int. j. of developmental disabilities, Vol. 58 No. 3, November, 2012, 159–175

Examining In-Home Care Needs and Work Responsibilities for Parents with Children with A Rare Trisomy Condition

Deborah Bruns and Carly Schrey
Southern Illinois University Carbondale, USA

Balancing in-home care needs and work responsibilities can present many challenges for families. This can be especially true with a family member with a disability. Mothers with children with severe disabilities are often not able to continue working in the job they held before their children's birth or had hoped to attain after becoming a parent. The present study explores parent perspectives related to in-home care needs and work responsibilities for their child with a rare trisomy condition, themselves, and their spouses. Results describe the interplay of children's daily caregiving needs, time management, mothers' change in work status, and spouse's work outside the home. Daily caregiving largely contributed to mothers not working outside of the home due to their children's basic and medically related needs as well as a lack of suitable caregivers. In addition, parents in this study often had to reduce work hours or leave the workforce to care for their child. A call for additional research is offered coupled with implications for service providers working with families with a member with a rare trisomy condition.

keywords Trisomy, care needs, medical caregiving, work responsibilities

Introduction

Balancing in-home care needs and work responsibilities can present many challenges for families. This can be especially true when a family member has a disability (Fidler et al., 2000; Kuhlthau and Perrin, 2001; Trute et al., 2007). In their framework, Summers et al. (2005) draw attention to family functioning related to having a member with a disability. Zuna et al. (2009) also emphasize the interrelationships among family members and varied types of support needs (e.g., emotional, financial). Authors note the need to closely examine the severity of the child's disability and related care needs and their correlation with parent needs and outcomes (Wang et al., 2004).

 DOI 10.1179/2047387712Y.0000000002

Providing daily care for a child with severe disabilities includes bathing, dressing, and feeding. These tasks often extend beyond the early childhood years and, sometimes, require specialized training. For example, Curran et al. (2001) describe caregiving for 16 children with severe disabilities in the UK including cerebral palsy. Mothers discussed their child's daily needs as considerably distinct from children without disabilities in areas such as administering enteral (tube) feedings and positioning needs. Mothers also noted an interest in working outside the home but being unable to due to their children's caregiving needs. McManus et al. (2011) identified similar results. Over 12 000 parents with children with developmental disabilities in the USA completed the National Survey of Children with Special Health Care Needs. Data indicated that caregiving was often regarded as a burden especially with children with complex health needs and limited availability of appropriate health care. Parents in the sample with low socio-economic status, from a minority group or without insurance most strongly expressed the burdens associated with the caregiving needs of their children.

Parents report that two wage earners are preferred but not always possible due to their child's daily care as well as behavioural, care, and/or medical needs (Curran et al., 2001). Available literature highlights the greater likelihood that a parent, most often a child's mother, remains at home to ensure proper care for their child with severe disabilities (Thyen et al., 1999; Powers, 2001; Porterfield, 2002; Parish, 2006; Foster et al., 2011;). Across these studies, mothers indicated employment before their children's birth and reduced opportunities to work outside of the home due to their child's daily caregiving needs. Another finding is described by Shearn and Todd (2000). In their sample of 18 mothers with children with intellectual disabilities in the UK, some mothers worked but reported 'being on call' to respond to their children's needs at all times which negatively affected their employment. Mothers worried about their ability to successfully fulfil the dual responsibilities of meeting their children's caregiving needs and work demands.

Mothers with children with severe disabilities are often not able to participate in the work-related opportunities they either had before the birth of their child or had hoped for before becoming a parent. Mothers with children with Down syndrome discussed this phenomenon as 'role restriction' (Roach et al., 1999). Mothers emphasized caregiving responsibilities as the basis for limited opportunities outside of the home. Additional studies have also identified this factor including Loprest and Davidoff (2004) and Seltzer et al. (2001). Again, samples focused on parents caring for children with more intensive care needs are needed to learn more about 'role restriction'.

One quarter of parents in Kuhlthau et al.'s (2005) sample ($n=750$) in Massachusetts reported reducing work hours to care for their child and 13·2% stopped working completely. Conversely, financial exigency was cited as the reason to continue working for 18% of the sample especially for parents with a child with severe disabilities. Porterfield (2002) and Seltzer and colleagues (2001) described similar findings in their samples. In addition, Lewis et al. (2000) describe changes to full-time work status in order to meet children's caregiving needs. Parents took on one of four patterns (modified single earner, one-and-a-half earners, dual earners,

and flexible dual earners) to support general financial needs and also provide essential basic and medical care for their child with an intellectual disability.

Parish (2006) examined mothers with adolescents with severe disabilities in North Carolina. While work responsibilities sometimes conflicted with caregiving responsibilities, working outside the home was viewed as beneficial to mothers' mental health and was also described as financially necessary. Participation in the work force was also viewed as a welcome opportunity for adult interaction in a study by Shern and Todd (2000). Mothers in their sample shared the belief that time spent away from their child was necessary in order to provide optimal in-home care. In sum, research indicates that balancing the in-home care needs and work responsibilities for parents with a child with severe disabilities is challenging.

