

## FAMILIES

# Families of children with Down syndrome: What we know and what we need to know

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This paper provides a brief overview of what is currently known about families of children with Down syndrome. In addition, it highlights a number of issues that require further research if we are to have a thorough understanding of the impact of a child with Down syndrome on families as a system and on the individuals who make up that system. Some of these issues include the need for: 1) a more balanced perspective – one that acknowledges both positive and negative aspects of the experience, 2) greater attention to the experiences of fathers, 3) more cross-cultural research and studies focused on cultural perspectives, 4) increased attention to the change in demands for families as the individual with Down syndrome ages, 5) more longitudinal studies, 6) greater variety in methodological approaches, for example greater use of qualitative approaches and observational methods, and 7) an increase in the use of statistical approaches that model change and test hypotheses about predictors of change in both parents and children.

From a family systems perspective, the relational life of families provides the central ecological context in which children are nurtured<sup>[1]</sup>. The family system is influenced by the attributes each individual brings to family relationships as well as by family members' perceptions of those attributes. The birth of a child with Down syndrome is likely to affect the family system in many ways, from the micro level of dyadic interaction to the macro level of the cultural views guiding parent perceptions about a developmental disability.

Much research has indicated that a child with Down syndrome has effects on the family<sup>[2]</sup>. Some effects relate to the likelihood that a child will display the cognitive and behavioural phenotypes considered to be typical of children with Down syndrome. These include specific patterns of strengths and weaknesses in information processing, social interaction, expressive language, receptive skills, motor skills, and motivation<sup>[3,4,5]</sup>. Such patterns and/or the expectations of such patterns may influence the behaviour of caregivers in dyadic interaction with the child. Other effects may be due to increased difficulties and/or diminished satisfactions in the parental role and to parents' reduced opportunities in other spheres of life such as career success. In contrast, positive effects are possible through the parents' identification of a particular purpose in

life, or the development of particular talents that may not have been called upon in other circumstances<sup>[6,7,8]</sup>.

Parental adaptation to a child with Down syndrome has also been studied extensively in relation to parental well-being, especially the psychological reactions of parents. Many studies on parental adaptation to a child with Down syndrome have focused primarily on stress. The literature is often unclear about the precise meaning of stress. At least three interpretations have been applied to research on parental adaptation. Some authors use stress to refer to the emotional responses of parents to the demands of the parenting role (e.g., feeling isolated, entrapped, overwhelmed with responsibility). Others focus on the demands brought about by the child's temperament and behaviours (e.g., demandingness, soothability, activity level). A third approach focuses on parental mental health and psychological functioning (e.g., depression, anxiety, self-acceptance, mastery). This latter approach provides more useful data as it is possible for families to face increased demands but to meet these effectively (see, for example, REF 8). Clearly, however, parenting responsibilities, child demands and psychological outcomes are related.

While there is some evidence that parents of children with Down syndrome experience similar levels of well-being to

those experienced by parents of typically developing children<sup>[9]</sup>, most studies suggest that parents of children with Down syndrome experience lower levels of well-being than parents of typically developing children of similar age (see, for example, REFS 10,11). However, it should be noted that, in these studies, the majority of the parents fell into the non clinical range on instruments measuring psychological functioning (e.g., REFS 11,12). Nevertheless, as Singer pointed out, even mild levels of depression can have detrimental effects on individuals and on their families<sup>[13]</sup>.

Despite these increased difficulties in comparison to families where all children are developing typically, as a group, parents of a child with Down syndrome experience fewer negative effects and more positive effects than parents of children with other disabilities<sup>[14-17]</sup>. Many studies compare parents of children with Down syndrome with those who have a child with autism (e.g., REFS 18,19), a comparison that may increase the likelihood of this outcome. There are also studies which use families of children with other conditions as a comparison group (e.g., REFS 16,17) and the results generally show that parents of children with Down syndrome have higher levels of well-being. In a recent study examining "the Down syndrome advantage," Stoneman reported that the findings generally replicated

the Down syndrome advantage found by other researchers<sup>[20]</sup>. However, once the variance attributable to income was removed, the Down syndrome advantage disappeared. Another possible explanation for the Down syndrome advantage is that families whose children with Down syndrome are functioning more poorly than the average or those who have severe behaviour problems (possibly as a consequence of co-morbid conditions such as autism) may choose not to participate in research (see REF 21).

