Research Article

# Association of Social, Paternal and Maternal Factors with Intellectual Disability: A Case-Control Study

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## Abstract

**Background:** Pakistan has a high number of intellectually disabled people, who cannot learn and function at expected levels. The scarce research done here shows consanguinity, malnutrition, low socio-economic status, and maternal illiteracy to be risk factors.

**Objectives:** To identify risk factors for Intellectual Disability in Pakistani children and to recommend preventive measures for parents and policymakers.

**Methods:** This was a case-control study conducted in Lahore, Pakistan from February to May 2022. The total sample size was 378 with 126 cases and 252 controls (ratio 1:2), aged 2-19 years. Parents were interviewed and data was entered into a structured questionnaire. Data was entered in SPSS 26 and analyzed.

**Results:** Significant differences between cases and controls were observed for consanguinity (p=0.001), educational level (p=0.001), socioeconomic status of parents (p=0.001), and for paternal genetic and neurological factors (p=0.001). Multiple logistic regression analysis showed that a history of consanguinity (p=0.001) and low maternal education (p=0.001) had a significant effect on ID. Low socioeconomic status showed no significant association with ID, nor did breastfeeding. Adjusted Odds Ratio (OR) showed that the risk of developing ID increases by a factor of 7.8 and 4.1 respectively for history of consanguinity and low maternal education.

**Conclusions:** Consanguinity emerged as a major risk factor for intellectual disability. This should be discouraged by pre-marital counseling and health education.

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## Introduction

Intellectual Disability (ID) is a condition where "…there are limits to a person's ability to learn at an expected level and function in daily life".[1] It involves severe deficits, arising in the developmental period, in two domains: intellectual functioning or general intelligence like learning and problem solving, and also in adaptive functioning which includes daily life skills needed for an independent life, communication, and social skills.[2] Intelligence can be assessed with the Intelligence Quotient (IQ), which can be measured by numerous tests, the most widely used being the Wechsler Adult Intelligence Scale (WAIS) for adults and the Wechsler Intelligence Scale for Children (WISC).[3] However, other factors like environment and language must also be considered during the assessment of intellectual

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disability. Various tests are also available to determine adaptive functioning.

Estimates showed only two databases provide comprehensive estimates of global disability prevalence in children and adolescents. UNICEF household surveys showed moderate-to-severe disabilities of all types in functional status prevalent in 28.9 million children aged 0-4 years, 207.4 million aged 5–17 years, and 236.4 million children aged 0–17 years. They estimate a prevalence of 10.1% in the 0–19-year age group which amounts to 266 million children. The Global Burden of Disease 2019 showed 49.8 million under 5 years, and 241.5 million between 5-19 years had mild-to-severe disabilities of all types, amounting to 291.3 million (11.3%) children <20 years old. Both reports showed South Asia and Sub-Saharan Africa carrying more than half the burden of global disabilities.[4]

Pakistan is a Low and Middle-income country marred with economic and social difficulties. According to the last census in 2017, Pakistan has a population of 207 million.[5] It is estimated that the burden for all types of disabilities is 371,8336.[6] Latest UNICEF data shows that 13% of its households have at least one member with a disability.[7] Children with intellectual disabilities represent a high proportion of Pakistani youth, a cause of concern for parents, the already strained health care system, and society.

Much research has been done in the developed world and the risk factors for ID identified thus far include advanced paternal or maternal age, congenital infection, prematurity, low birth weight, low Apgar score, premature membrane rupture, fetal distress, multiple caesarian sections, and more than 30 minutes on assisted ventilation.[8] Very little research exists in Pakistan, and research done in the Western World is not universally applicable due to differences in health care systems and culture.[9] The scarce research done here shows that the risk factors are consanguinity, malnutrition, socioeconomic status, and maternal illiteracy. In a significant number of children with ID, no known cause was identified.[10] In addition to the economic and emotional burden of a disabled child, the social stigma leads to delay in diagnosis and treatment, and social isolation of the child and family. Lack of awareness, particularly about risk factors and prevention, exacerbates the problem. In a country like Pakistan, already facing multiple economic challenges, more research is needed to understand the factors contributing to ID and to be able to identify the points of intervention that could arrest the already high disability burden.[7] The aim of this study was to identify risk factors for Intellectual Disability in Pakistani children and to recommend preventive measures for parents and policymaker.

## Methods

This case-control study took place at 3 institutes in Lahore, Pakistan from February to May 2022. The calculated sample size was 378 with 126 cases and 252 controls (ratio 1:2), a significance level of 5%, power at 80%, anticipated probability of exposure (cases) at 73%, and anticipated probability of exposure (controls) at 60%[11]. Cases consisted of Intellectually Disabled students from the Rangers' Institute of Special Education (RISE), already diagnosed and categorized by the institute into congenitally malformed, mentally retarded, autistic, Down Syndrome, and others e.g., visually impaired, and slow learners, aged 2-19 years. An attempt was made to interview all 127 students at RISE, and 126 cases were finally interviewed. After matching for age and gender, controls were selected and data was collected from them from Al-Kafeel School System (130 students out of a total of 500), and Ranger's Public-School Rise and Shine (122 students out of a total of 237.