As described above, children with severe disabilities comprise a diverse group and impact mothers' ability to work outside the home. Yet, a subgroup not discussed in previous or current studies is in-home care needs and work responsibilities in families with a member with a rare trisomy condition such as trisomy 18 or Edward syndrome (t18), trisomy 13 or Patau syndrome (t13) or trisomy 9 (t9). Trisomy conditions are the most common type of chromosomal anomaly. Trisomy occurs when an extra copy of a chromosome is present due to nondisjunction during meiosis (stage of embryonic cell division). For example, trisomy 18 means there are three copies of chromosome 18. Trisomy 21 or Down syndrome is the most common type followed by trisomy 18 and trisomy 13. Prevalence numbers vary with t18 as the second most common trisomy condition after trisomy 21 (Down syndrome) with one per 3500–6500 live births. Children with t13 are less common (see Brewer et al., 2002; Irving et al., 2011). The majority of existent studies focus on mortality rates reporting many children dying before their first birthday (Brewer et al., 2002; Crider et al., 2008; Irving et al., 2011; Lakovschek et al., 2011; Rasmussen et al., 2003).

Surviving children with rare trisomy conditions typically experience many medical issues include cardiac defects, respiratory complications, and feeding difficulties (Jones, 2006; Bruns, 2010, 2011a, b; Carey, 2010). Most children's developmental skills are similar to those of an infant or young toddler in the areas of communication, social interaction and fine motor skills (Bruns, 2010, 2011a). Recently, an increasing number of studies have reported the outcomes of medical intervention such as cardiac repair surgery (Graham et al., 2004; Kosho et al., 2006; Kaneko et al. 2008; Courtwright et al., 2010; Maeda et al., 2011). These studies point to some affected children living beyond their early childhood years.

What is largely missing in the available literature is a focus on caregiving and work options for parents caring for a child with a rare trisomy condition. Investigating caregiving and work responsibilities is needed to inform professionals working with this unique population. Recently, Bruns and Foerster (2011) reported on the support needs of 20 parents with a child with t18, t13, or t9. An additional study addresses this issue with a larger sample (Janvier et al., 2012). Both studies touch on in-home care needs and work responsibilities, but more information is needed.

The framework of Summers et al. (2005) and Zuna et al. (2009) provide direction related to family functioning amid caregiving and responsibilities outside the home. The present study explores parent perspectives related to their children's in-home care needs and work responsibilities for themselves and their spouses.

Method

Begun in 2007, the Tracking Rare Incidence Syndromes (TRIS) project seeks to increase awareness and knowledge for families and professionals and facilitate improved decision making for optimal services and supports for affected children and their families (http://web.coehs.siu.edu/Grants/TRIS/). To date, data for over 350 children and adults with a rare trisomy condition have been collected through online surveys (TRIS Full Survey for children living 2 months or longer, TRIS Modified Survey for infants living up to 60 days, and TRIS Follow-up Survey which is completed annually after the TRIS Full Survey).

The TRIS project is distinctive in its focus of collecting data over time in multiple areas including family experiences, medical needs, and developmental outcomes.

Instrument development

The results presented here utilized the TRIS Family, Friends and Finances Protocol. The protocol is based on family support items on the TRIS Full and Follow-up Surveys and an in-depth review of the family support literature (e.g. Heiman, 2002; Trute et al., 2007). Development of the TRIS Full Survey has been described elsewhere (Bruns, 2010, 2011a, b). The TRIS Family, Friends and Finances Protocol was developed specifically for this study. As such, no reliability or validity data are available at this time.

The protocol is comprised of seven sections (Child's diagnosis, Support from immediate family members in your home, Support from extended family members, Support from friends, Finances/Sources of income, Support from workplace, and Community supports). Item responses are a combination of closed (Yes/No) and open-ended items in each section. Many items request specific examples. Data presented here are from the Finances/Sources of income section of the TRIS Family, Friends and Finances Protocol (see Bruns and Foerster, 2011 for results from the Support from immediate family members in your home and Support from extended family members sections; see Appendix for TRIS Family, Friends and Finances Protocol items).

Participants

Parents asked to complete TRIS Family, Friends and Finances Protocol met the following criteria: (1) completed the Full TRIS Survey between 1 February 2007 and 31 October 2008; (2) child was living at time of survey completion; and (3) their child has been diagnosed with a subtype of t18, t13, or t9. Children with rarer trisomy types such as trisomy 6p were excluded because of their limited representation in the TRIS project database.

Ninety-one Full TRIS Surveys were completed in the time period. From this group, 68 parents were invited to participate in the present study. Twenty parents (29·4%) completed the protocol. The 20 parents (18 mothers, and 2 fathers) represented seven children with t18 (35%), seven with t13 (35%), and six with t9 (30%). The majority were married ($n=17$, 85%) and completed some post-secondary education ($n=16$, 80%). The mean age of mothers at time of conception

TABLE 1

DEMOGRAPHIC CHARACTERISTICS (N=20)

Characteristic	Mean (± SD)
Child age/status	Survivors (n=20, 100%)
	138·5 months (± 80·7 months)
	Range: 40–370 months
Mother's age at conception	32·9 years (± 6·24 years)
	Range: 22–42 years
Father's age at conception	34·9 years (± 6·38 years)
	Range: 24–46 years
	n (%)
Marital status	
Single	1 (5%)
Long-term relationship	1 (5%)
Married	17 (85%)
Divorced	1 (5%)
Widowed	
Education level	
10–12 years	4 (20%)
13–16 years	6 (30%)
17–20 years	7 (35%)
More than 20 years	3 (15%)
Income level*	
Low	3 (15%)
Medium	14 (70%)
High	3 (15%)

NOTE: *Income level is not presented in dollar figures due to geographic location of project participants: USA (n=17), Canada (n=1), England (n=1), and Scotland (n=1).

was 32·9 years and fathers 34·9 years. Table 1 provides additional aggregate information.