There have now been several important studies that have collected longitudinal data related to the parenting experience of parents of a child with Down syndrome. Hauser-Cram, Warfield, Shonkoff and Krauss found that over a seven year period (child age 3 years to 10 years) the demands associated with raising a child with Down syndrome increased for both mothers and fathers, and for mothers, this increase was greater than that experienced by mothers of a child with a motor impairment or mothers of a child with a developmental delay of unknown origin<sup>[22]</sup>. At the initial data point (3 years) the mothers of a child with Down syndrome had lower child related demands, but these demands increased so that by age 10 they were higher than those reported by the other two groups. Very similar findings were reported by Most, Fidler, LaForce-Booth and Kelly<sup>[23]</sup> who compared the trajectories of child-related demands in mothers of a child with Down syndrome with that of mothers of a comparison group of children with intellectual disability of mixed aetiology using the child related aspects of the Parenting Stress Index (PSI)<sup>[24]</sup>. The children were between 12 and 15 months on the first occasion, 30 months on the second occasion and were 45 months when the third wave of data was collected. While mothers of a child with Down syndrome reported lower stress levels on the PSI than the mothers in the comparison group at 12 months, when their child was 45 months of age there were no differences. The mothers of a child with Down syndrome showed, at the group level, an increase in stress that was not apparent in the other group. While the groups were initially matched on developmental level, by Time 3 the children with Down syndrome were functioning at a lower cognitive level than the children in the

comparison group.

An increase in stress as the child with Down syndrome ages was also reported by Eisenhower, Baker and Blacher<sup>[25]</sup>. These authors initially saw families when the child was aged 36 months and then collected data again at 48 and 60 months and found an increase in problem behaviour in the children with Down syndrome which was associated with an increase in maternal stress. Both Most and colleagues<sup>[23]</sup>, and Eisenhower, Baker and Blacher<sup>[25]</sup>, suggested that the comparative protective effect often attributed to Down syndrome (in contrast to other types of disability) may apply only in the early years of life. The longitudinal studies indicate that stress levels of parents of children with Down syndrome increase over the early to middle childhood period, suggesting that more research should be directed at delineating moderators of stress beyond the infant and toddler years. As Most and colleagues point out, however, the majority of studies are cross sectional and match children on developmental level at the age being studied<sup>[23]</sup>. If trajectories of development differ, over time, the children will cease to be matched, which may account for the changes in parental status vis a vis the comparison group.

The same association between behaviour problems and parental stress is evident in these parents as in other parents of a child with a disability<sup>[14,16]</sup>. Generally, however, children with Down syndrome exhibit fewer behaviour problems than children with intellectual disability from other causes<sup>[25,26]</sup>, although more than typically developing children and siblings<sup>[26]</sup>. Both the Hauser-Cram and Eisenhower studies reported that the increase in stress levels were related to the increase in child-related demands experienced by parents<sup>[22,25]</sup>.

It is still the case that the majority of research regarding family functioning is conducted with mothers. Several factors contribute to this pattern. One is that mothers continue to take the primary child caring role with respect to children with Down syndrome<sup>[27]</sup>. In an interview study with fathers of a child with Down syndrome that focused on their experiences of parenting, 46% spontaneously stated that their wives took almost all responsibility in relation to the child with Down syndrome<sup>[28]</sup>. Nevertheless, research on fathers of children with Down

syndrome indicates that they are contributing to family life in a variety of ways. Hedov and colleagues found that fathers of a child with Down syndrome took a greater share of the child care (when days off work to care for a sick child were examined) than did fathers in families where all children were developing typically<sup>[27]</sup>. In one of the few studies focusing on fathers' perceptions, Ricci and Hodapp found that fathers of children with Down syndrome reported their child to have more positive personality traits and fewer behaviour problems than fathers of children with other types of intellectual disabilities<sup>[16]</sup>. The older children with Down syndrome, however, were found to be less reinforcing and acceptable to fathers than the younger children.