Data was collected after getting authorization from relevant institutions. The Ethical Review Committee of CMH Lahore Medical College gave ethical permission for the study (#.732/ERC/CMH/LMC). The data collection tool was a structured questionnaire largely based on a pre-validated, published study, which took approximately 20 minutes to fill[11]. It consisted of demographic data followed by questions regarding parental education, socioeconomic status, parental consanguinity, paternal history of intellectual disability, and history of breastfeeding. Socioeconomic status was defined by the following cut-off values: Low (< Rs. 50,000), Middle (Rs 50,000-100,000) and High (> Rs 100,000). A history of Breastfeeding was taken to mean that the child had been breast feed for at least 6 months, whether exclusively or not. The questionnaire was in English and interviewer administered. Interviewer bias was minimized by using trained interviewers from both genders fluent in English

as well as local languages who could translate the questionnaire for the respondents. The parents of the selected children were interviewed, after getting informed consent and assurance of confidentiality. Recall bias was sought to be minimized by checking records kept at the institutes. Data was analyzed with SPSS version 26. Frequencies and percentages were calculated for categorical variables. Logistic regression and Chi-square test of significance were used to see associations between intellectual disability and risk factors. P value < 0.05 was considered statistically significant.

## Results

Out of a total of 378 participants, there were 126 (33.3%) cases and 252 (66.7%) controls. Age was between 2-10 years in 67 (53.2%) cases and 229 (90.9%) controls; and between 11-19 years in 59 (46.8%) cases and 23 (9.1%) controls. The cases consisted of 55 (43.65%) boys and 71 (56.34%) girls. The majority of mothers in both groups, 119 (94.1%) in cases and 220 (87.3%) from controls were housewives. Among cases, 106 (84.1%) had a history of consanguinity among parents, compared to 102 (40.5%) in controls; this difference was statistically significant (p=0.001). First-cousin marriage between parents was found for 169 (44.7%) students. Educational levels of fathers and mothers varied in cases and controls and this difference was also statistically significant in both categories (p=0.001). Similarly, Parents' economic status also showed a statistically significant difference between cases and controls respectively (p=0.001). Table 1 depicts the association of social factors with intellectual disability.

**Table 1:** *Association of Social Factors with Intellectual Disability (n=378)*

| Variables | Cases (n=126) | | Controls (n=252) | | P-Value |
|---|---|---|---|---|---|
| | n | % | n | % | |
| Consanguinity | 106 | 84.1 | 102 | 40.5 | 0.001 |
| **Educational status of mothers** | | | | | |
| Illiterate | 28 | 22.2 | 10 | 4 | 0.001 |
| Primary-Matriculation | 75 | 59.5 | 93 | 36.9 | |
| Graduate & above | 23 | 18.3 | 149 | 59.1 | |
| **Educational Status of Father** | | | | | |
| Illiterate | 10 | 7.9 | 0 | 0 | 0.001 |
| Primary-Matriculation | 88 | 69.8 | 38 | 15 | |
| Graduate & above | 28 | 22.2 | 214 | 85 | |
| **Economic Status of Parents** | | | | | |
| Low (< Rs. 50,000) | 124 | 98.4 | 202 | 80.2 | 0.001 |
| Middle (Rs. 50,000-100,000) | 2 | 1.6 | 47 | 18.7 | |
| High (> Rs 100,000) | 0 | 0 | 3 | 1.2 | |

Paternal genetic and neurological factors also varied in cases and controls and this difference was statistically significant (p=0.001) as depicted in Table 2.

Multiple logistic regression was applied to check the significant combined effect of all study variables on intellectual disability. Results are depicted in Table 3.

**Table 2:** *Association of Paternal Genetic and Neurological Factors with Intellectual Disability (n=378)*

| Variables | Cases (n=126) | | Controls (n=252) | | P-Value |
|---|---|---|---|---|---|
| | n | % | n | % | |
| **Congenital Malformations** | | | | | |
| Yes | 12 | 9.5 | 4 | 1.6 | 0.001 |
| No | 114 | 90.5 | 248 | 98.4 | |
| **Mental Retardation** | | | | | |
| Yes | 9 | 7.1 | 3 | 1.2 | 0.001 |
| No | 117 | 92.9 | 249 | 98.8 | |
| **Autism** | | | | | |
| Yes | 3 | 2.4 | 1 | 0.4 | 0.001 |
| No | 123 | 97.6 | 251 | 99.6 | |
| **Down Syndrome** | | | | | |
| Yes | 2 | 1.6 | 1 | 0.4 | 0.001 |
| No | 124 | 98.4 | 251 | 99.6 | |
| **Any Other (history of any inherited psychiatric illness e.g. schizophrenia or inherited physical disability)** | | | | | |
| Yes | 22 | 17.5 | 7 | 2.8 | 0.001 |
| No | 104 | 82.5 | 245 | 97.2 | |

