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*Original Article*

# Health and well-being of children with congenital cardiac malformations, and their families, following open-heart surgery

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**Abstract** Infants who survive open-heart surgery are at risk for developmental disability, which may impact on the well-being not only of the child, but also the family. The objective of our prospective study, therefore, was to determine the long-term health-related quality of life of children with congenital cardiac malformations following open-heart surgery, and to describe the persisting level of stress in their families. To this end, 49 parents completed the Child Health Questionnaire, the Parenting Stress Index, and the Child Behaviour Checklist as part of a developmental follow-up protocol when their child was 5 years of age. Mean scores on the Child Health Questionnaire were in the normal range, with physical well-being equal to 53.5, psychosocial well-being 50.9, with only 6.4 percent and 8.5 percent of subjects, respectively, falling within the suboptimal range of less than 40. The distribution of scores on the Parenting Stress Index, however, were more variable, with over one-quarter of parents indicating a high level of stress, with almost one-fifth having low levels of stress, and just over half scoring in the normal range, with the group mean being 52.6 plus or minus 32.3. An abnormal neurologic examination before surgery was associated with lower physical health ($\beta$ equal to $-5.5$, p equal to 0.02, $r^2$ equal to 0.18), whereas lower arterial saturations of oxygen, less than 85 percent preoperatively, was associated with lower psychosocial health ($\beta$ equal to $-6.6$, p equal to 0.01, and $r^2$ equal to 0.14). The internalizing and externalizing behaviours of the child were significantly correlated with psychosocial well being, with r ranging from $-0.32$ to $-0.52$, and p less than 0.05. Parental stress also correlated with psychosocial health (r equal to $-0.48$ and p equal to 0.0009). Overall, the perception by the parents of the health-related quality of life of their child is favourable 5 years following open-heart surgery during infancy. Many parents, nonetheless, continue to feel either stressed or defensive about their child, particularly if their child exhibits behavioural difficulties. Our findings suggest that strategies need to be considered to enhance family well-being in the planning and delivery of health services to this population at high risk.

Keywords: Congenital heart defects; quality of life; parental stress

CONGENITAL CARDIAC MALFORMATIONS ARE frequently encountered in newborns, and can be lethal or severely debilitating if not repaired surgically early in life. The use of cardiopulmonary bypass and deep hypothermic circulatory arrest in such neonates and young infants over the past 20 years has resulted in a dramatic increase in survival for those born with these malformations.[1] There is, however, increasing concern for the neurologic integrity of those surviving such surgery. These young infants may be exposed to hypoxic-ischaemic insults prior to, during, and/or following surgery, posing a risk for cerebral injury, with lifelong developmental consequences. There are few prospective studies delineating the developmental sequels associated with cardiac surgery carried out during infancy. One that has

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been performed indicated that global developmental delays, and subtle neurologic deficits, are common, although severe disability is rare.[2] Further elucidation of the extent of developmental disability using a range of measures determining outcome is needed so that parents may be counselled appropriately, and have realistic expectations for their child. Furthermore, better appreciation of the anticipated challenges for the child and family as the child grows and develops following repair enables us better to plan health services that meet the ongoing needs of the family. In particular, knowledge of the impact of this life-threatening condition on the long-term health and well-being of the child and family is critical if appropriate access is to be provided to resources and support so as to optimize the quality of life of those at risk.

Quality of life may be characterized as the perception by the individual of their well-being and general satisfaction with life. Health-related quality of life specifically focusses on aspects of quality that relate to health, such as areas of functioning, which are observable, in addition to the perspective by the patient, or in the case of young children, their proxy, of their well-being and affective experience, which is non-observable. Few studies have described the health and well-being of children with complex cardiac malformations who survive open-heart surgery. Recent studies on adults with such cardiac defects indicate that, overall, state of health and quality of life is good, comparable to that of the general population.[3,4] Conversely, other studies indicate that physical and psychosocial well-being is sub-optimal in adults when compared to controls, even though most do not have moderate-to-severe disability.[5,6] Similarly, recent studies on quality of life in children with congenital cardiac malformations presented conflicting findings. Two qualitative studies on adolescents reported a difficult period of transition, with concerns noted in social, psychological and physical domains, particularly with respect to issues of autonomy and social integration.[7,8] In another recent study, using the PedsQL generic and cardiac modules to describe quality of life in a large cohort of children between the ages of 2 to 18, significant differences were found from controls in all domains except for physical functioning. In contrast, other studies found little difference in quality of life from normative data in this population.[9,10]