Children were at least 3 years old at the time of the present study. The oldest was 30 years old. Specifically, within trisomy type, the age ranges were 3–15, 6–15, and 4–30 years, respectively for children and adults with t9, t13, and t18. Individuals with t13 and t18 represented primarily full and mosaic types. The group with t9 included with three with the mosaic form of the syndrome, one partial and two individuals with an affected p arm. Individuals with a full trisomy typically have greater care needs than those with mosaic, partial, or p arm types. It is also critical to emphasize that each child has a unique array of daily care and medical needs.

Potential participants were contacted up to three times after the initial invitation. Responses were tracked in an Excel spreadsheet. Beyond the 20 who returned the protocol, an additional six agreed to participate but did not complete

the TRIS Family, Friends and Finances Protocol. Reasons for non-participation included timing of request (holiday season), child-related responsibilities, or family situations such as health issue of a relative. Review of the demographic information of non-participating parents yielded no notable demographic differences.

Procedure

An initial email message was sent to parents with information about the study (n=68). If a parent was interested in participating, a copy of the consent form and the TRIS Family, Friends and Finances Protocol were sent electronically. A reminder email was sent to parents who agreed to participate but did not return the protocol after 2 weeks. Non-respondents were also sent a follow-up email message 2 weeks later.

Initial contacts were staggered in order to encourage responses through frequent communication with one group at a time. Parents with a child with a t9 variant were contacted in November 2008 and parents with a child with a t13 variant were sent the initial email message in December 2008. Finally, parents with a child with a t18 variant were contacted in January 2009.

For the t9 group, 18 parents were initially contacted. Sixteen responded 'yes' for participation. Six surveys were completed and returned for analysis (33%). Twenty-four parents with a child with t13 were contacted. Seventeen responded 'yes'. Seven surveys were returned (41%). In the t18 group, a total of 26 parents were invited to participate. Twenty responded 'yes' with seven surveys returned (35%). The overall completion rate was 29%. Participant information was compiled in Excel spreadsheets (one for each trisomy type) including TRIS project identification number, name, phone number, address, email address, child's name, current age, and trisomy type. Spreadsheets were updated as new information was received.

If a parent preferred a paper copy of the TRIS Family, Friends and Finances Protocol, the materials were sent to the mailing address in the TRIS project database. The majority of completions were electronic (n=17, 85%). All resulting data from the electronic and paper protocols were entered into Word documents organized by protocol item.

Data analysis

Qualitative analyses were used to identify themes. Both authors completed multiple readings of the data. Items with fewer than five responses were removed from analysis. Preliminary themes emerged after multiple readings (Lincoln and Guba, 1985; Huberman and Miles, 1994; Miles and Huberman, 1994). The first author and the TRIS Research Assistant then met and reviewed preliminary themes. Categories within themes were also discussed. Consensus was reached when all categories were mutually exclusive (Patton, 2002; Corbin and Strauss, 2008). A graduate student not affiliated with the TRIS project was asked to review 25% of the returned protocols and generated themes. The three readers reviewed themes and were in agreement and inter-rater reliability was established (Corbin and Strauss, 2008).

Demographic information on the 20 participants was available from the TRIS project database. Parents complete demographic items on the TRIS Full Survey including age at child's conception, marital status, and highest level of education. SPSS 16·0 (2008) was used to create a data set for the resulting data and run aggregate frequencies and descriptive statistical analyses.

Results

The following sections examine parent perspectives of the interplay of in-home care needs and work responsibilities.

Child's caregiving needs

Parents that decided to stay home with their child to be the primary caregiver (n510) did so for a variety of reasons. Their child's high level of care was expressed by several parents 'caring for Ally is basically like caring for a nine month old baby. She has no self- help skills so I take care of everything relating to feeding, clothing and hygiene'. Another parent commented 'Our daughter is three years three months old but she is developmentally more like an 18 month old…I do have to do the basic duties to feed her, clothe her, change diapers etc…'. Consistent with previous studies, parents noted the pervasive and ongoing nature of caregiving (Curran et al., 2001; McManus et al., 2011).

Parents also commented on the lack of suitable in-home caregivers that would be needed if they were to return to work including 'Irene is tube fed, so she'd need nursing care in order for me to go back to work', a mother who made the decision to leave her job as a clerical worker shared '[I] get to be with him every day so I know he's being taken care of'. Another mother remarked, 'I was unable to find a suitable daycare situation for her while I worked so I quit my job and returned to school at night'.

Some parents were able to find suitable child care and return to work (n56). For most, child care was provided inside the home by a nanny, sitter, or a family member rather than a child care centre or other form of group care. A father commented, 'Neil stays at home 95% of the time unless at school or day camps. I drop him off and pick up if he spends the evening at a friend's house. Our house is accessible, so often it's much easier [for childcare]'. 'Although I don't work, I do lots of volunteer work and have a sitter that takes care of my kids' was another response on the topic from a mother with a child with t13 mosaic.

An additional finding was voiced by six mothers stating that the cost of child care was difficult within their family's finances. One mother commented that the cost, '… leaves my income close to zero after paying for childcare'. This statement highlights the trade-off necessary for suitable child care. Several families that were unable to pay for child care noted they received funding for respite care to assist with necessary in-home care.

Flexibility in the workplace was lacking for most parents in the sample. One mother's experience exemplified this, 'I don't work outside the home. It is impossible to find a job that will allow me to be gone for long periods of time if my child is ill or hospitalized or take time off for all his appointments, etc.'. This

mother left her job at as a data conversion operator for the postal service to be a full-time caregiver.

