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Original Article

A Prospective Study of the Hearing Loss in Global Developmental Delay Children

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Abstract

Background: Developmental disabilities are a group of related chronic disorders of early onset estimated to affect 5–10% of children. Global developmental delay is a subset of developmental disabilities defined as a significant delay in two or more of the following developmental domains: Gross/fine motor, speech/language, cognition, social/personal, and activities of daily living.

Aim of the Study: The aim of this study was to describe the clinical profile and audiological profile in children with global developmental delay presenting to the pediatric ENT unit.

Materials and Methods: The study sample size was a total of 121 children with global developmental delay. Children with complaints of global developmental delay underwent a detailed ENT examination including examination under microscope of ear which is the standard of care. Hearing loss was assessed by audiological tests such as behavioral observation audiometry (BOA), otoacoustic emission, brain stem evoked response audiometry (BERA), and tympanometry (Tymps). The degree of hearing loss was classified using the American Speech-Language-Hearing Association classification.

Observations and Results: Among 121 children with global developmental delay, there were 72 (59.5%) males. The mean age of the study group was 3.2 years. The youngest child in the study was 6 months old and the oldest child being 14 years old. 25 (20.6%) children participating in the study had syndromic association. Of 121 children, only 36 (29%) presented with speech delay and suspected hearing loss. BOA done in 242 ears showed 56 (23%) ears with normal hearing, 68 (28%) with hearing loss, and inconsistent report in 38 (15.5%) ears. In the 80 remaining ears (33%), test could not be done.

Conclusions: The mean age of referral was 3.2 years in global developmental delay children who were referred for the evaluation of speech delay. Among the 121 global developmental delay children included in the study, 36 (29%) had hearing loss with speech delay. Our study detected a higher incidence of undetected hearing loss of 144 ears (59.5%) in children with global developmental delay.

Key words: Behavioral observation audiometry, Brain stem evoked response audiometry, Global developmental delay, Hearing loss, Tympanometry

INTRODUCTION

Developmental disabilities are a group of related chronic disorders of early onset estimated to affect 5–10% of children.[1-3] Global developmental delay is a subset of developmental disabilities defined as significant delay in two or more of the following developmental domains: Gross/fine motor, speech/language, cognition, social/personal, and activities of daily living.[4-6] Children with global developmental delay require complex, individualized therapy to maximize their long-term quality of life. One subset of children with special needs includes those with both developmental delays and deafness. One of the major causes of global developmental delay is cerebral palsy (CP). CP was first described in 1862 by an orthopedic surgeon named William James Little.[7] CP is a condition caused by damage to the brain, usually occurring before, during, or shortly after birth. "Cerebral" refers to the brain and "palsy" refers to a disorder of movement or posture. CP is a central nervous system (CNS) disorder of movement,

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coordination, and posture, reflecting a non-progressive abnormality or insult to the immature brain.[8,9] CP is neither progressive nor communicable. It is also not curable, although education, therapy, and applied technology can help people with CP to lead productive lives. Children with CP have an organic complication in the peripheral and CNS.[10] Therefore, CP is often accompanied by other disorders and problems of cerebral function, in particular speech and language impairment, intellectual impairment, disorders of vision and hearing, attention, vigilance, and behavior.[11,12] Consequently, the majority of the affected individuals cannot participate and find their place in normal society.[13-15] Advances in the treatment of prenatal infections and improved medical facilities for neonatal care have resulted in more surviving infants but with complications that may include hearing impairment. In particular, reduced mortality rates of premature babies with very low birth weight are responsible for more infants with complications including hearing impairment. Hearing impairment estimates as high as one-third population of CP; a study done by Susan *et al.* shows 4–13% hearing loss in children with CP![16,17] Hearing impairment is the most common in children with very low birth weight or severe hypoxic-ischemic insults.[18-20] The existence of hearing problems with unique motor problem in CP presents a range of special educational and psychological needs, to an even greater degree than for children with single disability. Untreated reduced hearing acuity during infancy and early childhood compounded with additional disability may have a more deleterious effect on communication abilities, speech and language, and cognitive development that can severely interfere with their parent-child and peer-child interactions, low self-esteem, and linguistic, auditory perceptual, and educational development.[21,22] However, the effects of hearing impairment are amenable to the treatment and rehabilitation strategies if identified at an early age and effective intervention program is initiated. Thus, overall future and success of a child can be improved by reducing the complications of hidden disability of hearing impairment.[23-25] However, there are no data available in the Indian context, with regard to the degree and type of hearing impairment in children with global developmental delay. Hence, we propose to study the presence of hearing impairment in children with global developmental delay. The aim is to increase the awareness of possible correctable audiological impairment that hinders development and learning in children with global developmental delay children.