Parents of children with chronic health problems are at risk for experiencing ongoing stress, undermining the well-being or overall stability of the families, as well as their ability to cope. Factors contributing to parental distress are variable, but do not appear to be related to the severity of the malformation suffered by their child.[11] Rather, parental stress appears to be more closely associated with the psychosocial adjustment and behavioural skills of the child, albeit that the evidence needs further validation.[12,13]

As part of a prospective study, therefore, we have described the acute and long-term developmental state of a consecutive sample of young infants who underwent open-heart surgery early in life. Our primary objective was to determine their long-term health-related quality of life. In addition, the level of stress of their parents 5 years after first open-heart surgery was used as an indicator of family well-being. We also analysed the predictive value of factors either evident during the initial peri-operative period, or evident at long-term follow-up, so as to identify those who are at increased risk for poor physical or psychosocial well-being or greater parental stress. We selected the period of five years for follow-up as this corresponds to the end of the preschool years, this being the time when children begin formal schooling, and hence representing a point of transition in the development of the child.

## Methods

In this prospective study, we recruited a consecutive sample of young infants with a congenital cardiac malformation admitted for open heart surgery at the Montreal Children's Hospital. We evaluated their developmental state before and after surgery, prior to discharge, and 12 to 18 months following surgery. These findings have been reported previously.[14–17] The cohort has now been reassessed at 5 years of age, at the end of the preschool years. The study was approved by the hospital Institutional Review Board, and written consent was obtained from parents of participants.

Those participating included either newborns with a congenital cardiac malformation admitted to the neonatal intensive care unit and requiring open-heart surgery in the first weeks of life, or infants with a congenital lesion who underwent surgical repair in infancy, with all infants below the age of 2 years, typically less than 6 months. Subjects were excluded if they had any known risks for developmental delay, that included:

- prematurity defined as gestational age of less than 37 weeks
- small for gestational age
- evidence of perinatal asphyxia as defined in Limperopoulos et al., 1999[14]
- identified congenital brain malformations or genetic syndromes associated with delay
- previous open-heart surgery
- hypoplastic left heart syndrome.

We recruited 131 young infants into our cohort at the time of first open-heart surgery. Of these, 16 subsequently died, 18 were lost to follow-up or lived too far away to arrange testing, and 3 refused further follow-up. At 5 years of age, 94 of the cohort were assessed by an occupational therapist, psychologist and/or neurologist using a wide range of measures for outcome. Developmental outcomes are described elsewhere, with additional manuscripts in preparation.[18] In order to ascertain the well-being of the child and their family, we used the Child Health Questionnaire to determine the health-related quality of life of the child, with parental stress being measured using the Parenting Stress Index. Parents were given these standardized questionnaires, available in either English or French, at the time of the follow-up at 5 years, and were asked to complete them at home, returning them in a stamped envelope provided to them.

The Child Health Questionnaire Parent Form, 50-item version, is a generic measure of health-related quality of life for children at or above 5 years of age.[19] This measure quantifies the physical and psychosocial well-being of children, and has been validated for use in children with a variety of chronic health conditions. This norm-referenced standardized measure provides detailed information on 12 specific attributes of health, and provides overall ratings for physical well-being and psychosocial well-being, with a normative mean of 50 and standard deviation of 10. It has excellent internal consistency, reliability and validity.

The Parenting Stress Index is a tool that identifies the magnitude of stress in the parent-child system.[20] We used the 36-item short form in our study. A Likert-type scale, ranging from 1, for strongly disagree, to 5, for strongly agree, is applied to each item, and then grouped into Parent–Child Dysfunctional Interaction subscale measuring, for example, the feelings of the parents for their child, the feelings of the child for their parents; a Difficult Child subscale measuring, for example, the mood of the child, and a Parental Distress subscale measuring, for example, the ability of the parents to enjoy life, as well as a score for total stress. Higher scores indicate greater levels of stress. For this measure, a total score greater than the 85th percentile indicates high stress, whereas a score below the 15th percentile designates a defensive response, with scores in between considered as normal. The instrument has been shown to possess excellent psychometric properties, and has been widely used in studies of outcome for children.[21]

A number of medical, surgical, and developmental characteristics were examined as possible factors associated with greater risk for poor life quality and family stress.