Time management

Finding time for themselves or outside of the home was a problem for approximately half of the sample (n59 of 16 responses, 56%). One mother, who is her child's caregiver said, 'She follows me everywhere. This can be a problem when I need time to myself'. Many parents felt that not having time for themselves was a typical part of parenting, 'Does anyone have enough time for themselves? I get done what needs to be done and move on to the next thing just like everyone else' and 'Time for myself? I gave that up when we decide to have children'. Yet, having time to regroup is necessary to accomplish all that is needed for their child and beyond. One parent commented, 'I need to schedule breaks and getting together with friends…Have to do things when Neil is asleep, at school, or with care provider'.

Beyond seeking time for themselves, parents needed time for other in-home responsibilities. Several comments addressed accomplishing what was necessary to maintain family functioning, 'I found that if I want to get things done, my only real option is to sleep less. I have to stay up late or get up early' and 'the house isn't always clean but everyone is fed and comfortable'. An additional trade-off was described in terms of difficulties completing all that was required for the family when their child's care needs took precedence, 'I just put off things until I feel like getting them done. My son is my first priority'. Another parent added, 'I wish I had a few more hours in which I was not tired' to describe the continuous nature of caregiving.

Interestingly, few parents discussed time with their spouse. Comments, such as the ones presented above, focused on time for child caregiving. The few remarks represented distinct aspects of the issue, 'I have time for myself when she is at school and I have a very supportive husband' and 'Admittedly, time for our marriage has suffered more even than time for myself'.

Change in work status

In addition to in-home care needs, two work-related themes emerged including change in work status and spouse's work outside the home.

Only two parents (n520, 10%) reported not working before having their child with a rare trisomy condition. Representative occupations were secretary, bank manager and purchasing agent and worked between 35 and 50 hours a week. Following the birth of their child, an additional six parents remained home (n58, 40%). This alteration to work responsibilities was also noted in Kuhlthau and Perrin (2001) and Kuhlthau et al. (2005) In addition, one stay-at-home parent stopped working in order to pursue a bachelor's degree in special education.

Slightly over half of parents in the sample responded that their child with a rare trisomy condition affected their primary occupation (n511/19, 58%). After their child was born, mothers reported their primary occupation being a stay at home mom/full-time caregiver or working from home. Conversely, mothers who returned to work often missed many hours of work because of their responsibilities

outside the workplace. One mother commented, 'I missed more work than ever before due to her hospitalizations, illnesses, and I'm not as focused at work due to the stress of the situation in general'. In addition, another mother stated, 'I work part time…because I want to spend as much time as I can with my kids and I need to get my daughter to therapies and work with her 1 : 1'.

Similar to Parish (2006) and Shern and Todd (2000), one mother with a daughter with t9 mosaic noted 'Working does help me keep everything in perspective. If I were home full time I could see myself getting consumed with our daughter's care…This is a nice balance for me…'. This mother switched from full-time to part-time work in marketing. Most responses from other participants did not discuss the option for this type of flexibility in work hours.

Spouse's work outside the home

For parents with a spouse, 15 (n517, 88%) stated that their spouse worked outside the home in fields including retail sales, line technician, roofer, marketing, and stockbroker. It was because of this income that many mothers were able to stay home to provide care for their children with t9, t13, or t18. One mother stated, 'I am always at her beck and call, thanks to my husband's salary'. Another perspective was shared related to financial concerns such as 'one wage is just enough for the basic necessities. Any additional expenses like medical bills or special equipment require extra income'. This mother used to work outside the home as a computer programmer before her child was born. The child's father is an engineer. This mother also notes 'I have no regrets leaving work to take care of her'.

Six mothers indicated a trade-off in that their spouses working outside the home in their reduced or limited involvement in caregiving needs for their child such as 'Watches her if I go out for a couple of hours and is able to do her feed and that's it'. Yet, the converse was also evident, 'He is pretty involved when he is home but he travels three to four days per week. He is very helpful on the weekends'. The dual responsibilities of in-home care and work responsibilities for the spouse were also explained this way, 'When home, he does what is needed but he works 80 hours a week [as a military pediatrician]'. In addition, 12 mothers reported their spouse having little to no involvement in medical caregiving such as tube feedings. 'I make all of the appointments and usually attend the appointments alone'. Another mother added, [He is] not very involved [in playing, interacting, etc.] He works a lot'. This mother's husband works full time as a line technician. Many responses highlight the trade-off between the need for spousal income versus participation in caregiving.

Taken together, the results indicate a variety of experiences for parents with a child with a rare trisomy condition. Financial concerns ranked high in providing necessary caregiving and locating suitable child care. Limited time for one's self was articulated by many in the sample but largely described as a necessary part of being a parent regardless of their child's genetic status. Parents also reported their perspectives about their spouse working outside the home. Additional responses are shown in Table 2.