Aim of the Study

The aim of this study was to describe the clinical profile and audiological profile in children with global developmental delay presenting to pediatric ENT unit and to determine the type and severity of hearing impairment in children with global developmental delay.

MATERIALS AND METHODS

This prospective study was done on children with global developmental delay who presented to the ENT 2 outpatient care (OPD) between October 2017 and April 2018. Patients were selected as per the inclusion and exclusion criteria. The study was explained to every patient in detail and they were included in the study after obtaining informed written consent. The sample size was calculated from the statistical input taken from the reference article by Susan *et al.* in 2011. Taking $P = 81.8\%$ and precision as 6, the sample size was calculated using Master software version 2.0. The study sample size was a total of 121 children with global developmental delay.

$$n = \frac{Z^2_{1-\frac{\alpha}{2}}\, p(1-p)}{d^2}$$

Where,

- p: Expected proportion
- d: Absolute precision
- $1-\alpha/2$: Desired confidence level.

Inclusion Criteria

(1) All children with global developmental delay from 6 months to 16 years attending ENT-2 OPD and (2) parent giving consent to be part of the study were included in the study.

Exclusion criteria

(1) Children less than 6 months of age and (2) parents not giving consent to be part of the study were excluded from the study.

Clinical evaluation

Children presenting to ENT 2 OPD with the complaints of global developmental delay underwent a detailed ENT examination including examination under microscope of ear which is the standard of care. Hearing loss was assessed by audiological tests which included behavioral observation audiometry (BOA), otoacoustic emission (OAE), brain stem evoked response audiometry (BERA), and tympanometry (tymps). BERA was done using intelligent hearing screening machine to assess hearing loss. In BERA, click stimulus of 60–90 dB above hearing threshold in the frequency of 2–4 KHz and tone burst both are used. The stimulus rate used was 30.1 clicks/s. Active electrode (red) was placed over the forehead, reference electrode (black) was placed over ipsilateral mastoid or ear lobe, and ground electrode was placed over contralateral mastoid. Tympanometry was done in all children to use Grason Stadler 61 tympanometer. The degree of hearing loss was classified using the American Speech-Language-Hearing Association (ASHA) classification [Table 1].

The numbers are representative of the patient's hearing loss range in decibels (dB HL).

Outcomes measured were as follows: (1) Otomicroscopy findings of tympanic membrane, (2) the degree of hearing loss in global developmental delay children using the ASHA classification, (3) OAE–distortion product OAE, and (4) type of curve in tympanogram curve.

- Type A - Normal curve having a single peak of admittance between 150 and 100 daPa and a volume of 0.2–1.8 mL.
- Type B or flat curve - Flat curve with no admittance peak.
- Type C - Admittance peak shifted to negative pressures.

All the patients were assessed, and findings were noted down.

Appropriate statistical tests were used to analyze the data. For categorical data, the number of patients and percentage were presented. Chi-square or Fisher's exact test was applied to the data. All tests were two-sided at $\alpha = 0.05$ level of significance. All analyses will be done using Statistical Package for the Social Services software Version 21.0 (Armonk, NY: IBM Corp).

OBSERVATIONS AND RESULTS

A total of 121 children meeting the inclusion criteria were recruited in the study after parental consent. All the children underwent detailed ENT examination including examination under microscope of ear and audiological assessment.

