Initial Symptom of Different Developmental Difficulties: Language Delay

Farklı Gelişimsel Problemlerin İlk Bulgusu: Konuşma Gecikmesi

Pelin ÇELİK¹, İclal AYRANCI SUCAKLI¹, Halil İbrahim YAKUT²

² Department of Pediatrics, University of Health Sciences, Ankara Child Health and Diseases Hematology and Oncology Training and Research Hospital, Ankara, Turkey



ABSTRACT

Objective: The aim is to evaluate sociodemographic characteristics, differential diagnoses and risk factors of children with language delay.

Material and Methods: This retrospective study was included 222 children with language delay referred to a tertiary developmental-behavioral pediatrics clinic. Children were evaluated with the Bayley Scales of Infant and Toddler Development 2nd Edition and Guide for Monitoring and Child Development.

Results: Most of families (78%) firstly admitted to pediatricians. Only 38.7% of the children had expressive language disorder, while 39.6% had cognitive delay and 14% had autism spectrum disorder (ASD). 56.8% of the children did not have book. Book ownership decreased with increasing sibling count and lower parents educational level. Kindergarten/ pre-school education was in only 3.2% of the children. 15.8% of the children did not have any opportunity for peer relationship. Most of the children (82.4%) had daily screen time above 4 hours. Mental Developmental Index scores were lower in children with lack of books, and parents with ≤8 years of education.

Conclusion: Serious neurodevelopmental problems including cognitive delay and ASD should be considered in children with language delay. Pediatricians play a central role in early diagnosis, appropriate management and should guide the families about protective factors that affect language development.

Key Words: Autism spectrum disorder, Cognitive delay, Early diagnosis, Language delay, Risk factors

ÖΖ

Amaç: Bu çalışmada konuşma gecikmesi yakınması ile başvuran çocukların sosyodemografik özelliklerinin, ayırıcı tanılarının ve konuşma gecikmesi ile ilişkili risk faktörlerinin incelenmesi amaçlandı.

Gereç ve Yöntemler: Gelişimsel pediatri polikliniğine "konuşma gecikmesi" yakınması ile başvuran 222 çocuğun tıbbi kayıtları retrospektif olarak gözden geçirildi. Çocuklar Gelişimi İzleme ve Destekleme Rehberi (GİDR) ve Bayley Bebekler İçin Gelişimsel Değerlendirme Ölçeği II kullanılarak değerlendirildi.

Bulgular: Çocukların %78'i ilk olarak çocuk sağlığı ve hastalıkları uzmanlarına başvurmuşlardı. Olguların sadece %38.7'sinde ifade edici dil gelişiminde izole gecikme saptanırken, %39.6'isında bilişsel gecikme, %14'ünde otizm spektrum bozukluğu saptandı. Çocukların % 56.8'inin evde kendisine ait kitabı yoktu. Kardeş sayısı arttıkça ve ebeveyn eğitim düzeyi düştükçe kitaba sahip olma oranının düştüğü görüldü (p<0.05). Çocukların sadece %3.2'si okul öncesi eğitim alıyordu, %15.8'ünün oyun alanı/park gibi yaştı ilişkilerinin sağlanacağı ortamlarda bulunma fırsatının olmadığı saptandı. Çocukların çoğunluğunun (% 82.4) ekran maruziyeti günde 4 saat ya da daha fazlaydı. Evde kendisine ait kitabı olmayan ve ebeveyn eğitim süresi 8 yıl ve altında olan çocukların MDI puanları daha düşük saptandı (p<0.05).

Sonuç: Konuşma gecikmesi ile başvuran olgularda bilişsel gecikme, otizm spektrum bozukluğu gibi ciddi nörogelişimsel sorunların da göz önüne alınması gerekmektedir. Çocuk sağlığı ve hastalıkları uzmanları dil ve konuşma alanındaki gecikmelerin erken saptanmasında, uygun yönlendirilmesinde ve dil gelişimini etkileyen değiştirilebilir koruyucu faktörler hakkında ailelere rehberlik etmede önemli role sahiptirler.