There are a number of variables that have been found to contribute to parental outcomes, and it seems likely that mothers and fathers respond differently to the pressures associated with raising a child with Down syndrome (see, for example, REF 29). Krauss reported that although mothers and fathers of infants and toddlers with developmental disabilities (including Down syndrome) did not differ in overall levels of stress, their patterns of stress were distinctly different<sup>[30]</sup>. Mothers reported higher levels of stress related to the parenting role whereas fathers reported more stress related to their feelings of attachment to the child. Similarly, Keller and Honig found that maternal stress was more related to the demands of child care, while fathers' stress was more related to the child's acceptability<sup>[31]</sup>. Further support for differences between mothers and fathers is provided by Saloviita, Itälänna and Leinonen<sup>[32]</sup>. They found that, although for both parents a negative view of their situation was most associated with the experience of stress, mothers were most influenced by behavioural problems in the child while fathers were more influenced by their child's low social acceptability. It is clearly important to learn more about fathers' experiences of parenting a child with Down syndrome and to examine their contributions to family life in ways that move beyond the limited possibilities of providers of economic and spousal support.

## Parental satisfaction and self-efficacy

Studies on parenting efficacy focus on parents' confidence in their ability to fulfil the parenting role and successfully nurture children's development. Parenting efficacy incorporates dimensions of confidence in parenting with satisfaction with the parenting role. Greater parenting efficacy has been associated with more positive perceptions of the child<sup>[33]</sup>. Hassall, Rose and McDonald found that parents of a child with intellectual disability who felt satisfied with their parenting role reported lower levels of stress than did those who were less satisfied<sup>[34]</sup>. Gilmore and Cuskelly<sup>[35]</sup> compared the scores of mothers of a child with Down syndrome with normative data they had collected for the Parenting Sense of Competence measure<sup>[33]</sup> and found that, when children were young (between the ages of 4 and 6), mothers' reports of their satisfaction with parenting were not different from those of mothers of children who were developing typically. Confidence in their parenting skills, however, was lower for the mothers of a child with Down syndrome.

Self-efficacy in the parenting role reflects parents' confidence that they have the skills to provide the necessary guidance to their child<sup>[36]</sup>. Laws and Millward suggested that, for parents of a child with Down syndrome, parental identity is tied up with being their child's educator<sup>[37]</sup>. Parental self-efficacy for these parents may therefore be partially based on their child's acquisition of skills. In the study discussed above, Gilmore and Cuskelly found that confidence was not related to competence as measured by an IQ test<sup>[35]</sup>. Measures of IQ, however, may be inadequate to capture increases in skills, and adaptive behaviour measures may be more useful for this purpose.

## Family functioning

An additional view of parental adaptation can be gained by a consideration of family functioning. The importance of family relationships to the well-being of all family members has been highlighted by the review of scientific evidence of early childhood by Shonkoff and Phillips<sup>[38]</sup>. The way in which the family functions may serve as a buffer or promoter of parenting stress. For example, trajectories

in maternal parenting stress of children with developmental disabilities have been found to be reduced in cohesive families (i.e., families in which members feel connected to and supported by each other)<sup>[22]</sup>. Another aspect of family functioning, perceived quality of family life, has recently been investigated. Investigations of quality of life provide a broad view of several domains of family life including health, financial well-being, family relationships, spiritual and cultural beliefs, social support, leisure enjoyment, and community involvement. Brown, MacAdam-Crisp, Wang, and Iarocci investigated the quality of life for families in which a child had Down syndrome or autism in comparison to families of typically developing children<sup>[39]</sup>. Although parents of children with autism reported the lowest overall quality of life, the families of children with Down syndrome reported a lower quality of life in relation to health, financial well-being, social support, and career opportunities (for parents) than those in the comparison group.