Table 1: The ASHA classification

Degree of hearing loss	Hearing loss range (dB HL)
Normal	–10–15
Minimal	16–25
Mild	26–40
Moderate	41–55
Moderately severe	56–70
Severe	71–90
Profound	91+

ASHA: American Speech-Language-Hearing Association

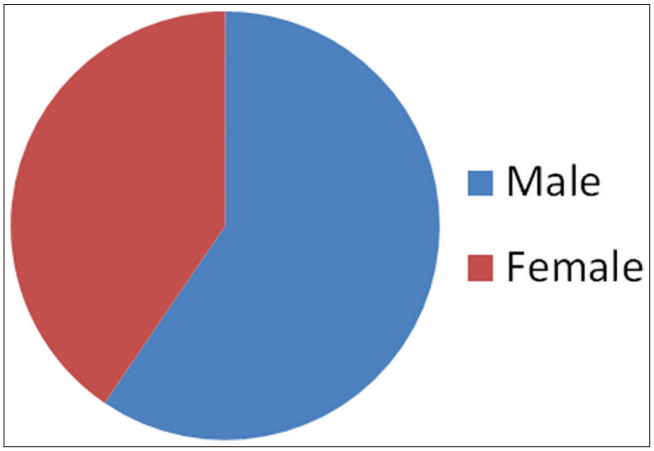

Figure 1: Gender distribution

Gender and Age Distribution

Majority of the participants was male, 72 (59.5%) [Figure 1].

The mean age of the study group was 3.2 years. The youngest child in the study was 6 months old and the oldest child being 14 years old. Nearly three-forth (76%) of the participants were under the age of 5 years [Figure 2].

Syndromic and Autism Spectrum Disorder Association

About 25 (20.6%) children participating in the study had syndromic association [Figure 3].

Of 25 syndromic children, two children had congenital rubella syndrome, 12 children had Down's syndrome, three children with mucopolysaccharidosis-associated syndromes, two children with West syndrome, one child each with fragile syndrome, opercular syndrome, Cornelia de Lange syndrome, orofacial digital syndrome, opercular syndrome, oculo-auriculo-vertebral spectrum syndrome, and Schinzel syndrome, and 10 children (8.2%) had autistic features [Figure 4].

CP: More than half of the children in the study had CP (55.6%). Of the 67 children with CP, 14 (20%) had hypotonic CP, 33 (49.25) had spastic CP, 13 (19.4%) had dystonic CP, and 7 (10%) had mixed variety [Figure 5].