TABLE 2

REPRESENTATIVE RESPONSES

	Representative responses
Child's caregiving needs	'I have found caring for Taylor challenging not because being a [child with] t18m but also because of people's attitudes.' (trisomy 18 mosaic)
	'As I'm aging and she's growing it's [caregiving] getting a little more challenging for both of us (meaning our daughter and us)...' (full trisomy 18)
	'Her dependency on us will last a lot longer than for most kids' (trisomy 9 mosaic)
Time management	'My daughter's need for countless doctors' appointments and therapy sessions require me to work less than full time. So, I feel like I am not succeeding at anything. Not enough time to give my daughter all the support she needs, not enough money to keep up with expenses of caring for her plus paying childcare on the day I do work' (trisomy 9 mosaic)
	'My house always feels like it is in disarray.' (trisomy 13 mosaic)
	'I just put off things until I feel like getting them done. My son is my first priority' (full trisomy 13)
Change in work status	'I can't devote continuous time in the laboratory…But it's hard to say if things would have been different is Addy has been born without the disability' (full trisomy 13)
	'I have no regrets on leaving work to take care of Irene. Those were precious times' (trisomy 18 mosaic)
	'It wasn't easy finding an employer who was understanding of the time off needed for medical care' (trisomy 9 mosaic)
	'I need to find an employer that will understand the demands of my family' (full trisomy 13)
Spouse's work outside the home	'We need two wage earners or we'd have to rely on less-than-optimal social services' (trisomy 9 mosaic)
	'He does everything; diapers, feeding, bathing, staying up with her at night' (trisomy 18 mosaic)
	'75% me and 25% him [caregiving] because he works full time' (trisomy 9 mosaic)
	'...my husband switched his position at his job so that he could work nights and be home with her during the day' (trisomy 9 mosaic)

Discussion

The results presented here offer a view of in-home care needs and work responsibilities for parents with a child with a rare trisomy condition. Caregiving contributed to not working outside of the home due to their child's basic and medically related needs as well as a lack of suitable caregivers. These sentiments are found in the existent literature including Curran et al. (2001) and Kuhlthau and colleagues (2001, 2005). Working mothers in this sample expressed a sentiment similar to Scott (2010),'…[mothers] experienced great challenges and hardship in trying to meet their children's needs while juggling paid work… working for employers who understood their circumstances…encountered rare empathy and workplace flexibility necessary to juggle care work and wage work' (pp. 690–91). Summers et al. (2005) and Zuna et al. (2009) also address these issues in their framework of family functioning.

In contrast, role restriction as described by Roach et al. (1999) was not a focus of parents' responses, while Lewis et al. (2000), Porterfield (2002), and Shearn and Todd (2000) notion of 'being on call' was expressed by several mothers. This, in turn, made mothers less desirable to employers because they would need to leave or miss work frequently and on short notice. Parents also expressed that they needed to be outside the home for adult contact and time off from caregiving responsibilities.

This need for balance is similar to those voiced in other studies with samples from the USA, Israel, and Canada (Heiman, 2002; Trute et al., 2007; Foster et al., 2011, respectively). Caregiving can become consuming when a child requires around-the-clock assistance with basic care and provision of medical treatments. The child's needs are primary and directly impact her survival. As emphasized in Bruns and Foerster (2011), 'several parents commented on the unease and fearfulness of family members regarding their child' (p. 367), assistance in care is often limited from those who would typically provide caregiving support. Janvier and colleagues (2012) examined parents with children t18 and t13 and found that the majority of mothers did not work and remained home to provide care for their children.

Change in work status was also voiced. Similar to literature focusing on families with a member with a significant disability, parents in this study often had to reduce work hours or leave the workforce to care for their child (Curran et al., 2001; Kuhlthaum and Perrin, 2001; Porterfield, 2002; Seltzer et al., 2001; Wang and Barnard, 2004). As such, the impact of caring for a child with a significant disability extends to all aspects of parenting and ability to work outside the home. For the sample described here, these circumstances are also coupled with the often dire prognoses of a brief lifespan for the child with a rare trisomy condition (Courtwright et al., 2010; Rasmussen et al., 2003). Even if not fiscally sound, parents may not work primarily in order to spend the most time possible with their child.

The last point, though not explicitly expressed by parents in this sample, targets the uncertainty surrounding the child's medical and overall life trajectory and its influence on parent decision-making to work outside the home. A diagnosis of trisomy 18, for example, is often wrought with uncertainty by parents and, oftentimes, professionals. The knowledge base is limited in scope especially for survivors and their families. The results presented here, while similar to some perspectives voiced by parents with children with other genetic syndromes such as Smith Magenis (Fidler et al., 2000; Foster et al., 2011), raise the question of how parents can give voice to a mostly positive outlook within the context of their child diagnosed with a condition viewed, as Walker et al. (2008) states, 'incompatible with life' (p. 15).

Limitations

A concern with the present study is the size of the sample (n=20). Approximately 70% of parents did not return the protocol due to time constraints, child with rare trisomy condition's health and other family-specific factors (Bruns and Foerster, 2011). The results of the study are not generalisable beyond this study. It is unclear

what can be done to counteract these circumstances as some parents were willing to complete the protocol but did not return it after numerous reminders. The authors understood that children's needs received priority over completion of the protocol. Data were aggregated rather than analysed separately by rare trisomy condition due to the small sample. Findings were presented in general terms rather than attempting to identify differences across groups. Beyond Janvier and colleagues (2012), a related concern is the lack of similar studies for comparison.

It is also uncertain if the data collection method affected the sample size. Copies of the TRIS Family, Friends and Finances Protocol were sent electronically as an email attachment. This necessitates the ability to download the document, enter responses and return to the researchers by electronic means. Face-to-face interviews were not viable as participants were from all areas of the USA as well as Canada, England, and Scotland. The prevalence of children with these conditions reduces the likelihood of locating a suitable sample within driving distance of the authors. The time and cost of phone interviews were also prohibitive due to limited TRIS project personnel at the time of data collection. In addition, data shared here were from protocols originally completed in 2008 and 2009. The researchers decided not to contact parents to finish incomplete or omitted items due to the passage of up to several years since completion (Bruns and Foerster, 2011).