Anahtar Kelimeler: Otizm spektrum bozukluğu, Bilişsel gecikme, Erken tanı, Konuşma gecikmesi, Risk faktörleri

Correspondence Address:/ Yazışma Adresi:

¹ Division of Developmental-Behavioral Pediatrics, Department of Pediatrics, University of Health Sciences, Ankara Child Health and Diseases Hematology and Oncology Training and Research Hospital, Ankara, Turkey

INTRODUCTION

Language is a symbolic and structured communication system which allows people to share their ideas, thoughts, emotions, and beliefs. Receptive language is referred to the ability to perceive and understand words, sentences, and dialogs, whereas expressive language is referred as the ability to produce words, sentences, and dialogs. The development of language and communication skills is important as it has a key role to learning and social skills. Speech and language delays/disorders are among the primary referral reasons for comprehensive developmental evaluation and account for approximately 40% of the cases (1). The prevalence has been reported between 5% and 12% in children 2 to 5 years of age (2).

Detailed evaluation of children with language delay is important for differential diagnosis and early and appropriate intervention as well as prognosis.

In this study, we aimed to evaluate sociodemographic characteristics, risk factors, and differential diagnosis of children referred to a tertiary developmental and behavioral pediatrics clinic with the chief complaint of language delay.

MATERIALS and METHODS

This retrospective study was conducted in Ankara Child Health and Diseases Hematology and Oncology Training and Research Hospital and ethical committee of approval was obtained from Ethical Committee of same hospital.

Children ≤42 months referred to the developmental-behavioral pediatrics clinic because of "language delay", "inability to speak", "delay in language", and "inability to form sentences" between 1 January 2017–1 September 2018 were included. Children >42 months age, children with previously known neurological, genetic and metabolic disesases; children with speech sound disorder only, and non native Turkish speaking children were excluded.

Medical records were retrospectively reviewed. Sociodemographic data, linguistic environment (presence of books, daily screen time, frequency of participating in an environment where allows peer relationship, preschool education, maternal depression), developmental evaluation results, and suggested early intervention services were recorded.

The developmental evaluation was conducted based on familycentered holistic developmental evaluation principles. Both The Guide for Monitoring Child Development (GMCD), and Bayley Scales of Infant and Toddler Development 2nd edition (BSID-II) were used for all children for developmental evaluation (3, 4). Firstly, the GMCD were administered. The GMCD, produced in Turkey by Ertem et al. (5), provides a method for developmental monitoring and early detection of developmental difficulties in low and middle income countries. The GMCD is a brief, openended, precoded interview with the primary caregiver. The first question is to find out parental concerns. Next, the clinician asks the 6 open-ended questions about following developmental domains: expressive language and communication, receptive language, gross and fine motor, relationship (social-emotional), play, and self-help skills (for children older than 12 months). Caregiver's spontaneous responses to the open-ended questions are applied to the milestones whenever possible.

After evaluating GMCD, BSID-II was administered. BSID-II is comprised of two scales, the Mental Developmental Index (MDI) and the Psychomotor Developmental Index (PDI). MDI measures the combination of the nonverbal cognitive and language skills, and does not assess cognitive, expressive and receptive language subscale separately. So we used both of BSID-II and GMCD together. During evaluation, children were observed in terms of their free play, relationship and interaction additionally.

Receptive language, expressive language, play and self-help skills were evaluated by the GMCD and cognitive and motor development were evaluated by BSID-II. Delay in receptive language, expressive language, play and self-help skills were defined as two standard deviations or more below in the relevant area according to GMCD. Speech sound disorder and autism spectrum disorder (ASD) diagnoses were performed according to diagnostic criteria of DSM-V. Children who had difficulties in the acquisition and use of language (putting words and sentences together to express thought and ideas, recalling words) across modalities (spoken and sign language or other) and the difficulties which were not attributable to any sensory impairment, neurological or medical condition or cognitive impairment were defined as expressive language disorder. Cognitive and motor delay were defined as <70 points according to MDI and PDI.