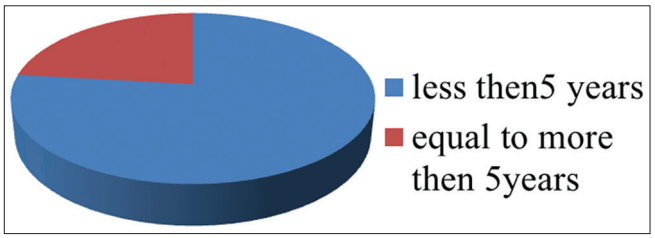

Figure 2: Age distribution

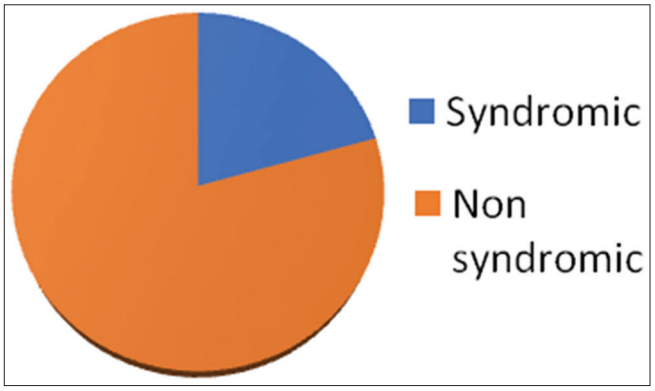

Figure 3: Syndromic and non-syndromic distribution

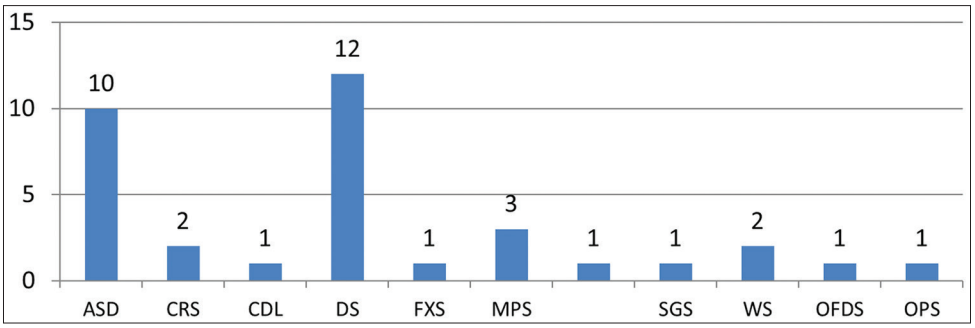

Figure 4: Various syndrome and autism spectrum disorder distribution

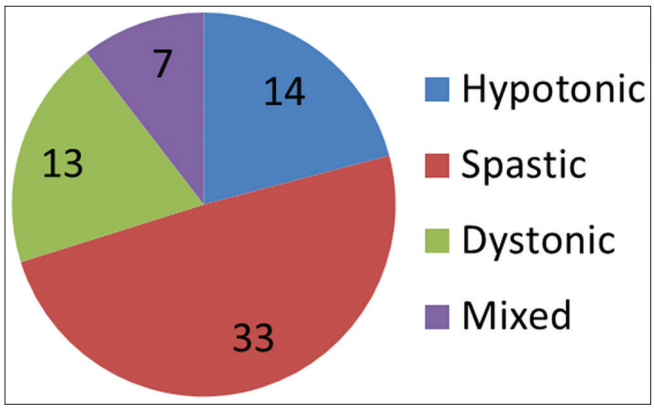

Figure 5: Types of cerebral palsy

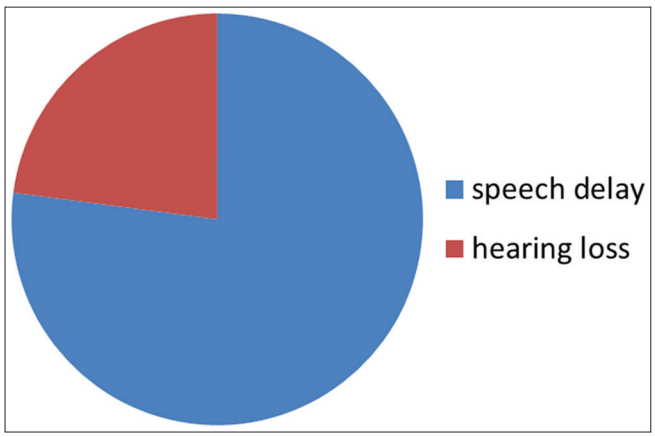

Figure 6: Presenting complaints

Presenting Complaint

Of 121 global developmental delay children all presented with speech delay, only 36 (29%) presented with suspected hearing loss with speech delay [Figure 6].

Otomicroscopy findings

In this study, a total of 242 ears were examined. Otomicroscopy revealed an intact and normal tympanic membrane in majority of the subjects, 233 (96.2%), 7 (2.8%) had a dull tympanic membrane, and one (0.4%) ear had otitis media with effusion. One child in the study had unilateral grade 3 microtia [Figure 7].

Hearing Assessment

(1) BOA: Of 242 ears assessed with BOA, 56 (23%) ears had normal hearing, 68 (28%) had hearing loss, and 38 (15.5%) ears had inconsistent report. In the eighty remaining ears (33%), test could not be done as the children were either uncooperative for the test or an auditory brain evoked response audiometry was done instead [Figure 8].

Of 68 ears examined for hearing loss, the severity was found to be profound in (29%), severe in (27%) and mild in (22%) [Figure 9].

OAE

Of 242 ears evaluated, OAE were absent in 109 (45%) ears and present in 56 (23%) ears [Figure 10].

BERA

Of 242 ears examined for BERA, 78 (32.2%) had normal hearing, 20 (8.2%) had minimal loss, 23 (9.5%) had mild hearing loss, 26 (10.7%) ears had moderate hearing loss, 3 (1.2%) ears had moderately severe hearing loss, 20 (8.2%) ears had severe hearing loss, 52 (21.1%) ears had profound, and BERA was not done in 20 (8.2%) ears [Figure 11].

Tympanometry

Of 242 ears, 145 (59.9%) ears had A type curve, 27 (11.1%) ears had B type curve, and 37 (15.2%) ears have C type curve [Figure 12].

Hearing status of children with CP

Of 67 children with CP with global developmental delay, hearing assessment by BERA was done in 126 ears. Of 134 ears, 46 (34.3%) ears had normal hearing, 12 (8.9%) ears with mild hearing loss, 15 (11.4%) ears with moderate hearing loss, 7 (5.2%) ears with severe hearing loss, and 34 (25.3%) ears with profound hearing loss. BERA was not done in 8 (5.9%) ears, as their hearing was already assessed by previous methods and further evaluation by BERA was not indicated [Figure 13].