### *Developmental factors*

- preoperative and acute postoperative neurologic examination (see Limperopoulos et al., 2000)[15]
- microcephaly at the time of surgery
- scores for developmental outcome at 5 years, including motor, measured using the Peabody Developmental Motor Scale; cognitive, measured using the Wechsler Preschool and Primary Scale of Intelligence; language, measured using the Peabody Picture Vocabulary Test; behavioural, measured using the Child Behaviour Checklist; and functional levels, measured using the Vineland Adaptive Behaviour Scale and WeeFIM.

### *Medical and/surgical factors*

- cardiopulmonary bypass time
- deep hypothermic circulatory arrest time
- type of cardiac lesion (cyanotic versus acyanotic)
- arterial saturation of oxygen at preoperative assessment
- age at surgery
- days hospitalized in the intensive care unit
- total days hospitalized.

Descriptive statistics were used to characterize performance on the measures. Correlations were used to assess the association between predictors and variables in outcome that are continuous, whereas chi square analysis and t tests were used for categorical predictive variables. Simple and multiple linear regressions were also carried out using the Child Health Questionnaire and Parenting Stress Index scores as outcome variables.

## Results

### *Sample characteristics*

Of 94 subjects who participated in the follow-up, the questionnaires were completed by 49, or 52 percent of parents, with one being a foster parent, the remainder biological parents, and with 81.8 percent completed by the mother. The mean age at follow-up was 64.2 plus or minus 11.3 months, representing 61.5 plus or minus 10.1 months since the first open-heart surgical procedure. Over half of the children (56.5 percent) were attending preschool, whereas 28 percent were in kindergarten, and the remainder were either not yet at school (4.3 percent), or were in the first (6.5 percent) or second (4.3 percent) grades at school. Half of the sample was first-born children. Of 49, 34 had cyanotic lesions. Of the overall group, 46 had defects that were corrected at surgery, whereas only 3 were palliated. Saturations of oxygen were below 85 in 29.5 percent at preoperative neurologic examination. The principal cardiac diagnosis

Table 1. Characteristics of the group.

| **Baseline characteristics at surgery** | **Mean ± SD (range)** |
|---|---|
| Age at surgery (months) | 2.6 ± 4.2 (0.3–26.9) |
| Days in intensive care | 11.0 ± 6.9 (2–33) |
| Days hospitalized | 23.4 ± 18.5 (5–115) |
| Cardiopulmonary bypass (minutes) | 151.6 ± 50.3 (76–301) |
| Deep hypothermic circulatory arrest (minutes) | 18.9 ± 23.5 (0–74) |

was tetralogy of Fallot in 15 infants, while 13 had transposition, 6 had septal defects, and 5 had anomalies of the aortic arch. Less frequent diagnoses included double outlet right ventricle, common arterial trunk, Shone's syndrome, totally anomalous pulmonary venous connection, and various malformations resulting in functionally univentricular physiology. Mean age at surgery was 2.6 months, although 20 out of 49, or 41 percent, had surgery in the newborn period. Table 1 summarizes their characteristics at baseline. Overall, the children of those parents who did not complete the questionnaires, but participated in follow-up assessments, were not significantly different on baseline demographic, medical or surgical variables, such as age at surgery, type of lesion, days hospitalized, or saturations of oxygen, with the exception of acute peri-operative neuromotor assessment, with those with abnormal neuromotor scores more likely not to complete the questionnaires. Children of parents not completing the forms also had lower overall adaptive functioning reflected by a Vineland mean total score of 88.9 versus 95.9 (p equal to 0.038), but were otherwise similar on all other developmental and functional measures when compared to those who completed the questionnaires. There was a trend for parents of children palliated not to complete the questionnaires, with 9 out of 12 in the group of those undergoing palliation not completing (p equal to 0.06); and also for children of non-responders to have a lower full scale intelligence quotient, with a mean of 86.3 versus 95.4 (p equal to 0.079).

