RESEARCH ARTICLE

Intellectual disability among special children and its associated factors: A case control study, Lahore Pakistan

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Abstract

Objective: To study the association of maternal, paternal and social factors with intellectual disability in special children.

Methods: The case-control study was conducted in four schools of Lahore, Pakistan, from September 2014 to September 2015, and comprised intellectually disabled children aged 6-15 years, and an equal number of matching healthy subjects. group. Interviews were conducted with the help of a pre-tested structured questionnaire.
SPSS -17 was utilised to analyse the data.

Results: Of the 298 participants, 149(50%) each were cases and controls. Overall, there were 83(56%) boys and 66(44%) girls. Significant association of intellectual disability was found with consanguinity (p=0.001), father's educational status (p=0.03), paternal history of mental retardation (p=0.01) and history of delayed cry (p=0.001). Breastfeeding (depicted a protective relationship (p=0.03).

Conclusion: Parental, social, environmental and familial causes contributed to intellectual disability among the subjects.

Keywords: Intellectual disability, Factors, Advanced maternal age, Consanguinity, Asphyxia, Trauma. (JPMA 69: 684; 2019)

Introduction

Intellectual disability (ID) is defined as a significant limitation in intellectual functioning and adaptive behaviour covering many everyday social and practical skills, originating before the age of 18 years.[1] Intellectual functioning refers to general mental capacity, learning, reasoning, problem-solving measured by an intelligence quotient (IQ) score. A score of around 70-75 indicates a limitation in intellectual functioning that may include children of Down's syndrome, autism, mental retardation etc.[1] ID, a stigmatising condition of mental origin, not only affects the sufferer but has major implications on the family, society and country as a whole.[2] Global prevalence of ID is 1.04%[3] and is predicted to increase by 15% till 2020.[4] In Pakistan, estimated ID prevalence is 17% with an 8% contribution of mental retardation.[5] Severe mental retardation is quoted to be 1.9%, mild 6.5%, and is affected by various socioeconomic statuses and geographical distributions.[6]

ID faces the dilemma that among the cases so diagnosed, causes can be identified only in 25%.[7] These can be attributed to autosomal recessive chromosomal disorders.[8] Genetic, acquired, socio-demographic and cultural factors are also highly associated with ID.[9] Consanguinity is associated predominantly with undiagnosed cases.[10] Prenatal, perinatal causes in mothers and perinates like low birth weight (LBW), asphyxia, delayed cry, anoxia, trauma and kernicterus at the time of birth and postnatal causes in infants have a considerable share as well.[10] Early initiation, exclusivity and duration of breastfeeding have an impeccable impact on the IQ scores and academic performance of children.[11]

The most authentic and standardised method to measure intelligence globally and for research is Wechsler intelligence scale for children (WISC) aged 6 -16 years.[12] It is unique as it focuses on conduction of multiple numbers of tests measuring the quality via performance rather than quantity of intelligence.[12] Scoring criteria is based on performance and verbal questions. Highest range is 130 and above; gifted, 120-129; high intelligence, 110-119; normal intelligence, 90-109; average intelligence 70-89. Anything 70 and below indicates borderline intelligence or ID.[13]

Public health can play a vital role in reducing the impact of ID by applying levels of prevention, essentially tertiary prevention, by advocating development of special centres and rehabilitation to improve both physical and mental functioning of the disabled.[14] Most studies have been conducted in the Western world, and there is a need to determine the association of geographic, demographic and cultural factors in countries like Pakistan as these factors are modifiable and can be targeted in prevention

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programmes to reduce the burden of this disability. Role of perinatal factors needs to be highlighted. Scarcity of data, especially in our part of the world, needs to be addressed so as to promote evidence-based practices. The current study was planned to determine the association of maternal, paternal and social factors with ID in Pakistani special children.