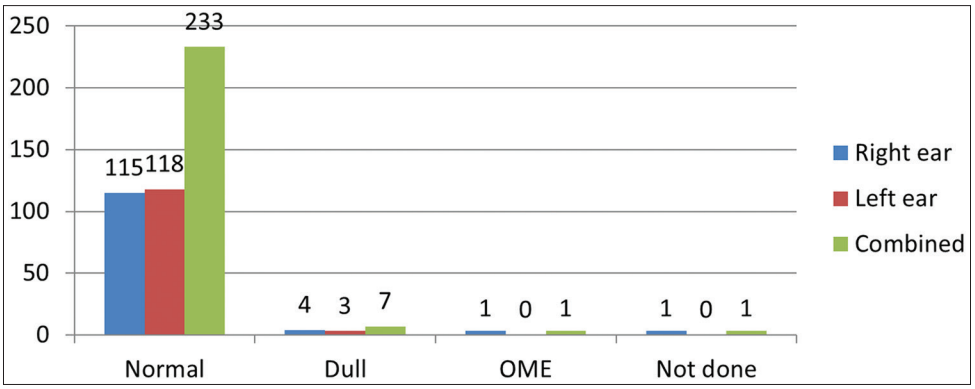

Figure 7: Otomicroscopy finding

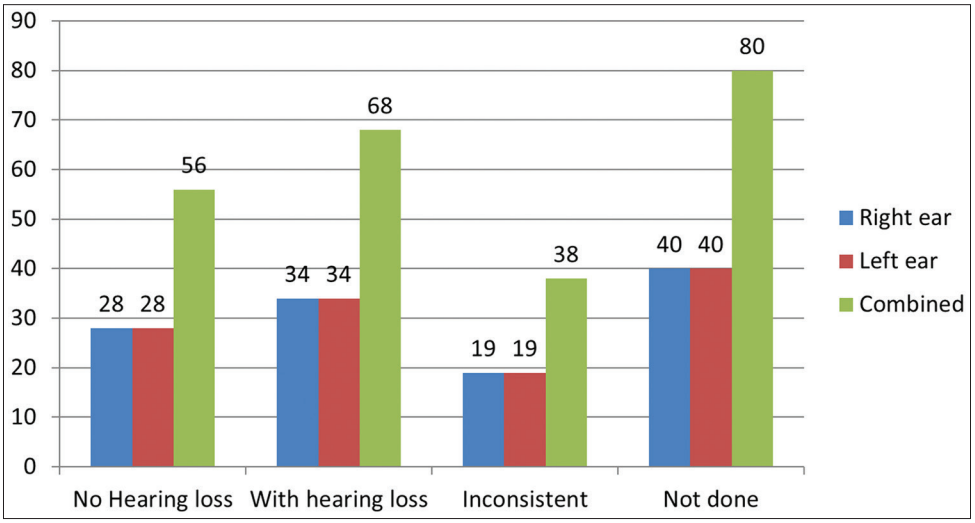

Figure 8: Behavioral observation audiometry hearing level distribution

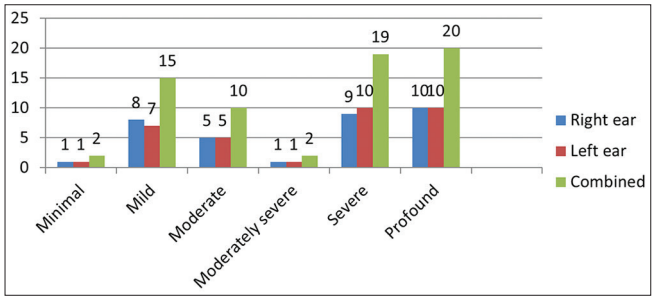

Figure 9: Degree of hearing loss distribution

DISCUSSION

The cross-sectional observational study was done in tertiary care center from October 2017 to April 2018 to find the clinical and audiological profile of children with global developmental delay attending to otorhinolaryngology OPD after obtaining informed consent. Children were recruited in this study according to the inclusion and exclusion criteria of the study. Various studies have attempted to understand the association between CP, delayed development, and hearing loss; mainly, the data come from developed countries.[16] Among those studies, a majority of the studies are retrospective studies. Moreover, the focus seems to be on CP a subset of global developmental delay but is limited with lack of detailed information regarding the audiological status of the children and the objective way of assessing hearing. The paucity of studies on hearing assessment in global developmental delay children probably reflects that the concept of audiological profile of children with global developmental delay has not yet been understood well. Our study addressed the hearing in children diagnosed with global developmental delay. Our aim was to determine the pattern and frequency of hearing loss in a cohort of children with confirmed diagnosis of global developmental delay. Children with global developmental delay require complex, individualized therapy to maximize their long-term quality of life. If hearing loss is undetected early in developmental stage, it affects the speech and language and adds on to delay. Moreover, speech delay sometimes is considered as part of developmental delay, ignoring the need for hearing assessment and rehabilitation. Children with global developmental delay usually report to