**Table 3:** *Multivariate Logistic Regression (n=378)*

| Variable Name | Adjusted Odds Ratio | P-Value |
|---|---|---|
| Consanguinity | 7.8 | 0.001 |
| Low Maternal Education Status | 4.1 | 0.001 |
| Low Economic Status of Parents | 3.9 | 0.09 |
| Breast Feeding | 0.68 | 0.16 |

Significant differences between cases and controls were observed for consanguinity (p=0.001), educational level (p=0.001), socioeconomic status of parents (p=0.001), and for paternal genetic and neurological factors (p=0.001). Multiple logistic regression analysis showed that after adjusting for low socioeconomic status and breastfeeding, a history of consanguinity (p=0.001) and low maternal education (p=0.001) showed a significant effect on ID. Low socioeconomic status showed

no significant association with ID, nor did breastfeeding. An adjusted Odds ratio (exp.β) of 7.8 shows that the risk of developing ID increases by a factor of 7.8 if someone has a history of consanguinity compared to not having consanguinity. Similarly, the Adjusted Odds Ratio of 4.1 shows that low maternal education status increased ID risk by 4.1 times.

## Discussion

It is a matter of great concern that cousin marriages are contributing to producing disabled children. Deficiencies in research lead to difficulties in identifying the causes. This problem was also observed in an Indian study where no definitive cause was found in 25% of cases[12]. This research was designed to focus on social as well as maternal and paternal etiological factors for ID.

In this study, over 75% of cases had a history of parental consanguinity, showing a highly significant association with the development of ID. The same was observed in a study carried out by Omar and Kokab in Lahore, Pakistan, where 73% of cases gave a history of consanguinity between their parents.[11`] Other studies from India also showed fairly high levels of consanguinity, 37.02%, and 29.14% respectively.[13,14] Consanguinity is quite common in Pakistan, reflected in both culture and religion, with figures as high as 62.7%, with higher figures seen from rural areas.[15]

This study emphasizes the role played in the prevention of ID by maternal education; 25% of the mothers of cases were illiterate and over 66% were just matriculate. These results are almost similar to the Lahore study where 15% of mothers of cases were illiterate and 52% were just matriculate.[11] These are also supported by a review carried out by Kaufman, Ayub, and Vincent where, even after adjusting other variables, a relationship was observed between maternal education and ID[16]. Similarly, fathers of cases were less educated as compared to fathers of controls and this difference was statistically highly significant.

Differences in social status were observed as nearly 99% of cases belonged to lower socioeconomic class. A systematic review studying the association of home socioeconomic circumstances from poorer countries supports this observation, which could be either a cause or a result of the disability.[17] A very low percentage of cases indicated a neurological or genetic origin, however, all paternal neurological factors such as congenital malformations, mental retardation, autism, and Down Syndrome were significantly associated with ID. These findings are almost similar to Omar and Kokab's study where paternal genetic factors such as mental retardation and Downs Syndrome were significantly associated with ID.[11] As far as the role of breastfeeding in the prevention of ID is concerned, the majority of controls in our study were found breastfed and while adjusted Odds Ratio was <1, the result was not significant. This finding is consistent with a Scottish study where the majority of controls revealed a history of breastfeeding.[18]

Multivariate logistic regression found that after adjusting for low socioeconomic status and breastfeeding, the following factors showed a significant association with intellectual disability: a history of consanguinity and low maternal education. Socioeconomic status and breastfeeding were shown to have no significant effect on ID.

Our study has a few limitations: our researchers did not conduct our own IQ tests for cases but utilised those provided by the institutions. Moreover, a small sample size and a case-control design means that the study does not imply causation.

## Conclusion

Mental health is a neglected field in Pakistan, and our study helps create awareness of the risk that social factors pose for mental illness, especially consanguinity, in addition to genetic or neurological factors. These factors can be prevented by pre-marital counseling and public awareness. Pre-marriage screening tests should be declared mandatory. Policy makers should also address the low levels of female education and high poverty in the country.

**Ethical Approval:** The Ethical Review Committee CMH Lahore Medical College & Institute of Dentistry approved the study vide letter No. Case #. 732/ ERC/ CMH/ LMC.

**Conflict of Interest:** The authors declare no conflict of interest.

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**Authors' Contribution:**

**MAC:** Conception & design, analysis & interpretation of data, drafting of article, critical revision for important intellectual content, final approval

**ZO:** Conception & design, critical revision for important intellectual content, final approval

**HA:** Conception & design, drafting of article, final approval

**SR:** Analysis & interpretation of data, drafting of article, critical revision for important intellectual content, final approval

**UAS:** Conception & design, drafting of article, final approval

**BA:** Analysis & interpretation of data, critical revision for important intellectual content, final approval

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