Even with the limitations described above, this study offers a much needed perspective on the issues facing families with a member with a rare trisomy condition. The majority of the existent literature focuses on survival status and medical needs (e.g. Brewer et al., 2002; Crider et al., 2008; Courtwright et al., 2010; Vendola et al., 2010). There is a dearth of studies addressing in-home care needs and work responsibilities for this unique population.

Future research

There is a need for additional research on this topic and with this unique group of families. Longitudinal studies are needed to examine changes in in-home care needs and work responsibilities over time. Efforts are underway to analyse data from the TRIS Follow-up Survey to increase understanding of the findings described here. In addition, larger samples of families with children with trisomy 9, 13, and 18 across countries are needed to confirm or disconfirm the themes reported here (Janvier et al., 2012). Data could then be analysed in aggregate and disaggregated forms by trisomy type to draw preliminary conclusions about in-home care needs and work responsibilities. This would also provide reliability and validity data for the TRIS Family, Friends and Finances Protocol. Finally, it would also be an addition to the literature to conduct studies pairing parents such as the ones described here with parents with a child with another genetic condition such as Down syndrome or Smith–Magenis syndrome to determine similarities and differences. Further examination of paternal involvement is also warranted (Simmerman et al., 2001; Skotko et al., 2011).

An additional aspect worthy of study is articulated by Wang et al. (2004). The authors state '…it is possible that a child with severe and multiple disabilities who has no mobility or language might actually be less challenging to a family…'

(p. 90). Research with the group described here offers a means to test this statement in regard to caregiving and financial concerns. The latter has been studied by Kuhlthau et al. (2005) and Loprest and Davidoff (2004) and merits further investigation. The need to keep working to meet financial needs coupled with limited availability of qualified child care options points to a need to better understand how parents navigate these circumstances over the short and long term. In addition, research should address the interplay of parents and their employers in decision making within the context of 'being on call' to address their children's care and medical needs.

Implications

There are a number of implications to be drawn from the results reported here. First, there is a need to increase awareness about families caring for a child with a rare trisomy condition. This needs to be coupled with a move beyond focusing on survival/mortality (e.g. Irving et al., 2011) and medical interventions (e.g., Graham et al., 2004) for this group. The foci must extend to family functioning including areas such as changes in the marital relationship and sibling interactions (Summers et al., 2005; Zuna et al., 2009). At this time, beyond this study and findings reported in Bruns and Foerster (2011), little is known about the unique needs and circumstances these families encounter on a day-to-day as well as long term basis.

This study and others like it also provide information to increase awareness of professionals working with families with children with rare trisomy conditions. This is especially true in light of improved survival rates (e.g. Bruns 2010, 2011a, b). There is a greater likelihood that medical professionals, early intervention providers, teachers, and therapists will encounter these children and their families and understand their unique needs. Family support from professionals will be needed as more children with rare trisomy conditions are surviving the immediate postnatal period and beyond.

As evidenced here, families with children with a rare trisomy condition are in need of more skilled caregivers and respite care providers. The need for additional time for caregiving and other responsibilities was repeatedly shared. In-home care needs required much time, but many parents were unable to find individuals to assist them in this area. Owing to their children's ongoing care and medical needs (Carey, 2010), caregivers must have the requisite training to work with children with t18, t13, and t9. Limited knowledge of these children's needs limits the support professionals can offer families. With longer survival, it becomes imperative to provide training and professional development to a range of professionals across medical, early intervention/education, and therapeutic areas.

Finally, raising awareness of this population of children and their families must extend beyond those directly affected. For example, employers should increase their understanding of the possible effects a child with t18, t13, or t9 will have on the ability of their parents to continue full-time employment. This can only occur when professionals working with these families and parents themselves offer information about day-to-day responsibilities. Telecommuting, as appropriate, could be an alternative to a traditional workplace setting. On a societal level, views of individuals with disabilities, especially those with a poor prognosis combined

with significant care and medical needs, is largely negative. Yet, as indicated here and in the literature, the worth of the child to their family is beyond reproach (Wang et al., 2004). Efforts must be taken to focus on the positive contributions the children described here and with similar conditions offer their families, communities, and beyond.

Conclusion

The perspectives of parents described here bring attention to unique circumstances related to in-home caregiving responsibilities and opportunities for work outside of the home. These results merit continued study and an increase in understanding of professionals interacting with such families.

References

Brewer, C., Holloway, S., Stone, D., Carothers, A. and Fitzpatrick, D. 2002. Survival in trisomy 13 and trisomy 18 cases ascertained from population based registers, J. Med. Genet., 39, 54–56.

Bruns, D. 2011a. Birth history, physical characteristics, and medical conditions in long-term survivors with full trisomy 13, Am. J. Med. Genet. Part A, 155A, (11), 2634–2640.

Bruns, D. 2011b. Presenting physical characteristics, medical conditions and developmental status of long-term survivors with trisomy 9 mosaicism, Am. J. Med. Genet. Part A, 155A, (5), 1033–1039.

Bruns, D. A. 2010. Neonatal experiences of newborns with full trisomy 18, Adv. Neonatal Care, 10, 25–31.

Bruns, D. and Foerster, K. 2011. We've been through it all together: supports for parents with children with rare trisomy conditions, J. Intell. Disabil. Res., 55, (4), 361–369.

Carey, J. 2010. Trisomy 18 and trisomy 13 syndromes, in Management of genetic syndromes, (ed. S. B. Cassidy and J. E. Allanson), 3rd edn; Hoboken, NJ, Wiley-Blackwell, 555–568.

Corbin, J. and Strauss, A. 2008. Basics of qualitative research: techniques and procedures for developing grounded theory, 3rd edn; Los Angeles, CA, Sage.

