

Blinding Retinopathy of Prematurity in Western India: Characteristics of Children, Reasons for Late Presentation and Impact on Families

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Objective: To ascertain why children with end-stage retinopathy of prematurity (ROP) become blind, and to explore the impact of blindness on families.

Design: Mixed quantitative and qualitative methods.

Setting: Tertiary-care eye hospital in India.

Participants: Children with end-stage ROP and their carers.

Intervention: Cases presenting between June 2009 and July 2016 were identified from medical records and data extracted. Carers were contacted for missing information, if required. Data were analyzed to explore where failure had occurred in the process of screening and treatment. A subset of carers were selected for in-depth interviews to explore the impact of having a blind child. Interviews were recorded, transcribed and analyzed using a thematic framework.

Main outcome measures: Parental perceptions.

Results: 66 children were included: median age 4.3 y (range 3 mo– 6 y), 58% boys. 74% were blind due to 'screening failure', which was associated with lower maternal education ($P=0.03$). Of the 17 cases of treatment failure (24.6%), majority (12, 70%) had aggressive posterior ROP. A subset of carers of 18 children (50% boys) were interviewed, mostly mothers. Most reported impoverishment as a result of having a blind child, and many reported lack of access to special education, negative attitudes of others and concerns about the future.

Conclusions: Screening for retinopathy of prematurity needs to be expanded and counselling improved. Access to special education and rehabilitation need to be improved.

Keywords: Health expenditures, Perceptions, Screening, Qualitative studies.

Retinopathy of prematurity (ROP) is becoming an important cause of blindness in children in middle income countries [1]; approximately 2,900 preterm infants are likely to become visually impaired or blind every year in India [2]. Improvement and expansion of neonatal care in India over the last two decades has led to more preterm infants surviving, and thus at risk of ROP [3,4]. Control of visual loss from ROP requires high-quality neonatal care, to reduce exposure to known risk factors; timely screening of preterm infants at risk, followed by urgent treatment if required. The National Neonatology forum of India (NNFI) published guidelines for ROP in 2010 [5], but screening and treatment are not being universally implemented, even in the major cities in India [6].

Progression to the blinding stages of ROP can occur because of failure of screening, or failure of treatment, which may be due to service provider factors, such as regular screening not being in place; patient factors, such as failure to attend for screening and/or treatment after the infant has been discharged from the neonatal unit, or

disease factors, such as the presence of aggressive posterior ROP (AP-ROP) which has a poor prognosis despite treatment.

This study was undertaken to explore the reasons why children became blind from ROP, and to gain insights into the points of failure in the health system. A second aim was to assess the impact on families of caring for a ROP blind child.

METHODS

The study was undertaken in a tertiary-care eye hospital in Pune, India in June and July 2016. The hospital is a large, not-for-profit, eye hospital that charges user fees which are reduced or waived for poor patients. Ethical approval was obtained from the ethics committees of the study hospital and the London School of Hygiene & Tropical Medicine. Informed consent was taken over the phone in the presence of two witnesses, or face-to-face. All carers interviewed, whether over the telephone or personally, were offered free services such as counselling or guidance on special education, if required.

To establish reasons why children who presented with end stage ROP (Stage 4b or 5) became blind, quantitative methods were used. A convenience sample of potentially eligible children were identified from the hospital's electronic database and medical records. Eligibility criteria were children presenting with end stage ROP with very poor vision in both eyes between June 2009 and July 2016. Exclusion criteria were unilateral disease, those with Stage 4a who may have been visually impaired but not blind, and those with incomplete clinical data. For all children who present to this hospital with advanced ROP, the protocol requires that a pediatric optometrist assesses the visual acuity, including light perception in a darkened room, and that carers be asked where their child received neonatal care and whether their child was screened and/or treated for ROP. Data are extracted from neonatal discharge summaries when available. For this study, data extracted from the records of eligible children included: age at presentation, mother's age at delivery, visual acuity and ocular findings at presentation, gestational age (GA), birth weight, the neonatal care unit where the child received care, whether screening and/or treatment for ROP had been documented as recommended or having taken place, and the findings, if applicable. If information about ROP was not recorded in the medical records carers were contacted by telephone if the information was not considered sensitive (*e.g.*, level of education) or by face-to-face interview for sensitive information (*e.g.*, to explore whether screening had been recommended). During interviews, parents were asked to describe the whole sequence of events in relation to ROP from NICU admission to discharge *i.e.*, whether ROP was mentioned, whether screening was recommended or undertaken while in the unit or after discharge; and whether treatment was recommended or undertaken. If screening or treatment were recommended but not undertaken they were asked to explain why this was the case. All this information was used to assess why the child became blind. Type of NICU (public and private) were categorized by their location into 'metropolitan NICUs' (*i.e.*, in Pune metropolitan area) and 'non-metropolitan NICUs' (*i.e.*, in smaller cities and towns).