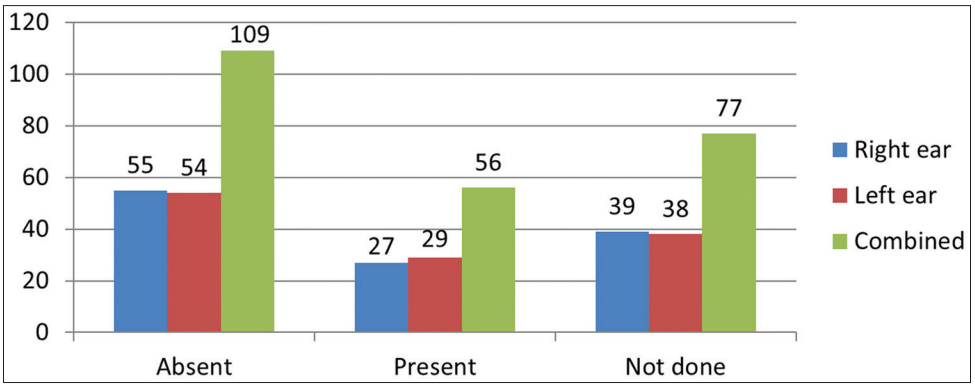

Figure 10: Otoacoustic emission

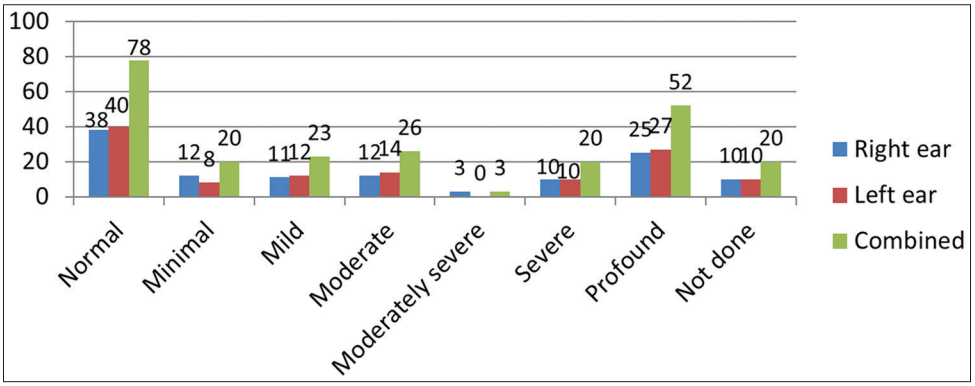

Figure 11: Brain stem evoked response audiometry degree of hearing loss

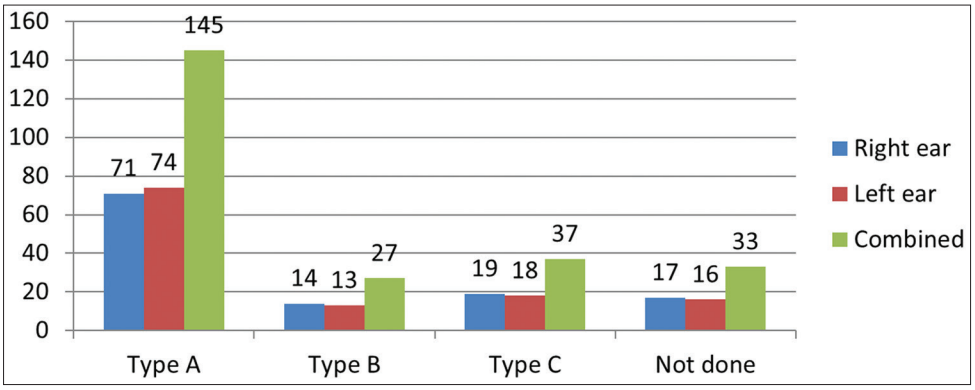

Figure 12: Tympanometry type distribution

developmental pediatrics outpatient clinic for intervention of delay in achieving developmental milestones, either in the motor domain or with problems in feeding. Speech delay is a common problem in these children which is often attributed to the intellectual delay and other oromotor problems. Hearing loss being a potential cause for speech delay is often overlooked in these groups of children as the parents misread the responses of the child to think that he has good hearing. When a general pediatrician consult is sort for speech delay, hearing tests are rarely suggested. Tests with very low sensitivity such as clap test are done by the general pediatrician who can rarely tell if the child is hearing or is the response due to visual or tactile stimuli. Hence, it is very essential to educate the pediatrician looking after the child that hearing assessment is a vital tool in the diagnostic armament of evaluating the child, especially, with the suspected delay in development. For the young child, the term global developmental delay has emerged to describe a disturbance across a variety of developmental domains and such a child has limitations or delay in the acquisition of developmental and functional skills that are both observable and measurable within the context of the natural progression of infants and young children. The latest consensus definition used by the American Academy of Neurology practice parameter statement defines global developmental delay operationally, as a significant delay in