## Marital functioning

One of the central relationships within families is that between mothers and fathers. A meta-analysis conducted by Risdal and Singer found that the published data on marital functioning in families with a child with a disability were somewhat skewed by the negatively biased assumptions of researchers, and that when these biases were addressed, the negative consequences for this relationship were substantially less than heretofore thought<sup>[40]</sup>. Van Riper and colleagues found no differences in marital functioning (or family functioning) between families with a child with Down syndrome and comparison families where all children were developing typically<sup>[9]</sup>. Kersh, Hedvat, Hauser-Cram and Warfield also reported that married mothers and fathers of 10-year-old children with motor impairment, Down syndrome or developmental delay reported no differences in their marital quality, although all three groups had lower levels of marital quality than couples in the general population<sup>[41]</sup>. Higher marital quality was related to lower levels of stress and fewer depressive symptoms for both mothers and fathers as well as to greater efficacy for mothers.

In a study of the perceived burden of care of mothers of children with a developmental disability (one third of whom had Down syndrome), Erickson and Upshur found that the mother's perception of the caretaking burden was lighter when the father participated in tasks and provided emotional support<sup>[42]</sup>. Building partially on that study, Simmerman, Blacher, and Baker studied marital satisfaction of parents with a child with severe intellectual disabilities in middle to late childhood<sup>[43]</sup>. They found that mothers' satisfaction with fathers' help, not the actual amount of help provided, predicted both mothers' and fathers' marital adjustment. Fathers' help was most frequently reported to be in the areas of playing, nurturing, discipline and decision-making about services and less frequently in areas of hygiene, dressing, feeding, teaching, therapy and taking a child to appointments. These studies suggest that, although the dyadic relationship between mothers and fathers has multiple dimensions, maternal satisfaction with fathers' participation may guide the relationship.

## The sibling experience

The relationship between siblings is considered to be one of the most enduring relationships within families. There are a number of studies that have examined the impact on siblings of having a brother or sister with Down syndrome, although these generally focus on children who are in middle childhood or older. These studies typically focus on behaviour or other adjustment problems in the sibling and/or on the relationship between the typically developing child and the child with Down syndrome. While there were some initial reports of adjustment difficulties (e.g. REF 44) more recent research has found that the siblings have favourable self-concepts<sup>[45]</sup> and that many believe they have developed additional strengths because of their sibling with Down syndrome<sup>[45,46]</sup>. Also, findings suggest that there are no important differences in the adjustment of the siblings of a child with Down syndrome and children in families where all are developing typically<sup>[26,47]</sup> and that relationships are as good as or better than in these families<sup>[48,49]</sup>. Good sibling relationships are often perceived by mothers as evidence of good parenting<sup>[12]</sup>. We have very little understanding of how parents



accomplish this task, although it is clear that the majority do so.

Some of the challenges for researchers interested in examining the sibling experience include the lack of longitudinal studies with large, diverse samples. Most of the existing sibling studies are cross-sectional studies conducted with small, convenience samples of white, middle to upper-class siblings. Many families of children with Down syndrome have more than one typically developing sibling, so another challenge for researchers in this area is deciding which sibling to include in the study. A final challenge for sibling researchers is the lack of good measures to evaluate the sibling experience, especially measures designed to assess positive outcomes. For a more complete review of the challenges facing researchers interested in the sibling experience, see reviews by Hodapp, Glidden and Kaiser, Stoneman, and Van Riper<sup>[50,51,52]</sup>.