Subjects and Methods

The case-control study was conducted at four schools in Lahore, Pakistan, from September 2014 to September 2015. The sample size was calculated while keeping significance level at 95%, prevalence at 50% (due to unavailability of data), power of the study at 80% and considering $p<0.05$ significant.[15]

Intellectually disabled children aged 6-15 years suffering from Down's syndrome, autism and mental retardation with an IQ score less than 70 constituted the cases. Children without intellectual disability securing good academic grades (>75%) constituted the controls.[16]

Cases were taken from the Rising Sun Institute and the National Special Education Centre, Lahore. The total number of students at both institutes was 900. A list of students suffering from mental retardation, diagnosed with the help of WISC-IV, autism, diagnosed with the help of Diagnostic and Statistical Manual of Mental Disorders (DSM) IV,[17] and Down's syndrome, diagnosed with the help of distinctive facial and physical features, and having an IQ score of 70 or less determined with the help of WISC-IV was prepared, and cases were raised from within this pool A list of all the public schools of Lahore was prepared and they were visited. A second list of schools giving permission to interview the parents was prepared. By simple random sampling technique, Latif Academy High School and Naveed Foundation were selected. Students securing good grades (>75%) from classes 1-10 were identified, 15 students from each class fulfilling the inclusion criteria were selected to raise the control group.

Data was collected by the investigator through personal interviewing of the parents using a pretested structured questionnaire, which was tested before initiating the data-collection on 10 cases and controls and no changes were needed to be incorporated in the questionnaire. About 25-30 minutes were spent in filling up the questionnaires. Matching for age and gender was taken in consideration during the data-collection stage. Records of the institutes visited were also rechecked to reduce the element of recall bias. SPSS 17 was used to analyse the data. Frequencies and percentages for categorical variables were calculated. $P<0.05$ was considered statistically significant. Logistic regression was applied.

Ethical clearance was taken from institutional review board (Fatima Memorial College of Medicine And Dentistry, Lahore, Pakistan). Permission from the respective institutions and consents from the parents of the subjects were taken.

Results

Of the 298 participants, there were 149(50%) each in the cases and control groups. There were 83(56%) boys and 66(44%) girls overall. Among the cases, 109(73%) had consanguinity compared to 89(60%) in the controls. Educational levels varied in mothers and fathers of both the cases and the controls (Table-1).

Majority of the mothers in both groups — 136(91%) cases and 143(95%) in controls — were housewives. Among the cases, 33(22%) fathers were unskilled, while 34(23%) were skilled labourers and professionals. Among the controls, 102(68.8%) fathers belonged to either of these categories. Age of the mother at the time of marriage ranged 14-24 years in 136(92%) cases and 141(95%) controls. Mothers age at the time of the birth of the participant ranged 18-30 years in 117(79%) cases and 121(81%) controls. Besides, 129(87%) cases and 138(93%) controls were born to fathers aged 20-39 years (p=0.08). Overall, 55(37%) mothers in the sample had a parity of more than five. Participants in the order of firstborn were more; 52(34.8%) cases and 48(32.3%) controls compared to second in 48(32%) cases and 42(28%) controls, and third-born 26(18%) and 30(20%) respectively. Further, 113(76%) cases and 123(83%) controls were delivered in

Table-1: Social factors associated with intellectual disability.

Variables	Cases (n=149)		Controls (n=149)		p value
	n	%	n	%	
Economic Status of parents					
Low	42	28	32	22	
Middle	78	52	116	78	<0.001
High	29	19	1	1	
Educational Status of Mother					
Illiterate	23	15	13	9	
Primary - Matriculation	77	52	128	86	<0.001
Graduate and above	49	33	8	6	
Educational Status of Father					
Illiterate	17	11	5	3	
Primary - Matriculation	68	46	109	73	0.001
Graduate and above	64	43	35	24	
Consanguinity					
Yes	109	73	60	40	0.001
No	40	27	89	60	
Relationship to Spouse (first cousin)					
Yes	77	52	37	25	0.001
No	72	48	112	75	

Table-2: Paternal genetic factors associated with intellectual disability.

Variables	Cases (n=149)		Controls (n=149)		Odd's Ratio	95% CI	p value
	n	%	n	%			
Congenital Malformations							
Yes	3	2	1	0.7	3.0		0.31
No	146	98	148	99		0.313-29.57	
Mental Retardation							
Yes	23	16	1	0.7	27.0		<0.001
No	126	85	148	99		3.598-202.88	
Down's Syndrome							
Yes	4	3	0	0	2.0		0.04
No	145	97	149	100		1.806-2.277	

Table-3: Perinatal factors associated with intellectual disability.