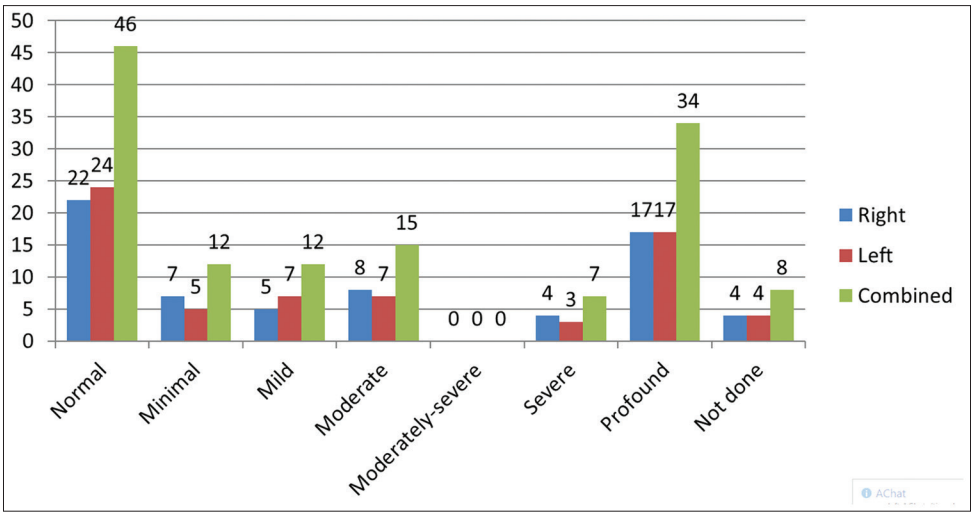

Figure 13: Brain stem evoked response audiometry results in cerebral palsy

two or more developmental domains (e.g., gross/fine motor, cognitive, speech/language, personal/social, and activities of daily living). Typically, if there is a delay in two domains, this implies a delay across all domains.[26,27] In our study, we found that all the children with global developmental delay were referred from developmental pediatric clinic for speech delay and only 36 (29%) children were suspected to have hearing loss by the parents; the result is similar to the study done by Jane and Tipayno. According to this retrospective study, 1023 children were referred for BERA testing by various departments and when analyzed for the reason for the referral; it was found that speech delay was the most common cause for referral.[28] There was a male preponderance in the study group with 72 (59%) males and 49 (40%) females. A study done by Coleen *et al.* also showed a male preponderance in their study group.[29] In our study, the children were grouped into two categories according to the age, first group of children <5 years and the second group of children of 5 years of age or more. Majority (77%) of the study population were in the under-five age group. This can be attributed to the obvious delay in milestones of the child which compels the parents or caregivers to seek medical help early. The mean age of the study population was 3.2 years. The youngest child in the study was 6 months and the oldest child 14 years. The mean age of 3.2 years in our study was delayed compared to studies done in developed countries. Moeller in their study commented that the mean age of identification of hearing impairment and speech delay was 18 months of age.[19] The studies from the developed countries show early diagnosis and rehabilitation probably due to the support offered by the health-care system. The youngest child in our study group was 6 months of age and was referred from developmental pediatrics after the request of principal investigator for audiological objective testing. The child showed an early sign of motor delay by not attending neck holding at 6 months of age and hence included in the study. However, this is not a standard practice of referral to otorhinolaryngologist for global developmental delay at first visit, as usual practice is to identify hearing status by startle reflex and clapping test. In a study done by Coplan, it was pointed out that, in those children with mental retardation and developmental disabilities, hearing loss was diagnosed late as 11–17 years.[30] Hearing loss was not considered during the initial assessment for speech delay similar to our study there was a child with global developmental delay and speech delay but underwent hearing tests for the first time at the age of 14 years. In our , we found that, of 121 global developmental delay children studied, majority of them are with CP 67 (55%) and one-fourth had syndromic association; the most common is Down syndrome. 10 (8.2%) children had autistic spectrum disorder. These findings were similar to previous studies done elsewhere; the major cause of global developmental delay was CP and Down syndrome.[31,32] The most common type of CP was spastic type in this study, which was similar to studies done by Sankar and Mundkur and Singhi and Saini.