Children were categorised into four groups: Group 1: age-appropriate development, Group 2: expressive language disorder, Group 3: cognitive impairment with/without motor delay, Group 4: autism spectrum disorder.

Maternal depression was assessed using the RAND 3-question screen (6).

Statistical Analysis

Statistical analyses were performed using the SPSS statistical package for MAC (v. 20.0). Categorical variables between groups were analyzed using the chi-squared test. Comparison of mean between two groups was examined using a t -test where the data fit a normal distribution, and the Mann–Whitney U test where the data was non-normal. For comparisons of more

than two groups, ANOVA was used for normal distributions, and the Kruskal–Wallis test for non-normal distributions. A p value of 0.05 was deemed to indicate statistical significance.

RESULTS

222 children (female: n=65, 29.3% and male: n=157, 70.7%) were included. The median age was 30 (13-42) months. The median age of parents was 27 (15-42) years for mothers and 31 (18-48) years for fathers. 95 (42.8%) of mothers and 117 (52.7%) of fathers had completed >8 years of education. The employment rate was 17.1% among the mothers and 97.3% among the fathers. Consanguineous marriage rate was 22.2%. 12.7% of the cases had premature birth history and 16.6% had hospitalization in neonatal period, while the most common diagnosis was indirect hyperbilirubinemia (6.3%).

78% of the children were referred by general pediatrician and pediatric subspecialists, whereas 12% of the children were directly admitted to our clinic. At admission, 23 (10.4%) of the children could not produce any meaningful word and 124 of 186 children (66.7%) aged 24 months or older could not form two-word sentences. Table I shows developmental evaluation results as well as diagnoses, clinical features, and demographic characteristics. Only 7.7% of the children were found to have

age-appropriate development (group 1), while 38.7% had expressive language disorder (group 2), 39.6% had cognitive delay (group 3), and 14% had ASD (group 4). Additional diagnoses were speech sound disorder (26.7% of the cases in group 2) and motor developmental delay (11.4% of the cases in group 3). Majority of the children aged 24 months or older in group 3 (84.4%) and group 4 (88.9%) could not form twoword sentences. Receptive language delay was found in 61.3% of the children in group 3 and 90.3% of the children in group 4. According to the results of the BSID-II, group 3 and 4 had lower MDI scores than group 1 and 2, while group 3 had lower PDI scores than group 1 and 2 (p < 0.05). The rate of book ownership and having parents with >8 years of education were lower in group 3 (p<0.05). No difference was found between the groups in terms of sex, gestational week, and maternal depression.

Co-morbid disorders known at the time of the initial presentation were asthma (7.8%), hypothyroidism (1.2%), and food allergy (1.6%). During the evaluation iron deficiency and iron deficiency anemia were discovered 28% and 10.4% of the cases. These children were given the necessary treatment. Conductive hearing loss was obtained in 3 (1.3%). Genetic and metabolic evaluation was performed in 48 children and no one revealed a genetic or metabolic disorder.