Courtwright, A. M., Laughon, M. M. and Doron, M. W. 2010. Length of life and treatment intensity in infants diagnosed prenatally or postnatally with congenital anomalies considered lethal, J. Perinatol., 31, 387–391.

Crider, K. S., Olney, R. S. and Cragan, J. D. 2008. Trisomies 13 and 18: population prevalences, characteristics, and prenatal diagnosis, Metropolitan Atlanta, 1994–2003, Am. J. Med. Genet. Part A, 146A, 820–826.

Curran, A. L., Sharples, P. M., White, C. and Knapp, M. 2001. Time cost of caring for children with severe disabilities compared with caring for children without disabilities, Dev. Med. Child Neurol., 43, 529–533.

Fidler, D. J., Hodapp, R. M. and Dykens, E. M. 2000. Stress in families of young children with Down syndrome, Williams syndrome and Smith–Magenis syndrome, Early Educ. Dev., 11, (4), 395–406.

Foster, R. H., Kanotra, S., Stern, M. and Elsea, S. H. 2011. Educational and occupational aspirations among mothers caring for a child with Smith–Magenis syndrome, J. Dev. Phys. Disabil., 23, (6), 501–514.

Graham, E. M., Bradley, S. M., Shirali, G. S., Hills, C. B. and Atz, A. M. 2004. Effectiveness of cardiac surgery in trisomies 13 and 18 (from the Pediatric Cardiac Care Consortium), Am. J. Cardiol., 93, 801–803.

Heiman, T. 2002. Parents of children with disabilities: resilience, coping and future expectations, J. Dev. Phys. Disabil., 14, (2), 159–171.

Huberman, A. M. and Miles, M. B. 1994. Data management and analysis methods, in Handbook of qualitative research, (ed. N. K. Denzin and Y. S. Lincoln). 428– 444; Thousand Oaks, CA, Sage.

Irving, C., Richmond, S., Wren, C., Longster, C. and Embleton, N. D. 2011. Changes in fetal prevalence and outcome for trisomies 13 and 18: a population based study over 23 years, J. Matern. Fetal Neonatal Med., 24, 137–141.

Janvier, A., Farlow, B. and Wilfond, B. 2012. The experience of families with children with trisomy 13 and 18 in social networks, Pediatrics, 130, (2), 293–298.

Jones, K. L. 2006. Smith's recognizable patterns of human malformation, 6th edn; Philadelphia, PA, Elsevier Saunders.

Kaneko, Y., Kobayashi, J., Yamamoto, Y., Yoda, H., Kanetaka, Y., Nakajima, Y., Endo, D., Tsuchiya, K., Sato, H. and Kawakami, T. 2008. Intensive cardiac management in patients with Trisomy 13 or Trisomy 18, Am. J. Med. Genet. Part A, 146A, 1372–1380.

Kosho, T., Nakamura, T., Kawame, H., Baba, A., Tamura, M. and Fukushima, Y. 2006. Neonatal management of trisomy 18: clinical details of 24 patients receiving intensive treatment, Am. J. Med. Genet. Part A, 140, 937–944.

Kuhlthau, K., Hill, K. S., Yucel, R. and Perrin, J. M. 2005. Financial burden for families of children with special health care needs, Matern. Child Health J., 9, (2), 207–218.

Kuhlthau, K. and Perrin, J. M. 2001. Child health status and parental employment, Arch. Pediatr. Adolesc. Med., 155, 1346–1350.

Lakovschek, I. C., Streubel, B. and Ulm, B. 2011. Natural outcome of trisomy 13, trisomy 18, and triploidy after prenatal diagnosis, Am. J. Med. Genet. Part A, 155A, 2626–2633.

Lewis, S., Kagan, C. and Heaton, P. 2000. Dual-earner parents with disabled children: Family patterns for working and caring, J. Family Issues, 21, 1031–1060.

Lincoln Y. S. and Guba E. G. 1985. Naturalistic inquiry; Newbury Park, CA, Sage.

Loprest, P. and Davidoff, A. 2004. How children with special health care needs affect the employment decisions of low-income parents, Matern. Child Health J., 8, (3), 171–182.

Maeda, J., Yamagishi, H., Furutani, Y., Kamisago, M., Waragai, T., Oana, S., Kajino, H., Matsuura, H., Mori, K., Matsuoka, R. and Nakanishi, T. 2011. The impact of cardiac surgery in patients with trisomy 18 and trisomy 13 in Japan, Am. J. Med. Genet. Part A, 155, 2641–2646.

McManus, B. M., Carle, A., Acevedo-Garcia, D., Ganz, M., Hauser-Cram, P. and McCormick, M. 2011. Modeling the social determinants of caregiver burden among families of children with developmental disabilities, Am. J. Intell. Dev. Disabil., 116, (3), 246–260.

Miles M. B. and Huberman A. M. 1994. Qualitative data analysis: an expanded sourcebook, 2nd edn; Thousand Oaks, CA, Sage.

Parish, S. L. 2006. Juggling and struggling: a preliminary work-life study of mothers with adolescents who have developmental disabilities, Ment. Retard., 44, (6), 393–404.

Patton M. Q. 2002. Qualitative research and evaluation methods, 3rd edn; Thousand Oaks, CA, Sage.

Porterfield, S. L. 2002. Work choices of mothers in families with children with disabilities, J. Marr. Family, 64, 972–981.

Powers, E. T. 2001. New estimates of the impact of child disability of maternal employment, Am. Econ. Rev., 91, (2), 135–139.