To explore the impact on families of having a blind child, indepth face-to-face interviews of the select carers from aforementioned group were undertaken. As the impact on the family was likely to vary depending on the age and gender of the child, carers of children of different ages and both sexes were purposively selected *i.e.*, equal numbers of boys and girls aged 0-2 years, 2-5 years and more than 5 years. Carers of 19 children were invited to participate. An interview topic guide was developed to assess impact in five domains, based on the World Health

Organization's (WHO) International Classification of Functioning: impairment, activity limitation, participation, personal factors and environmental factors [7]. The principal investigator conducted the interviews in the local languages (Marathi/ Hindi) ensuring privacy. A trained, experienced female counsellor was present during all the interviews to provide support and guidance to participants and to note their body language and emotional reactions. Interviews were recorded and transferred to a computer after assigning a code number to ensure confidentiality. Field notes were made after each interview. Qualitative data were transcribed in the local language and then translated into English. Translations were checked by a co-researcher to ensure validity. Thematic analysis was undertaken, using the WHO International Classification of Functioning framework [10] as a starting point and then identifying additional themes or subthemes.

Statistical analysis: Quantitative data were entered into an excel spreadsheet, cleaned, transferred into STATA/IC/14 for analysis. Statistical tests of significance were carried out to establish the strength of association between parents' education, occupation, the type, level and location of NICU and reason for blinding ROP.

RESULTS

Medical records of 86 children with Stage 4 or 5 ROP were reviewed; 20 children were excluded as they were either unilaterally blind *i.e.* had Stage 4a in their better eye (12), parents refused to participate (3) or medical records were incomplete (5). Sixty six children were (58% males) included in the study, 26 carers each provided additional information on phone and by interview.

The median (range) gestational age of the 66 children was 31.2 (26- 36.3) weeks, birth weight was 1175 (700-2300) g, and the median age at presentation was 4.3 (range 2- 35) months. The median age of the mother at delivery was 22.8 (range 18-34) years. Education and occupation data of 14 parents could not be obtained. The majority of carers (40/66, 60.7% fathers; 43/66, 65.1% mothers) were educated to up to high school or less and all were literate. Most fathers (77%) worked while most mothers (68%) were homemakers. The affected child was an only child in 20 (30.4%) families. Most deliveries and NICU admissions (69.7%) were in private facilities and most (77.3%) had received care in a tertiary-level NICU. Most children were referred to the eye hospital by an ophthalmologist (51.5%) or self-referred (30.3%), and only 9% were referred by a pediatrician. Four children had been screened and treated as part of the study hospital's ROP program. Over three quarter of the

children presented with perception of light (PL) or no light perception (52, 78.8%) and over half (38, 57.6%) had stage 5 ROP.

The type and location of NICU where children had received neonatal care was strongly associated with ROP blindness, with children cared for in Pune being more likely to have received care in a public NICU (90%) whereas children from smaller cities or towns were more likely to have received private care (75.5%) ($P < 0.001$).

Reasons for blinding ROP were broadly divided into two categories: 'screening' or 'treatment' failure. Screening failure was the reason for blinding ROP in 49 (74%) cases; 33 (50%) children had received care in an NICU which did not have a program for ROP, 9% were not screened despite screening being in place, 9% were screened late, and 5% were not given an appointment for screening after discharge. Treatment failure was the reason for blinding ROP in 17 (26%) cases. The medical records of 12 of these children indicated that they had been treated for APROP. Parents of three children did not seek treatment despite advice, and two children had delayed treatment on account of late screening.

For the 52 mothers for whom information on education was available those educated up to high school only were more likely to have a child blind from 'screening failure' (90%) than children of more educated mothers, where the reason was likely to be 'treatment failure' (41.7%) ($P = 0.03$). Education of the father, occupation of the mother or father were not associated with the reason for blinding ROP.

Impact of having a child who is blind from ROP

Nineteen carers were invited for in depth interviews, 18 of whom agreed. Carers of 9 boys and 9 girls were interviewed and the children ranged in age from 4 months to 7 years (8 aged 0-2 years, 6 aged 2-5 years and 4 older than 5 years).