Table 1. The family-centered holistic developmental evaluation results, diagnoses, clinical reacties, and demographic characteristics.						
	Age appropriate development (Group 1)	Expressive language disorder (Group 2)	Cognitive impairment with/without motor delay (Group 3)	Autism Spectrum Disorder (Group 4)	Toplam	р
n(%)	17 (7.7%)	86 (38.7%)	88 (39.6%)	31 (14%)	222 (100%)	
Male/Female	10/7	59/27	62/26	26/5	157/65	p>0.05
GMCD results						
Expressive language delay n(%) Receptive language delay n(%) Delay in self help skills n(%) Delay in play skills n(%)	0 (0%) 0 (0%) 0 (0%) 0 (0%)	86 (100%) 1 (1.2%) 1 (1.2%) 3 (3.5%)	88 (100%) 54 (61.3%) 22 (25%) 24 (27.3%)	31 (100%) 28 (90.3%) 13 (41.9%) 26 (83.9%)	205 (92.3%) 83 (37.3%) 36 (16.2%) 53 (23.9%)	* * *
BSID-II results						
MDI Mean±SD Cognitive delay (MDI <70 points) n(%) PDI Mean±SD Motor delay (PDI <70 points) n(%)	87±8.1 0 (0%) 89.1±9.2 0 (0%)	80.6±8.7 0 (0%) 83.2±10.6 0 (0%)	54.3±6.7 88 (100%) 64.9±17.4 10 (11.4%)	52.2±8.9 29 (93.5%) 86±11.3 0 (0%)	68.2±15.9 117 (52.7%) 76.1±17 10 (4.5%)	p<0.05 ° * p<0.05 ° *
Speech sound disorder n(%)	0 (0%)	23 (26.7%)	7 (8%)	0 (0%)	30 (13.5%)	*
Book ownership n(%)	9 (52.9%)	45 (52.9%)	26 (29.9%)	15 (48.4%)	95 (43.2%)	p<0.05 °
Form two-word sentences in children aged 24 months or older n(%)	6 (100%)	41 (53.9%)	12 (15.6%)	3 (11.1%)	62 (33.3%)	p<0.05 d
Mother's education >8 years	11 (64.7%)	42 (48.8%)	25 (26.3%)	17 (54.8%)	95 (42.8%)	p<0.05°
Father's education >8 years	10 (58.8%)	54 (62.8%)	34 (38.6%)	19 (61.3%)	117 (52.7%)	p<0.05 f
Gestational week, median (min-max)	40 (29-40)	40 (32-43)	40 (31-42)	40 (36-41)	40 (29-43)	p>0.05
Maternal depression n(%)	1 (5.8%)	14 (16.2%)	15 (17%)	8 (25.8%)	38 (17.1%)	p>0.05

Table I: The family-centered holistic developmental evaluation results, diagnoses, clinical features, and demographic characteristics.

a: Group 1 vs 2, Group 1 vs 3, Group 1 vs 4, Group 2 vs 3, Group 2 vs 4, b: Group 1 vs 3, Group 2 vs 3, c: Group 2 vs 3, d: Group 1 vs 2, Group 1 vs 3, Group 1 vs 3, Group 1 vs 4, Group 2 vs 3, Group 2 vs 4, Group 2 vs 4, Group 3 vs 4, e: Group 1 vs 3, Group 2 vs 3, Group 3 vs 4, f: Group 2 vs 3, *Statistical analysis could not be performed because of insufficient patient number of some groups

43.2% of the families reported that they had books for reading their children. The rate of book ownership decreased from 63.2% to 34.4% as the number of children in the family increased from one to three or more (p<0.05). The children whose mother or father had completed >8 years of education had a higher rate of book ownership (p<0.05, 64.2% and 57.8%, respectively). Only 7 (3.2%) children were attending kindergarten/pre-school. 15.8% of the children did not engage in environments where allows peer relationship such as playgrounds or parks, while 35.6% engaged only once or twice a week. 17.1% of the mothers described ongoing major depression symptoms and were referred to an adult psychiatry specialist.

The children whose parents had completed >8 years of education had higher MDI scores than those whose parents had completed \leq 8 years of education (mother's education >8 years vs \leq 8 years: 71.8±16.6 vs 65.6±15, p<0.05; father's education >8 years vs \leq 8 years: 71.9±16.2 vs 64.4±14.8, p<0.05). There was no relation between PDI scores and parents' educational level.

MDI scores of children who had books were higher than those who had not (71 \pm 16.4 vs 66.3 \pm 15.4, p<0.05). Daily screen time less than 1 hour, 1-2 hours, 2-4 hours and more than 4 hours were in 3.9%, 7.8%, 5.9% and 82.4% of the children, respectively.

According to comprehensive developmental evaluation, 25 (11.2%) of the children were given suggestions to facilitate language learning in their home environment, while 57 (25.7%) of the children were recommended to attend kindergarten/ pre-school education in addition to the home environment suggestions. Early intervention services were suggested to 150 (67.6%) of the children, including language and speech therapy (n=34, 15.3%), special education (n=32, 14.4%), special education in addition to language and speech therapy (n=74, 33.4%) and physiotherapy (n=10, 4.5%).