[33,34] Otomicroscopy examination in our study revealed normal tympanic membrane in 233 examined ears (96.2%). Other findings included microtia with congenital aural atresia in one ear, dull tympanic membrane in seven ears, and OME in one ear which were confirmed by tympanometry. There were no features of cholesteatoma in any of the ears. Ho and Keller reported similar results in a retrospective series of 15 children with global developmental delay.[35] In this study, children with global developmental delay underwent hearing assessment by BOA, OAE, BERA, and tympanometry. Behavioral observational audiometry was not conclusive in this cohort of children because many children had motor impairment, cognitive impairment, and instinctual and/or learning

disabilities. In this study BOA was done 124 ears (51%), others were inconsistent and could not be done in 118 (48%) ears. Of 242 ears, BOA shows that 56 (23%) had normal hearing and 68 (28%) ears had hearing loss. OAE were absent in 109 (45%) ears, present in 56 (23%), and could not be done in 77 (31%) ears. BERA could not be done in 20 examined ears (8%). BERA showed that 78 ears (32%) had normal hearing and 144 ears (59.5%) had hearing loss. In our study 67 children with global developmental delay were associated with CP and the majority were spastic type, BERA was done in 63 children (126) ears and could not be done in 4 children (8) ears. We studied the finding of BERA in this subset of CP children with global developmental delay. 46 (36%) ears had normal hearing, and 80 (63%) ears had hearing loss. Various studies have been reported 30–40% prevalence of hearing loss in CP children. Our study showed 63% prevalence of hearing loss in CP children.[16] Among the children with CP with hearing loss, 34 (26%) ears had profound hearing loss, and the result was more than Dufresne *et al.* study which showed 12.7% sensorineural hearing impairment.[36] In our study children who attended otorhinolaryngology OPD with global developmental delay and speech delay, only 36 (29%) children had a suspicion of hearing loss. On objective hearing testing, we identified that 144 (59.5%) ears had hearing loss. A long-term prospective study with comprehensive data would be more accurate and reliable for the recommendation for objective hearing test in global developmental delay so that early identification of hearing loss can be done before the children present with speech delay and developmental delay.

Limitations

The major limitation of this cross-sectional observational study was that it looked at a point estimate of hearing in children with global developmental delay. A cohort of children followed up for a short period would have given a better estimate of the problem. A second limitation of the study is that it is a hospital-based study and not a community-based study excluding children whose caregivers who did not seek medical advice for their children.

CONCLUSIONS

The mean age of referral was 3.2 years in global developmental delay children who were referred for the evaluation of speech delay. Among the 121 global developmental delay children included in the study, 36 (29%) had hearing loss with speech delay. Our study detected a higher incidence of undetected hearing loss (144 ears, 59.5%) in children with global developmental delay. On the basis of this finding, parents, family physician, pediatrician, and otorhinolaryngology should work as a team. Our study recommends that every child of global developmental delay should have a detailed audiological workup to identify hearing loss and follow-up for the early identification of middle ear pathology so that timely intervention can be taken.

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