Experience of diagnosis and access to treatment: Over half the caregivers first noticed that their child had a problem at around three months of age with the remainder reporting an older age.

"During our follow up visits we even asked the pediatrician about any eye problem. He never told us anything. Later when she was 7-8 months old, something white appeared in her eyes." (6 years, female)

The mother or an immediate family member noticed a problem in six children, whereas an ophthalmologist or a pediatrician noted it in the rest. Some carers were aware of the serious nature of the disease but could not seek treatment owing to the high cost.

"Expenses for laser were 28000 Rupees (USD 413) for both eyes. The doctor in the private hospital told us that he may need 2-3 sessions of laser. We couldn't pay." (5 months, Male).

One mother expressed anger towards the neonatologist who had not advised screening for ROP and another hoped that a new treatment would be discovered to treat her child. No carer reported being able to access counselling or rehabilitation services for their child.

"If only I knew about ROP, I would have gone to any extent to get her treated. We even confronted the neonatology doctors. They started check-up for ROP in the NICU after that." (3 years, Female)

Impact on development: One mother felt that there had been an enormous negative impact on development of her daughter due to blindness. This girl had an unaffected twin brother which enabled comparison regarding their development.

"We feel (the difference)...everything...I mean, she has a twin brother. He is ahead in everything and she is like this." (3.5 year, Female)

Concern about activities- playing, reading: Some carers thought that their child did not play or try to reach out for toys, and one reported her child not being able to read. This mother was perturbed that despite her being a local teacher, her own son could not read due to his blindness.

"In our religion children start reading Quran (holy book) when they turn 5 but he cannot see." (7 year, Male)

Participation- social activities, schooling and independence: Some carers thought that other children excluded their child from social activities while one mother reported that her child avoided socializing.

"I mean other children (drops the sentence midway)...She keeps sitting at one place only. She does not get with others. Even if there is a birthday party in the neighborhood...she does not go...she herself says no (to go to the party). She says- Mummy, I do not want to go." (3 years, Female)

Two of the four children of school going age attended school. Most parents of the preschool children were extremely worried about their child's schooling.

"I think she will not get admission in any (regular) school. Teachers there will not understand her." (9 months, Female)

The mother of a six year old female child was anxious about her independence in day-to-day activities.

"We worry whether she will be able to do everything on her own (breaks down and starts crying inconsolably)." (6 years, Female)

This mother from a wealthy family in a small village was worried that, although the whole family was supportive of her daughter, they were overprotective. This proved to be a barrier for her enrollment in a residential school and becoming independent. Both the husband and wife were distressed and appeared not to have come to terms with their daughter's disability.

Personal factors- the caregiver: While sharing their thoughts about their child being able to lead an independent life, some carers thought that it was impossible, some were not sure.

"No...I don't think so. I feel...he cannot see...(choked voice)...nobody will make friends with him (starts sobbing)...nobody will play with him...(wipes tears, controls her emotions and stops)" (9 months, Male)

One mother attributed her own strong, independent character to her blind child.

"God brought this child into my life so I could become a strong, independent person. I was such a timid person.... (When he started going to school) I urged my husband to buy me a scooter. Today I take him to school on a scooter." (7 years, Male)

The mother of this boy, her third child, came from a poor background but was immensely positive about the situation. She was satisfied with the education her son was receiving at a school for the blind, and seemed confident about her son becoming independent and being a support to others in the future.

Interpersonal relationships: Two mothers mentioned that the circumstances of having a preterm baby and having a blind child had had an enormous impact on their interpersonal relationships.

"We fight frequently over money. We cannot take her to any hospital for treatment as we don't have money. I feel we are ruining her life." (1.5 years, Female)

Blame - role of family and community: One mother, who had separated from her husband, was living with her child in her parents' house. Her husband and his family were upset because the child was not only blind but also a girl. Another mother went through the trauma of divorce proceedings before reuniting with her husband after he agreed to leave the joint family.

"They blamed me after he was diagnosed with blindness. My in-laws even suspected my fidelity. I

offered to undergo a DNA test." (6 years, Male)

Several mothers experienced negative attitudes of from the community, and some reported being blamed by others for their child's condition. One mother reported that her blind daughter was treated differently from her normal twin brother.