DISCUSSION

Our study has demonstrated that serious neurodevelopmental issues such as cognitive delay and ASD should be considered and differential diagnosis should be made in children presenting with language delay. Only 38.7% of the children with language delay were found to have expressive language disorder which has an overall good prognosis.

There is a strong correlation between development of language skills and cognitive skills. Studies reported that 6-37% of children with delay in language development had also cognitive delay (7-10). In our study, 39.6% of the children with language delay had cognitive delay as well. We believe that our high cognitive delay rate might be related with the fact that the study was conducted in a tertiary developmental-behavioral

pediatrics clinic; most of children were referred by pediatricians and also had psychosocial risks.

Receptive language score was shown to be positively correlated with mean cognitive score and predict cognitive performance (11). It was reported that children with a receptive language score of >1 standard deviation below the mean were under the risk of comorbid cognitive delay (11). Children with expressive language disorder accompanied by receptive language delay at the age of 3 accounted for 84% of the cases with cognitive impairment at the age of 5 (8). It is known that impairment in receptive language skills is more common than impairment in expressive language skills among children with ASD (12). In our study, the prevalence of delay in receptive language development was 61.3% and 90.3% in the group 3 and the group 4, respectively. For this reason, a differential diagnosis of cognitive delay or ASD is necessary in children with language delay who cannot process age-appropriate commands without visual cues.

Several studies reported that boys are at higher risk of language delay than girls and language delay occurs 3 times more commonly in boys (13). The majority (70.7%) of the cases were boys in our study as well.

Autism and Developmental Disabilities Monitoring Network of the Centers for Disease Control and Prevention, estimated the prevalence of ASD as 1 in every 59 (1.7%) children (14). In a study from Turkey, 15% of the cases with language delay were found to have ASD (10). A similarly high prevalence (14%) was found in our study as well. Since language delay is a typical characteristic of ASD, it is not surprising to find a significantly higher prevalence in a selected group of children with language delay compared to the general population. For this reason, ASD must certainly be assessed in differential diagnosis of children with language delay. CDC reports that ASD is 4 times more common among males. Similarly, we found that ASD was 5 times more common among boys.

Language-based social interaction environments such as reciprocal interaction with a parent or caregiver, informal play opportunities with their peers and kindergarden/pre-school education are among the protective factors associated with language delay (13, 15). In our study, more than half of the children did not have opportunity to engage in environments such as playgrounds or park where allows peer relationship more than once or twice a week. Only 3.2% of the cases were attending kindergarten/pre-school. This was because the families could not afford a kindergarten due to financial difficulties and dwell in areas where free play and peer interaction opportunities are not available.

Another protective factor for language delay is reading and sharing books to children on a daily basis (13). Reading aloud to kids starting from an early age led to better pre-school language skills, higher interest in reading, and increased interaction between the adult and the child (13). The Reach Out and Read model, one of the most studied and wide-spread programs that promote reading, was found to change parental attitudes toward reading aloud, increase frequency of parents' reading to their children on a regular basis and have better child language development (16). 2011-2012 National Survey of Children's Health showed that 60% of the families with higher income level and only 34% of the families with lower income level read to their children on a daily basis (17). In our study only 43.2% of the children had books. This might be related with the low socioeconomic level of the families. A significant positive relation was found between having completed >8 years of education and book ownership. Also, a negative relation was found between the number of siblings and book ownership. This might be related with the limited time left for reading with the increasing number of children and the division of economic resources of the family.

It was reported that children of families with low educational level had language problems more commonly (15). In our study, 42.8% of the mothers and 52.7% of the fathers had completed >8 years of education. In a study from Turkey, the mother's education was the most significant controllable environmental factor affecting language development (18). The education level and the vocabulary size of the mother are known to be strong determinants for language and cognitive development of young children (19). It was found that fathers with a higher level of education interacted more sensitively with their children, developed more positive relationships with their spouses, thus allowing for a higher-quality relationship between the mother and the child, and children of such fathers had higher MDI scores (20). In line with the literature, the educational level of the parents was significantly lower in group 3, which involved children with cognitive delay. It should be noted that there may be an indirect hereditary effect if the parent has an unresolved language impairment that interfered with their own educational advancement. We couldn't evaluate this condition because of retrospective design of our study.