### *Well-being of the child and family*

The Child Health Questionnaire was used as an index of health-related quality of life, with a parent as the proxy reporter. Most scored in the normal range (Table 2). This scale is comprised of 12 attributes, or subscales, and overall, the group performed well (See Table 3). Concerns were most often noted in the areas of mental health, such as items related to feeling lonely, nervous, upset, or crying, and general health, reflected by items documenting health in relation to his or her peers, expectations for a healthy life, vulnerabilities to illness, or worry about health. With respect to the general condition of health for this cohort, parents reported that 31.1 percent had problems with attention, 23.9 percent had problems with speech, 21.7 percent had problems with learning, 15.2 percent had asthma, 15.2 percent presented with developmental delay or retardation, 13.0 percent with anxiety, 13.0 percent with behavioural problems, 11.1 percent had chronic allergies, 10.9 percent had visual impairment, 6.5 percent had hearing impairment, 2.2 percent had chronic respiratory problems, 2.2 percent had disturbances of sleep, and 6.7 percent had other chronic health conditions. None of the parents indicated that their child had: chronic orthopaedic problems, chronic rheumatic disease, depression, diabetes or epilepsy.

The Parenting Stress Index was used as an indicator of family well-being. Only just over half, 54 percent, of parents were functioning in the normal percentile range (Table 2). High stress was most often associated

Table 2. Mean scores for health-related quality of life and levels of stress in the family.

| **Outcome measure** | **Mean ± SD (range)** | **Percentage below cut-off*** |
|---|---|---|
| *Child Health Questionnaire* | | |
| 1. Physical summary score | 53.5 ± 5.7<br>(35.7–64.8) | 6.4% |
| 2. Psychosocial summary score | 50.9 ± 7.9<br>(27.7–63.0) | 8.5% |
| *Parenting Stress Index* | | |
| 1. Total score | 52.6 ± 32.3<br>(1.0–99.0) | 27% high stress<br>19% defensive |
| 2. Parental distress subscale | 45.1 ± 31.5<br>(1.0–97.0) | 21% high stress<br>27% defensive |
| 3. Difficult child subscale | 50.9 ± 32.3<br>(1.0–99.0) | 25% high stress<br>17% defensive |
| 4. Parent-child dysfunctional interaction subscale | 50.7 ± 29.1<br>(5.0–99.0) | 15% high stress<br>15% defensive |

*Cut-off for Child Health Questionnaire (as recommended in the manual) is <40. For the Parenting Stress Index, cut-offs used were >85th percentile (high stress) and <15th percentile (defensive).

Table 3. Percentage of children with scores more than one standard deviation below the normative mean for those aged from 5 to 7 years of age on the attributes of the Child Health Questionnaire.

| | |
|---|---|
| Physical functioning | 10.4% |
| Bodily pain | 14.6% |
| Role/social-physical | 6.4% |
| Role/social-emotional, behavioural | 17.0% |
| Self-esteem | 18.8% |
| Behaviour | 18.8% |
| Mental health | 25.0% |
| General health | 35.4% |
| Parental impact-time | 8.5% |
| Parental impact-emotional | 18.8% |
| Family cohesion | 12.5% |
| Family activities | 16.7% |

Table 4. Multivariate regression models that identify variables at baseline predictive of physical and psychosocial well-being.

| Predictor variables | $r^2$ | β | p value |
|---|---|---|---|
| *I. Physical well-being* | | | |
| Preoperative neurologic exam | 0.18 | −5.5 | 0.02 |
| Postoperative neurologic exam | 0.11 | −3.6 | 0.05 |
| *II. Psychosocial well-being* | | | |
| Arterial oxygen saturation levels below 85% | 0.14 | −6.6 | 0.01 |

with the subscale relating to the difficult child, whereas defensive responses were most often noted in the subscale for parental distress. Scores were most likely to be in the normal range for the subscale assessing dysfunctional interaction between the parent and the child.

### *Predictors of quality of life and family stress*

*Physical well-being.* The only predictive variable identified for physical functioning was neurological examination both prior to and following open heart surgery (Table 4). An abnormal baseline peri-operative examination was associated with a lower score for physical well-being. No modification was noted for age at surgery, type of lesion, or the period of deep hypothermic circulatory arrest, although the β changed somewhat, to −4.8, when adjusting for the confounding effect of surgery performed during the neonatal as opposed to the infant periods, but was still significant.