Rasmussen, S., Wong, L., Yang, Q., May, K. and Friedman, J. 2003. Population-based analyses of mortality in trisomy 13 and trisomy 18, Pediatrics, 111, 777–784.

Roach, M. A., Orsmond, G. I. and Barratt, M. S. 1999. Mothers and fathers of children with Down syndrome: parental stress and involvement in childcare, Am. J. Ment. Retard., 104, (5), 422–436.

Scott, E. K. 2010. 'I feel as if I am the one who is disabled': the emotional impact of changed employment trajectories of mothers caring for children with disabilities, Gender Soc., 24, (5), 672–696.

Seltzer, M. M., Greenburg, J. S., Floyd, F. J., Pette, Y. and Hong, J. 2001. Life course impacts of parenting a child with a disability, Am. J. Ment. Retard., 106, (3), 265–286.

Shearn, J. and Todd, S. 2000. Maternal employment and family responsibilities: the perspective of mothers of children with intellectual disabilities, J. Appl. Res. Intell. Disabil., 13, 109–131.

Simmerman S., Blacher J. and Baker, B. L. 2001. Fathers 'and mothers' perceptions of father involvement in families with young children with a disability, J. Intell. Dev. Disabil., 26, 325–38.

Skotko, B. G., Levine, S. P. and Goldstein, R. 2011. Having a son or daughter with Down syndrome: perspectives from mothers and fathers, Am. J. Med. Genet. Part A, 155A, 2335–2347.

SPSS. 2008. SPSS 16·0 for Windows; Chicago, IL, SPSS, Inc.

Summers, J. A., Poston, D. J., Turnbull, A. P., Marquis, J., Hoffman, L., Mannan, H. and Wang, M. 2005. Conceptualizing and measuring family quality of life, J. Intell. Disabil. Res., 49, (10), 777–783.

Thyen, U., Kuhlthau, K. and Perrin, J. M. 1999. Employment, child care, and mental health of mothers caring for children assisted by technology, Pediatrics, 103, (6), 1235–1242.

Trute, B., Hiebert-Murphy, D. and Levine, K. 2007. Parental appraisal of the family impact of childhood developmental disability: times of sadness and times of joy, J. Intell. Dev. Disabil., 32, 1–9.

Vendola, C., Canfield, M., Daiger, S. P., Gambello, M., Hashmi, S. S., King, T., Noblin, S. J., Waller, D. K. and Hecht, J. T. 2010. Survival of Texas infants born with trisomies 21, 18 and 13, Am. J. Med. Genet. Part A, 152A, 360–366.

Walker, L. V., Miller, V. J. and Dalton, V. K. 2008. The health care experiences of families given the prenatal diagnosis of trisomy 18, J. Perinatol., 28, (1), 12–19.

Wang, K.W. K. and Barnard, A. 2004. Technology-dependent children and their families: a review, J. Adv. Nurs., 45, (1), 36–46.

Wang, M., Turnbull, A. P., Summers, J. A., Little, T. D., Poston, D. J., Mannan, H. and Turnbull, R. 2004. Severity of disability and income as predictors of parents' satisfaction with their family quality of life during early childhood years, Res. Pract. Persons Severe Disabil., 29, (2), 82–94.

Zuna, N. I., Turnbull, A. and Summers, J. A. 2009. Family quality of life: moving from measurement to application, J. Policy Pract. Intell. Disabil., 6, (1), 25–31.

Appendix

Items from the finances/sources of income section of the TRIS Family, Friends and Finances Protocol

1. What was your occupation/profession before you had your child?
   a. On average, how many hours a week did you work?
   b. What career path were you pursuing?
2. What is your primary occupation/profession now?
   a. Is your primary occupation affected by your child's needs? If yes, specifically what types of needs?
   b. What would you like to be doing with your career?
3. If you've entered/returned to the work since your child's birth, who takes care of him or her while you are work?
   a. How do you feel about your child's caregiver(s)/setting? Please provide specific information.
   b. Is it within your family's finances to pay for your child's care?
   c. How many caregivers/settings did you visit, etc. before finding one that was able to meet your child's needs?
   d. How far away is your child's caregiver/setting?
4. If you've remained home with your child, tell me about caring for your child.
   a. What is the most enjoyable part of this role? Please describe.
      (i) Basic care

(ii) Medical care
(iii) Other

b. What is the most challenging part? Please describe.

(i) Basic care
(ii) Medical care including interactions with medical professionals
(iii) Community interactions such as recreation, church, etc.
(iv) Time for yourself
(v) Getting things done for your family

c. Do you think you'll return to work outside of the home?

(i) If yes, what needs to occur first?
(ii) Occur second, third, etc.?

d. Does your spouse or partner work outside the home?

a. What is his/her occupation/profession?
b. On average, how involved is he or she in the following areas?

(i) Basic caregiving
(ii) Medical caregiving
(iii) Playing, interacting, etc.

5. Does your family need two wage earners or is one sufficient to meet financial obligations such as mortgage payment, food, medical bills, gas, etc.? Explain.
6. If both of you work outside the home, has this arrangement 'worked' for your family?

a. Benefits
b. Disadvantages
c. Other thoughts

Notes on contributor

Correspondence to: Deborah Bruns, Department of Educational Psychology and Special Education, Southern Illinois University Carbondale, IL 62901, USA. Email: dabruns@siu.edu

Deborah A. Bruns, Ph.D., Associate Professor, Southern Illinois University Carbondale. Carly Schrey is an Undergraduate student at Southern Illinois University Carbondale.