The American Academy of Pediatrics recommends avoiding digital media use for children below 18-24 months and limiting screen exposure to 1 hour per day between the ages of 2-5 years (21). In our study, only 3.9% of the cases had a screen exposure of 1 hour or less per day as recommended, while the majority (82.4%) of the cases had a screen exposure of more than 4 hours per day. Various studies showed that excessive screen exposure in early childhood was correlated with language delay as well as cognitive delay and social/emotional delay (22-24).

Maternal depression prevelance is 20% in low-and middleincome countries (25). Women with maternal depression have weaker attachment, spend less time playing with their children, use less facial expressions, and engage in verbal communication less frequently. Maternal depression was reported to effect children's language and cognitive development negatively (26). Maternal depression prevalence was found in line with the literature (17.1%) in our study. Mothers possible had worries about their children's developmental delay, and increased stress due to concerns regarding language delay.

In this study, 78% of the children were referred by either a general pediatrician or a pediatric subspecialist. In other words, families whose children have language delay firstly admitted to pediatricians. Some physicians wait until 3 years of age for evaluation and treatment of patients with language delay in early childhood, which may cause delays in diagnosis and early intervention (9). Also, traditional beliefs in our country such as "his brother/uncle started to speak later, too", "boys start to speak later", or "he will speak eventually" may lead to delayed admission to a physician as well. It is known that children with isolated expressive language disorder at the age of 2 years may still not achieve the same performance with their peers at the age of 3 or 4 years (27). When the language delay is first detected, it is impossible to accurately predict whether the child will be able to catch up to their typically developing peers without early support services or continue to have language problems (28). Delays in language development at an early age may indicate learning difficulties at later stages of life (29). Language and speech delay in early childhood is associated with increased reading, writing, attention and behavioral problems (30). Language and speech therapy was shown to lead to increased lexical repertoire, vocabulary size, length of utterances, intelligibility, socialization skills, and decreased parental stress (31). Also, rather than an expressive language disorder which has an overall good prognosis, language delay may be the initial presenting symptom of more serious neurodevelopmental problems such as ASD or cognitive delay. Given the critical importance of brain development in early childhood and benefits of early intervention on affecting neural pathways, young children with suspected developmental delays should be diagnosed as early as possible so that they receive early support services on time and appropriately (32). For this reason, pediatricians play a central role in early diagnosis and appropriate management of language delays. Risk factors affecting language development should be reviewed. It should be kept in mind that language delay might be the first sign of different developmental problems such as cognitive delay and ASD. Therefore, all developmental domains should be evaluated with standardized tools in these children. If the child has normal hearing, age appropriate development in terms of relating and receptive language skills, parents should be adviced to improve language development such as reciprocal interaction, increasing the number of books in the home, reading to their children, decreasing "screen time" exposure, increasing opportunities for children to interact and play with their peers, early attendance to kindergarten/pre-school. Even after promoting linguistic environment, children with no

language improvement, suspicion of ASD and cognitive delay should be referred for further evaluation promptly.

The limitations of our study include the lack of a control group, the low socio-economic level of the study group, and the lack of long-term follow-up results. Another limitation of the study is that the children in the sample were more likely to have more serious developmental delays since they referred to the subspeciality clinic; therefore, the results of the study cannot be generalized to the society. Consanguinity rate was 22.2% and mostly second degree. Heredity is most common etiologic factor noted in children with expressive language disorder. However, we couldn't determine hereditary factors because retrospective design of the study.

Nonetheless, the strengths of our study are, large sample size and evaluation of all developmental domains such as language, cognition, motor, relating, play, and self-help by family-centered holistic evaluation-based principles using both the BSID-II and GMCD.

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