*Psychosocial well-being.* Arterial saturations of oxygen below 85 percent prior to surgery were significantly associated with lower eventual psychosocial well-being (Table 4). Surgery performed in the period of infancy rather than during the newborn period, and the time required for circulatory arrest, both modified the effect, whereas the presence of a cyanotic as opposed to an acyanotic lesion was a confounder. Due to the small size of our sample, however, there is insufficient power to determine which variable is most important in enhancing the strength of the relationship between saturations of oxygen and eventual psychosocial well-being. This relationship will need further exploration in future studies. When examining the predictive value of current developmental state, only child behaviour, measured using the Child Behaviour Checklist, was significantly correlated with psychosocial functioning. The total score was r equal to −0.52 and p equal to 0.0003, with scores for internalizing behaviours of r equal to −0.32, and p equal to 0.03, and for externalizing behaviours of r equal to −0.046, p equal to 0.005. In addition, higher levels of parental stress were associated with lower psychosocial well-being for the child, with r equal to −0.48 and p equal to 0.0009.

*Parental stress.* With respect to baseline characteristics, only preoperative arterial saturation of oxygen of less than 85 percent emerged as a predictor variable for parental stress when dichotomized as highly stressed, above the 85th centile, or in the normal range. Odds ratio from this logistic regression was 4.5, with a confidence interval of 1.05 to 19.02, indicating that the odds of parents being stressed at long-term follow-up increased by a factor of 4.5 if their child was cyanotic prior to the first open heart surgical procedure. Child behaviour at follow-up was also significantly correlated with level of parental stress. The total score was r equal to 0.63, with p less than 0.0001, with scores for internalizing behaviours of r equal to 0.43, p equal to 0.0035, and for externalizing behaviours of r equal to 0.57 and p less than 0.0001. Behavioural problems, therefore, were associated with higher stress. As noted above, parental stress was negatively correlated with the psychosocial well-being of the children.

## Discussion

It can now be expected that almost all children born with a congenital cardiac malformation will survive to adulthood, and therefore there has been a shift in focus from medical morbidities to promoting health in this high-risk population. The World Health Organization has recently emphasized that health encompasses physical, mental, and social well-being, and is not merely the absence of disease or medical condition. Physical health, emotional well-being, psychosocial functioning, and occupational performance are all critical elements of health and functioning, and together contribute to a good quality of life.[10,22–24]

The results of our study indicate that survivors of open-heart surgery performed during infancy, as perceived by their parents, are likely to experience a good quality of life at their entry to schooling. Mean scores were above the normative mean, and very few children exhibited poor physical or psychosocial well-being. Closer examination of the attributes that comprise these summary scores, however, reveal that mental health, specifically social and emotional functioning, and general health were more likely to be suboptimal in this cohort. These findings corroborate those of other studies that indicate that children with congenital cardiac malformations are likely to experience a good quality of life.[9,10] Although the study by Uzark et al.[25] demonstrated that, except for physical functioning, these children were significantly different from healthy children, their sample included children between 2 to18 years of age, and it is conceivable that specific age groups, such as older children, were more likely to have lower scores in the PedsQL instrument, decreasing the overall group mean.

As part of the Child Health Questionnaire, parents were asked to indicate whether or not their child had a variety of conditions relating to health. Overall, our sample had comparable prevalence in 11 of these 18 conditions when compared to norms for the general population of the United States of America, where n equalled 391, and subjects were aged from 5 to 18 years.[19] Parents, however, more often reported anxiety, at 13.0 percent versus 5.2 percent for the general population of children, with similar differences for problems of attention, at 31.1 percent versus 19.4 percent, developmental delays at 15.2 percent versus 2.8 percent, problems with learning at 21.7 percent versus 11.7 percent, and problems with speech at 23.9 percent versus 12.2 percent. This reflects the greater likelihood for developmental and psychosocial difficulties in this population, as perceived by the parents, when compared to healthy children. Problems with vision, and chronic allergies, were less likely to be reported in our sample.

The recent literature on quality of life in children with disabilities indicate that key factors important in achieving a satisfying life include positive mechanisms for coping within the family, the motivation and attitudes of the child, and resources and social supports available to the family. Factors relating to the extent and severity of the medical condition are less important predictors, as children may exhibit a good quality of life in spite of significant developmental deficits.[23,26,27] Children with similar levels of disability may adapt differentially to their deficits, and their families may use different strategies for coping, and exhibit different levels of stress.