## Positive outcomes for families

When given the opportunity, parents identify a range of positive contributions that the child with Down syndrome makes to family life (see REF 53). Unfortunately, that opportunity is often not provided as researchers have generally focused on the negative consequences experienced by family members. Most relationships comprise both positive and negative aspects, and the dominant quality may change over time as circumstances vary. Family life in families of children with Down syndrome is likely to contain some mix of hassles and uplifts, disappointments and great satisfactions, and it important that the uplifts and satisfactions be considered if an accurate picture of family life is to be gained. These positive aspects of family life are also likely to contribute to the ability to cope with the difficulties families may face<sup>[54]</sup>. Parents report increased feelings of empowerment, personal growth, and a rearrangement of priorities, as examples of the positive changes they attribute to their experience of parenting a child with a disability<sup>[55]</sup>.

Poehlmann and colleagues found that mothers saw their child with Down syndrome as having a number of very positive personal characteristics that acted to maintain and develop connections between family members and with oth-

ers<sup>[53]</sup>. Hodapp, Ly, Fidler and Ricci found no difference between parents of a child with Down syndrome and parents of typically developing children with respect to 'rewardingness'<sup>[56]</sup>. Cuskelly and colleagues found that mothers of a child with Down syndrome reported significantly more reinforcing aspects of their relationship with their child than did mothers of typically developing children<sup>[12]</sup>.

More work on investigating the satisfactions associated with parenting a child with Down syndrome needs to be done. This is important in order that a balanced perspective can be provided to families who are beginning their lives as a family with a child with Down syndrome. It is also important that the broader society is appraised of these experiences as they may contribute to attitude change and to therefore increasing the inclusiveness of our society.

## Connections to community

### Community values

Disability is typically viewed as a burden, and the general community holds fairly negative views about parenting a child with Down syndrome<sup>[57,58,59]</sup>. Naturally, many individuals who become the parent of a child with Down syndrome shared these values prior to the birth of their child. While many parents alter their view after the birth of their child with Down syndrome<sup>[60,61]</sup>, they remain embedded within a community for whom having a child with a disability can be viewed only as a tragedy. This is clearly somewhat of an overstatement – there are many individuals who do hold this opinion and families of a child with Down syndrome will have contact with individuals with a range of views. The impact of this dissonance between family and community values has rarely been examined explicitly; however, the enmeshment experienced by some families of a child with a disability and the realignment of friendships that many parents report are likely, in part, to be due to this mismatch.

At the macro-level, parents are embedded in a set of cultural and often spiritual beliefs. The broader culture in which families live and with which values are shared is likely to play some part in the ways in which parents cope and their level of coping with the increased demands associ-

ated with raising a child with a disability. Belief systems about the perceptions of developmental disability are integral to parents' views of the effect of their child with Down syndrome on their lives<sup>[62]</sup>. The role of spirituality or religiosity in the family is also often central to such perceptions<sup>[63]</sup>. Some cultures are more secular than others and different religious traditions view disability and its meaning quite differently (e.g., REF 64).

Religion is not the only way in which cultures differ with respect to their responses to and understandings of disability. An example of possible cultural effects on coping processes comes from work by Blacher and McIntyre who found that Latina mothers reported higher depression and lower morale<sup>[65]</sup>. However, Latina mothers also reported more positive perceptions of parenting and a more positive impact of the child on their family. The authors speculated that this association may be a product of differing attributions about the cause of the child's behaviour from those of the comparison Anglo-American mothers. Lam and Mackenzie discussed the highly competitive nature of Hong Kong society and the impact this may have on parents of a child with Down syndrome in that milieu<sup>[66]</sup>. Magaña, Seltzer and Krauss found that family problems were more strongly associated with depression for Puerto Rican mothers than for comparison mothers drawn from non-Latina Anglo groups<sup>[67]</sup>.