Variables	Cases		Controls		Odd's Ratio	95%CI	p value
	n	%	n	%			
Trauma to the participant at the time of birth							
Yes	4	3	0	0	2.03	1.806-2.27	0.04
No	145	97	149	100			
Asphyxia to the participant at the time of birth							
Yes	48	32	6	4	11.33	4.67-27.48	
No	101	68	143	96			<0.001
Delayed cry at the time of participant							
Yes	55	37	3	2	28.48	8.67-93.65	<0.001
No	94	63	146	98			
Use of Oxygen at the time of birth of participant							
Yes	27	18.2	5	3	6.37	2.38-17.05	<0.001
No	122	81.8	144	97			
Breast feeding (participant)							
Yes	115	77.2	134	90	0.38	o.196-0.73	0.003
No	34	22.8	15	10			

Table-4: Regression Analysis: Data was matched for age and gender.

Variables Name	Unadjusted		Adjusted		p value
	OR	Confidence Interval	OR	Confidence Interval	
Asphyxia	11.33	4.67 -- 27.48	0.63	0.11 -- 3.34	0.58
Breastfeeding	0.38	0.20 --0.73	0.40	0.17 -- 0.94	0.03
Consanguinity	4.04	2.48 -- 6.59	4.30	6.69 -- 8.07	<0.001
Delayed Cry	28.48	8.67 -- 93.65	47.31	6.69 -- 334.59	<0.001
Economic status	0.07	0.038 -- 0.116	1.39	0.70 -- 2.75	0.34
Maternal Educational status	1.91	0.93 -- 3.39	0.82	0.57 -- 1.20	0.31
Paternal Educational status	3.71	1.33 -- 10.33	1.46	1.03 -- 2.08	0.03
Paternal history of mental retardation	27.0	3.6 -- 202.9	16.06	1.89 -- 136.33	0.01
Maternal history of mental retardation	2.1	1.83 -- 2.33	539517.0	0.00 --1.0	0.97
Use of oxygen	6.37	2.38 -- 17.05	3.03	0.76 -- 12.09	0.12
Trauma to baby	2.0	1.81 -- 2.27	124527.11	0.00 -- 10	0.98

OR: Odds ratio.

institutions. Normal vaginal delivery was the most common mode of delivery in 94(63%) cases and 110(74%) controls, but caesarean section (CS) was more in cases 52(35%) than controls 32(21%), (p=0.02)

Paternal genetic factors were also noted (Table-2).

Perinatal factors were significantly associated with ID. At the time of birth, trauma (odds ratio [OR] 2.03, 95%

confidence interval [CI] 1.806-2.27, p=0.04), asphyxia (OR 11.33, 95%CI 4.67-27.48, p=0.001), delayed cry (OR 28.48, 95% CI 8.67-93.65, p=0.001), use of oxygen (OR 6.37, 95% CI 2.38-17.05, p=0.001) exhibited significant associations.

The impact breastfeeding was significant (OR 0.38, 95% CI 0.196-0.73, p=0.003), but the duration of breastfeeding had no significant impact (OR 0.53, 95% CI 0.35-0.79, p=0.270) (Table-3).

Logistic regression was applied and after adjusting, only history of consanguinity, delayed cry at the time of birth, and paternal history of mental retardation were found to have significant association with ID, while breastfeeding showed a protective effect in relation to ID (Table-4).

Discussion

Pakistan has been reported as one of the developing countries with the highest possible rate of intellectual disabilities in children.[18] Scarcity of research leads to difficulty in identifying the cause. As observed in India, 35% cases having no definitive cause were attributed to environmental, familial and psychosocial factors.[19] This current research was designed to be a case-control study, targeting maternal, paternal and social factors so as to add up to the causal pathway of ID.

In our sample, male predominance was observed in ID cases which is concurrent with an earlier study.[20] In the current study, more than half of the mothers of controls were matriculate (high school). It was supported by a review that indicated that mild ID was dependent on the level of maternal education, availability, accessibility of education and healthcare facilities.[20] The current study emphasises on the role of female education and awareness of mothers in aiding prevention of this disability.

Mostly fathers of controls were educated with varying levels of degrees compared to fathers of cases which is consistent with an Indian study in which father's level of education was higher compared to mothers in ID children.[21] A relationship between ID and paternal education has been highlighted even after adjustment in the current study which needs further exploration with the help of longitudinal studies.