Our findings indicate that, overall, acute medical and surgical factors were not strong indicators of the quality of life at 5 years of age. This would support existing evidence on children or adults with congenital cardiac malformations that the severity of the medical condition, reflected by the severity of defect or length of hospitalization, is modestly correlated at best, with outcomes concerning the quality of life.[4,9,23,28] This validates the literature on quality of life on other populations of children with or at-risk for developmental disability.[26,27] Furthermore, presence of delays in developmental areas in our sample did not necessarily contribute to greater risk for compromised health and well-being. It is possible that there is a recalibration of expectations due to chronic illness, resulting in greater acceptance of the developmental limitations of the children.[10] Not surprisingly, behavioural difficulties in the child at entry to school were strongly associated with enhanced parental levels of stress, and the perceptions of the quality of life of their children. In one study, parents of children with cardiac defects themselves reported a lower quality of life than parents of healthy children, in spite of favourable ratings for the quality of life of their children. Parental distress, helplessness, and other stresses were strongly associated with overall family well-being, whereas the severity of the defect, or the level of disability of the child, were not important predictors.[29]

The index of parenting stress identifies systems within the interaction between parent and child that are under stress. High levels of stress can affect the behaviour of the parents, and modulate the outcome for the children. Goldberg et al.[13] examined behavioural problems in preschool children with congenital cardiac defects, cystic fibrosis, or no chronic illness, and found that the total score for the index of stress was a powerful predictor, accounting for almost one-third of the variance in the group with congenital cardiac malformations. They suggest that reduction of parental stress would be an important preventative strategy for children at risk for behavioural difficulties. Our study further supports this strong relationship between parental stress and behavioural difficulties in the child. Our study, however, was not designed to ascertain the causal relationship between parental stress and behaviour of the child, albeit that this relationship is conceivably bi-directional. Reports on parental stress in families of children with disabilities indicate that the severity of disability is not an important factor for risk.[12,11]

We did find that children who had abnormal neurologic examinations in the acute perioperative period were more likely to exhibit lower physical functioning at the age of 5 years. Early clinical examination may thus be an important marker of persisting cerebral injury. We also found that infants with low arterial saturations of oxygen preoperatively were at greater risk for lower psychosocial functioning. This may relate to the long-term organic effects of chronic hypoxia, but may also relate in part to parental tendencies to be overprotective of their infant diagnosed with a cyanotic lesion, which may in turn impact on long-term psychosocial functioning.

Our study was carried out on a small sample, and therefore ideally requires further validation. Just over half of the available sample responded, and although respondents were similar to non-respondents for most of the medical, surgical and developmental variables documented, there was the potential for bias favouring children undergoing biventricular repair rather than palliation and with a trend for somewhat better neurodevelopmental outcomes. Parents acted as proxy responders, given that the children themselves were too young accurately to complete the measures used. For children of this age, parents generally provide a valid appraisal of the quality of life of the child. It should be emphasized, nonetheless, that their responses are likely influenced by their hopes and expectations for their child, and how well they are coping as a family.[19,30] Family functioning has an important influence on the experiences of life and happiness of the child, and therefore it is indeed appropriate and essential that providers of health services consider the parental perspectives when allocating resources for this population of children at high-risk.[22,25]

Comprehensive, longitudinal determination of the outcomes for children benefiting from new medical and surgical interventions is critical if we are to understand better the needs of the child and family through childhood in terms of health and development. Standardized developmental assessments are helpful in identifying impairments that may require targeted interventions to optimize functioning in everyday activities. When feasible, these evaluations should be complemented with determination of the personal perspectives of the children and their parents to best appreciate aspects of health and functioning that are sub-optimal, and may require specific resources targeted at health and social service.[22] Few studies have evaluated the health and well-being of children with congenital cardiac malformations and their families. Our findings would suggest that the health-related quality of life is very good at entry to school of those surviving open-heart surgery, albeit that ongoing stress can be expected in the family, and should be appropriately supported as part of the services offered by providers of care.[29] Children with behavioural difficulties may be less able to adapt to everyday demands, thus affecting their psychosocial well-being. Behavioural problems also contribute to parental stress, further exacerbating family functioning. Interventions such as medications, psychotherapeutic approaches for individuals and groups, and parental support groups are needed to address ongoing behavioural difficulties, but are not routinely part of current medical follow-up.

Better elucidation of the outcomes for the health of both the child and the family is important so as to counsel families effectively, and to provide the anticipatory guidance that is needed to minimize poor quality of life.[6,24] Psychosocial difficulties, and poor coping by the family, are new hidden morbidities for this population at high risk that should influence clinical decision-making and planning of health services in the future, so as to optimize the health and well-being of these children and their families.[25]

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