasonable sensitivity and specificity for identifying vestibular loss. Although these cutoffs fall within the normal range published by WHO (2006), 3.8–9.2 months for sitting and 8.2–17.6 months for walking, concern for vestibular loss should be piqued when the acquisition of these milestones extends beyond these cutoffs and particularly when these skills are acquired well beyond the range provided by WHO. For hearing loss severity, ROC analysis suggests that using a bilateral PTA cutoff of 40 dB has excellent sensitivity (80%) and a bilateral PTA cutoff of 66 dB has excellent specificity (91%). As shown in Figure 2, the cutoff value of 66 dB identifies all children with bilateral vestibular loss. Collectively, these factors appear to be more sensitive for identifying children with bilateral vestibular loss compared with children with mild-to-moderate vestibular loss; however, none of these values provides 100% sensitivity and specificity. Despite this, they do provide a framework for when vestibular testing might be recommended in an infant diagnosed with hearing loss.

Limitations

There were several limitations for the current study. First, this was a retrospective study design. Parents were not questioned prospectively; therefore, we have many missing data points in terms of the number of parents concerned about their child's gross motor development and the exact age the child sat and walked, among other variables. In addition, several of the factors reviewed were based on

subjective parental report and limited to the information provided in clinical documentation, which may have been accumulated over several visits. Because this was retrospective, a control group with normal hearing was not included. A prospective study is planned, which would increase data for each of the variables to improve statistical power and include a control group with normal hearing.

Second, a bilateral PTA was used to describe the overall severity of hearing loss. Although this metric is appropriate for children with bilateral, symmetric hearing loss, the bilateral PTA may not be an accurate reflection of hearing loss severity for children with unilateral, asymmetric hearing loss. The bilateral PTA was used because vestibular function was assessed in a significant number of children using rotary chair (88%). In instances of unilateral vestibular loss, rotary chair is unable to localize which ear has vestibular loss, precluding the ear with vestibular loss from being matched with the correct severity of hearing loss. For this reason, bilateral PTA was felt to provide the best overall assessment of hearing loss severity.

Third, the use of rotary chair in estimating the severity of vestibular loss is challenging, particularly in infants. In the group with mild-to-moderate vestibular loss, it is difficult to know the extent of vestibular loss based on phase lead alone, paired with either normal or borderline low gain. These subjects likely had unilateral vestibular loss. Although the presence of rotary chair phase leads has been associated with caloric asymmetries, the magnitude of the phase lead is not always related to the magnitude of the vestibular loss (Suzuki, Pulec, & Smith, 1989), which is why all children with phase lead as their only finding were lumped into one group. However, it is possible that some had a mild bilateral vestibular loss. Alternatively, a rotary chair phase lead may also indicate lesions affecting the brainstem and not only the peripheral vestibular system (Baloh, Yee, Kimm, & Honrubia, 1981). Isolated rotary chair phase leads do not determine the ear involved, which can be particularly difficult when testing children, and additional testing or a reliable case history cannot be gleaned. Given the relationship between vestibular loss and severity of hearing loss, the involved ear is generally the ear with greater hearing loss; however, as described above, this assumption was not made, and a bilateral PTA was used when investigating the relationship between hearing and vestibular loss. When testing infants in the rotary chair, there is the additional factor of reliability. In some children, rotary chair was completed at several frequencies (0.01–0.16 Hz), whereas in other children, rotary chair testing was limited to one to two frequencies based on child tolerability. Last, caloric or vHIT, and not rotary chair, was used to classify subjects, which may have resulted in misclassification due to differences in the frequencies assessed with each test.

Fourth, etiology was not evaluated as a factor in this study as genetic testing had not been completed in most subjects nor had a diagnosed cause for their hearing loss been determined. Previous reports on the prevalence of vestibular loss with different etiologies suggest that vestibular loss should be suspected in cases of meningitis,

cytomegalovirus, Pendred, Usher, and Waardenburg, among others; therefore, in cases where the etiology is clear and vestibular loss is a known association, vestibular evaluation and subsequent physical therapy should be recommended (Brookhouser et al., 1982; Cushing et al., 2013; Yan & Liu, 2010; Yoshimura et al., 2016; Zagolski, 2008). Vestibular loss has also been associated with severe hearing loss acquired after birth (Arnvig, 1955; Selz, Girardi, Konrad, & Hughes, 1996). Therefore, when etiology is unclear, the degree and time of onset of hearing loss can provide justification for vestibular evaluation.

Last, our inclusion of comorbidities was not exhaustive. We only reported the absence or presence of a select few comorbidities and were not able to quantify the relative contribution of each (i.e., if certain comorbidities were more or less likely to affect gross motor development). There are likely other comorbidities that will impact motor development or the presence of vestibular loss that were not present in this population or were not included.

Conclusion

Findings suggest that parental concerns for gross motor delay, delayed milestones for sitting and walking, and degree of hearing loss are indicators of vestibular loss in young children with hearing loss. These factors are particularly helpful for identifying children with bilateral vestibular loss. Therefore, it is recommended that these factors be incorporated in any case history or screening tool for children with hearing loss. We recommend that parents be questioned regarding concerns for gross motor development and gross motor skill acquisition. A referral for vestibular evaluation should be considered for children whose hearing loss is greater than 66 dB and particularly those who sit later than 7.25 months or walk later than 14.5 months or whose parents report concerns for gross motor development. Because of the benefit of physical therapy, children identified with vestibular loss should then be referred to physical therapy for further evaluation and treatment.

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