There is little cross cultural work or work within cultural psychology to guide our understanding of what aspects of culture are supportive or undermining of family functioning when there is a child with a disability in the family. The majority of studies on families of children with Down syndrome are focused on only a subset of families, largely those who are white and living in Western industrialised nations. This gap limits our knowledge about the many ways in which parents adapt to a child with Down syndrome and deserves a priority in setting an agenda for future research.

### Early intervention

While early intervention is usually viewed as an intervention aimed at improving child developmental outcomes, there is general recognition that it is part of a developmental system<sup>[68]</sup> and as such

often has important effects on other family members, particularly mothers. In a review of studies of early intervention (parent training models) with a child with autism McConachie and Diggle identified positive impact upon mothers as one of the outcomes<sup>[69]</sup>. Pelchat, Bisson, Ricard, Perreault and Bouchard found that parents of children with Down syndrome or cerebral palsy who were involved in early intervention had more positive perceptions of their child and of their parenting situation, had lower levels of distress and felt more supported<sup>[70]</sup>. The benefits accruing from involvement with early intervention are likely to reflect a number of processes including the mother's perceptions of progress for her child, the development of self-confidence in her skills for working with her child, and an increased sense of support as a result of sharing her concerns with professionals (see REF 71) and other parents.

In an explicit acknowledgement of the role of early intervention in supporting families, Bailey and colleagues identified five outcomes to aid in determining the effectiveness of early intervention, all of which were aimed at the family functioning rather than at child skill development:

*"(a) families understand their child's strengths, abilities and special needs; (b) families know their rights and advocate effectively for their child; (c) families help their child develop and learn; (d) families have support systems; and (e) families are able to gain access to desired services and activities in their community"*<sup>[72:p.227]</sup>.

Bailey, Scarborough, Hebbeler, Spiker and Mallik conducted a national study of early intervention services in the U.S. and reported that parents had a very positive view of the help provided to their family by early intervention services<sup>[73]</sup>. Most families indicated that early intervention had a significant positive impact on their family; 59% of those sampled stated that their family was "much better" because of early intervention services. Although this study was not based on a randomised design (due to ethical reasons), it was large in both its scope and the diversity of families and early intervention programmes included.

## Support

There is evidence that support by friends, relatives, and health care profession-

als plays an important role in assisting mothers in their role as parents of a child with a disability<sup>[74,75,76]</sup>. Van Riper found that maternal well-being and mothers' perceptions of family functioning were associated with their perceptions of the quality of the support they received from the professionals working with them and their child with Down syndrome<sup>[71]</sup>. We have discussed the important role of perceived spousal support earlier, especially for mothers, and will not repeat this here. Other family members, particularly grandparents, play a central supporting role for many families<sup>[77]</sup>. We know little, however, about their needs with respect to support. In addition, more work on understanding how best to provide support for fathers is clearly needed.

## Employment

Economic resources contribute to individual and family well-being<sup>[78]</sup>, and employment is the major source of these resources for most families. Employment contributes more than merely money to individuals, however. It also provides an important avenue of connection to the community, as well as an arena to demonstrate competence outside the parenting role.

There are a number of studies that have established that mothers of children with a disability are less involved in employment outside the home than are other mothers (see REF 79), although few studies have investigated this area specifically with respect to mothers of a child with Down syndrome. In one of the studies to do so, Hedov and colleagues found that this group of mothers were less engaged in outside work than were mothers of a child without a disability<sup>[10]</sup>. Warfield reported that about two thirds of mothers of 5-year-old children with Down syndrome, motor impairment or developmental delay were employed, and that employment status did not vary with the child's type of disability, although more mothers of children with Down syndrome were employed full time<sup>[80]</sup>. She further found that greater parenting demands related to greater absenteeism at work but not to lower levels of work quality. In contrast, greater interest in work and less work intensity were associated with lower parenting stress.

Thus, the relations among maternal employment, parenting demands, and

workplace expectations are complex, and the needs of working parents of children with Down syndrome and other disabilities demand more investigation. The impact of differences in employment is also not clearly established. Shearn and Todd have identified a number of consequences for mothers including a curtailment of their ambitions<sup>[81]</sup>. The role that employment outside the home (or lack of it) in mothers' lives requires more focused attention.