In majority of cases, mothers were housewives, while one-third fathers belonged to unskilled, skilled labour and professional class. A study observed that one-third of mothers were housewives compared to more than half the fathers belonging to unskilled, skilled labour and professional class.[21] Mothers of most of the cases and controls belonged to the 14-24 years age group at the time of marriage while one-fourth were categorised within the range of 31-50 years at the time of birth of the participant. Concurrent with the results of a study, mothers of more than half of the cases had the age above 30 years,[22] thus leading to the conclusion that although early marriage has no significant relationship, advanced maternal age can be considered an influencing factor in the development of ID. This study exhibited a significant association between father's age with development of ID. Majority cases were born to fathers in the 40-55 years age group. These results corroborate with results of a study conducted in Andhra Pradesh, India, which reported 1.8-fold rise in relative risk in fathers in the 30-34 years age group. Another study highlighted a dose-response relationship between advancing paternal age and ID.[23]

Disparities in social status were observed, as more than half the cases belonged to middle class and one-third to lower class respectively. This is supported by a systematic review of 19 studies conducted in 50 low and middle income countries.[23] In another study, severe ID (IQ<50) was more profound in illiterate mothers, rural areas and low socioeconomic status.[24]

Consanguinity (cousin marriage) is influenced by religion, ethnicity, culture and geography exhibiting high trends in Pakistan.[24] The current study results depicted more than two-third cases giving history of consanguinity between their parents, establishing a highly significant association, as supported by an Indian study where consanguinity was established as one of the major causes in one-third cases.[22] This study highlighted more than 70% cases with history of first cousin marriages. The same was observed in Iran where 77% cases had a history of consanguinity, 50% of these were first cousin marriages.[25] This is an important finding as it not only bridges the gap between socio demographic factors, but also emphasises the need for awareness among the masses via public health interventions. A very low percentage of cases indicated a genetic origin, and after adjustment only paternal history of mental retardation was associated significantly, emphasising the novelty of this study in aiming to highlight the association of maternal, paternal and social factors with ID.

It is noteworthy that this study highlights significant associations between perinatal factors and ID. More than half the cases had experienced trauma, one-third asphyxia and a quarter were resuscitated with oxygen at the time of birth. Supporting evidence comes from a study conducted in the United Kingdom in which an association was observed between resuscitation at the time of birth and lower IQ levels. Besides, need for resuscitation was associated with poor maternal education, LBW babies of primigravida delivered by caesarean section.[26] Another

study identified that resuscitation with assisted ventilation increased risk of ID.[27]

Delayed cry at the time of birth is significantly associated with ID, especially in developing countries. In an Indian study, one-third cases had a history of delayed cry at the time of birth.[11] Similar results were observed in this study with more than one-third cases having a history of delayed cry. This study is one of its kind having been conducted in Pakistan, as it throws light on perinatal factors globally recognised as responsible for ID, to be understood and identified in our settings.

The role of breastfeeding has been stressed upon by the current study. Majority of the controls had a history of having been breastfed, which is consistent with the findings of a Scottish study in which breastfeeding was observed in majority controls being discharged, concluding that ID may be linked to babies not being breastfed and discharged from special baby care unit.[28] It should be noted that no effect of duration of breastfeeding was observed which is contrary to the results of a study which concluded, that after adjusting a dose-response relationship it was observed, between IQ and nine months of breastfeeding.[29]

Multivariate logistic regression concluded that after adjusting for asphyxia at the time of birth, economic status of parents, maternal and paternal educational status, maternal history of mental retardation, use of oxygen and trauma to the baby, only history of consanguinity, delayed cry at the time of birth, paternal history of mental retardation had a significant association with ID among special children. Breastfeeding had a protective effect in relation to ID.

In Pakistan, limited literature is available on ID. The current case-control study highlights the cause-effect relationship between the factors under study and ID, thus opening up avenues for longitudinal studies to be conducted.

As the study was self -funded, a few limitations were encountered. The ratio of cases-to-controls should have been 1:2. Ideally, the IQ test of cases should have been conducted by the researchers, but only the results of the tests conducted at the institutions was taken for analysis. Similarly, for the controls, IQ test should have been conducted but students with good academic score were selected. Prospective study with a larger sample would is recommended.

Conclusion

In order to address the neglected avenue of mental health in Pakistan, the results of the current study are a help to understand that not only genetic but parental, social, environmental and familial causes contribute to ID, especially perinatal and paternal factors. These factors presumably can be prevented by conduction of premarital counselling, genetic screening sessions as well as health education and awareness regarding breastfeeding and provision of antenatal and perinatal care which is a challenging task.

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