"Neighbours blame us. They say, 'You neglected her. She would have been cured if you had sought care earlier. They blame us.'" (1.5 years, Female)

"People ask all sorts of questions such as 'why does she look like this? Why does she rub her eye like this?' We can't face such questions." (3.5 years, Female)

Financial impact: Most carers found it difficult to raise the money needed for treatment and follow up, and some reported loss of income. Nearly all had to borrow money from relatives/ friends, and some sold or mortgaged their gold jewelry. Several fathers had taken out a loan, either from their employer or from money lenders. The financial implications caused a great deal of anxiety and distress.

"...we survive on daily wages.....he (husband) is a driver....so...its all....(choked voice) dependent on daily earning....so we had to borrow money from people for his operation (starts crying but controls herself)" (9 months, Male)

"My husband had to take so many leaves for the child's treatment. He is on the verge of losing his job." (5 months, Male)

Age- and gender-differences: There were no age or gender differences in the financial impact on families, nor in carers' concerns about the future or the development of their child. However, mothers of older children (>3years) were more likely to report social exclusion and limitation of activities/participation. There were no differences in families where there were other siblings, and among the two sets of twins, one mother reported differences in the affected child's development whereas the other did not.

DISCUSSION

In this study, the main reason why children had become blind from ROP was because of lack of, or inadequate screening. Failure of treatment was the cause in approximately a third of children. Most cases of treatment failure were diagnosed as having APROP and were treated with laser. This highlights the potential for poor outcomes associated with APROP [8]. In Pune metropolitan area 'treatment failure' was the main reason, reflecting the wide coverage of ROP screening, whereas in smaller cities/towns 'screening failure' predominated.

The gestational age and birth weight of children

WHAT IS ALREADY KNOWN?

- Screening failure is an important reason for blindness associated with Retinopathy of prematurity in India.

WHAT THIS STUDY ADDS?

- This study reports the financial, personal and social impact on the families of children blind from retinopathy of prematurity, and the lack of counselling and rehabilitation services for children and their carers.

included in this study were similar to studies from north India [9, 10] and Mexico [11] but the median age at presentation was lower may be because most parents were educated and likely to seek care early. There were more boys than girls in the present study, which could reflect gender differences in health seeking behavior, as has been demonstrated in children undergoing surgery for bilateral cataract in Asian countries [12]. This could be because some families are more willing to pay for treatment of their sons than for their daughters.

Other studies from India have reported that more than 80% of blind children were never screened for ROP [9,10] and 3% [9] were lost to follow up after one screening episode. Half of the children in the study from Mexico [11] were never screened due to lack of a ROP program in the NICU, and a further 33.4% children either slipped through the screening net as they were not given follow up appointments or failed to attend. In the present study, failure of treatment was the cause in approximately a third of children which was higher than in other Indian studies (3.5% - 10.6%) [9,10], and in the Mexico study (14.6%) [11].

To our knowledge this is the first qualitative study to assess the impact on families of having a child blind from ROP, although other studies have assessed the impact of having a disabled child [13]. Health-related quality of life has been reported among caregivers of children with disabilities [14] and among parents of children enrolled in the CRYO-ROP trial [15]. In the latter study, almost a quarter of parents' health utility index scores had negative values, which represents extremely low quality of life [16]. In the present study carers had varied responses, with a few being positive but the majority were very distressed by their child's blindness and anxious about the future. A limitation of this study is the possibility of a bias as it was undertaken in a hospital where there are fees for services. The findings may not be generalizable to other parts of India or to studies undertaken in a government sector where care is free. The small number of caregivers of older children in the qualitative study may not have captured the full range of impacts on families, and may have under-estimated the

impact on social exclusion and participation, which was of greater concern to carers of older children. Direct interviews of older children could have provided greater insight into the impact on social, emotional aspects of their lives.

Services for screening and treatment of ROP need to be expanded, particularly in smaller cities and towns, and the importance of timely screening, treatment and follow up is communicated clearly to parents using language they will understand. A study from Southern India reports improved follow-up from rural area after interventions to address various barriers [17]. Counselling services, and rehabilitation for young children blind from ROP need to be established to reduce developmental delay. Awareness needs to be created about the financial support available from the Government of India's 'Rashtriya Bal Swasthya Karyakram' (RBSK) program which specifically includes ROP as one of the 30 childhood conditions it is designed to address [18].

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Contributors: SK: conception and design of the work; acquisition, analysis and interpretation of data for the work; and drafting the work, and revising it critically for important intellectual content; CG: conception and design of the work, interpretation of data, revising the draft critically for important intellectual content; MZ: design of the work, revising the work critically for important intellectual content; SA: collection and analysis of data; MD: interpretation of the analysed data, critical review of the manuscript and final approval of the version to be published. All authors approved the final version to be published.

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