## Interventions

There appears to be little research on direct interventions aimed at reducing stress or increasing well-being in parents of a child with Down syndrome. In one report Greaves<sup>[82]</sup> compared an eight week group programme teaching from Rational Emotive Therapy<sup>[83]</sup> with teaching related to Applied Behaviour Analysis and a no treatment comparison group. All mothers reported a reduction of stress arising from their responsibilities for the care and management of their child with Down syndrome over the course of the intervention. While there were significant improvements for the Rational Emotive Therapy group on some measures of stress immediately post training in contrast to both comparison groups, no follow-up data were able to be collected.

There is a well established association between the use escape-avoidance coping strategies and poorer outcomes for parents (for example REF 74) It seems that a more sophisticated view of problem solving strategies may be helpful if research is to provide assistance to families with a child with a disability. A closer examination of the helpfulness of which strategies under which circumstances is likely to be productive. There are some circumstances that cannot be changed and it may be less debilitating for parents to avoid thinking about that particular problem and to focus their problem solving on problems that are amenable to change. Both longitudinal studies of the benefits of certain problem solving approaches and systematic intervention studies are required to provide information to guide the practice of those providing direct services

## What do we still need to know?

The review above has identified a number

of issues that require further research if we are to have a thorough understanding of the impact of a child with Down syndrome on families as a system and on the individuals who make up that system. These will be briefly summarised here.

The nature of the association between phenotypic characteristics as displayed by the child and the impact on parents needs to be investigated from the perspective of parental adjustment, not solely from the perspective of the impact on those parenting behaviours likely to influence child development. Consideration needs to be given to parental expectations based on the stereotypic view of children with Down syndrome as well as to the 'true' phenotypic characteristics.

There is an increase in the number of studies that include fathers; however, there is much we do not know about the experiences of fathers in their parenting role. These require further exploration if we are to understand what services and supports would assist them to adjust to the circumstances of having a child with Down syndrome. In addition, a deeper knowledge of their contributions to family functioning would provide a more complete picture of the influences on other family members. It is important that this information is collected from fathers themselves, rather than using maternal views as a substitute. This will require some additional work on the part of researchers as they look for ways to involve fathers in research.

Investigations of family life must take a balanced perspective and allow all aspects to be acknowledged. This includes some concentration on the satisfactions of parenting a child with Down syndrome (or of being a sibling or grandparent). Grandparents are often an important resource for parents and as they are an integral part of the family, they also should be included in research. We know very little about how grandparents respond to having a grandchild with Down syndrome and how this changes over time. There are a number of questions about the most useful ways in which they can be involved in the family that is satisfactory for themselves and helpful for the family with the child with Down syndrome, as well as questions about how they might be better supported in their roles.

The demands associated with parenting a child with Down syndrome appear

to increase with age and it may be that the focus of research should be on these older age groups. The contributors to this increase have not been established, although child behaviour problems, child acquisition of skills and the parental relationship are potential candidates. Families with a child with Down syndrome who also has other conditions that increase the demands on families, such as autism or significant sensory disabilities, need to be included in research if we are to have a complete understanding of the implications for families. If we fail to do this, these families will be even more marginalised, as they will not be represented in the usual picture of families of a child with Down syndrome presented through our work.

Cross cultural research on family adaptation to a child with Down syndrome, as well as investigations that reveal cultural ideology in relation to family adaptation, should be a priority. In these investigations the processes or mechanisms that underpin differences should be the guiding question. Finally, the investigation of direct therapeutic approaches to assisting parents, and other family members, deal effectively with the additional demands associated with having a child with Down syndrome in the family, should be undertaken.

Several issues emerge in consideration of the methodology employed in studies on parental adaptation to a child with Down syndrome. As exemplified in this review, most studies on parental adaptation to raising a child with developmental disabilities are cross-sectional and comparative. The comparisons are made in relation to the parents of typically developing children (e.g., REF 27) or to the parents of children with other disabilities (e.g., REF 14). Such studies have yielded important information about the comparative well-being of parents. Although many of the early studies employed maternal reports as representative of the parent voice, current trends involve the reports of both mothers and fathers (e.g., REFS 22, 84).

Comparative studies usually employ conventional analytic approaches, but the dependent nature of the mother-father data requires more complex analyses. Mother-father pairs are often influenced by each other's responses and behaviours. Models that incorporate both individual

level and dyadic level predictors (or correlates) of outcomes are needed. Moreover, the outcomes themselves are not independent of each other and are more accurately modelled as bi-directional. In a study on the contributions of family resources, parenting challenges, work rewards, and work demands on the parenting stress of both mothers and fathers in two-earner families in which a child had a disability, Warfield addressed the critical issue of data dependency<sup>[85]</sup>. She employed hierarchical linear modelling<sup>[86]</sup> to account for the effects of mother and father reports nested within couple-level data. The analytic approach used in that study is likely to provide a more accurate approach to examining the mother-father dyad and may serve as a model for future work.

Another methodological issue relates to the type of measures selected to investigate parent adaptation. Parent report on scaled instruments has been the method of choice, and Likert scales are the most common response format. Ease in data collection and analysis is one of the advantages of such scales. Nevertheless, Likert scales have been criticised for limiting a respondent's ability to accurately express opinions<sup>[87]</sup>. If respondents have difficulty choosing between two options, they are likely to leave an item blank, producing possible bias in the set of recorded responses. The problems of missing data and dilemmas about ways to address 'missingness' are considered by Schafer, and although the approaches he suggests (e.g., imputation) are preferable to prior approaches (e.g., mean replacement), researchers need to aim for as little missing data as possible<sup>[88]</sup>.

Other than studies of mother-child interaction, other methodologies, such as observational studies, are seldom used in investigations of parental adaptation. Observational studies have been employed with other special populations, however. For example, Jacobs and Fiese observed family interaction during mealtime using a well-developed observational instrument to determine if families of children with asthma who were overweight displayed distinct interactional patterns<sup>[89]</sup>. Observational studies may provide more nuanced information about parental adaptation to a child with Down syndrome.

The field of psychology has been slow



to respond to research designs that deviate from conventional quantitative comparative approaches. Qualitative research studies, though relatively few in number, have added a new dimension to the work on family adaptation to a child with a disability. Drawing from an ecocultural framework which emphasises the use of the 'activity setting' as the unit of analysis, Gallimore and colleagues studied how parents adapt daily routines to accommodate their child with a developmental disability<sup>[90,91]</sup>. Others, such as Harry, have used ethnographic approaches to elucidate ethnic and cultural differences in views of normative development and disability<sup>[92]</sup>. Investigations using qualitative

approaches may offer new and distinct ways of delineating the ethno-theories that mothers and fathers themselves bring to the experience of parenting a child with Down syndrome. Such ethno-theories are critical to a more complete understanding of parent adaptation and may serve to assist in the development of improved interventions to enhance parental functioning.

In addition, population studies deserve priority if we are to ensure that a complete representation of family life is captured. Without this, the experiences of families with a child who is functioning less well than his/her peers with Down syndrome may be overlooked. Finally, longitudinal

studies, although costly and difficult to maintain, have an important place in the study of parent adaptation. Carr's study serves as foundational work for descriptive information on children and parents over time<sup>[93]</sup>. More recent studies (e.g., REF 22) have taken advantage of statistical advances that model change and test hypotheses about predictors of change in both parents and children. The field is now well poised to investigate more thoroughly the mediation and moderation of such trajectories. Such information is critical to a more fully developed science on parent adaptation to a child with Down syndrome.

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