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Katie M. Hart
Western University

Nicole Neil
nneil@uwo.ca

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Down Syndrome Caregivers' Support Needs: A Mixed-Method Participatory Approach

Katie Hart, M.A., & Nicole Neil, PhD, BCBA-D

Faculty of Education, Western University
1137 Western Road
London, ON
N6G 1G7
Canada
p. 519-661-2111 ext. 84761

Conflict of Interest

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Abstract

Background

The purpose of the study was to explore the support needs of caregivers of children with Down syndrome from their perspective using a mixed - method participatory research approach.

Methods

Concept mapping methodology was used to obtain caregiver perspectives. Twenty - one caregivers answered the question ‘Are parents of individuals with Down syndrome supported, why or why not?’ Caregivers were involved in the analysis of the data through concept mapping procedures.

Results

Sorted data were analysed with multidimensional scaling and cluster analysis. Participants generated eight thematic clusters representing the support needs of caregivers of children with Down syndrome. The themes included online and social support, support system gaps, areas where support is lacking, Down syndrome community support, financial support, advocacy needs, educational support and concerns for community programming.

Conclusions

Themes align with previous research on support needs of parents of children with developmental disabilities. The study highlights the need for more local organisations to offer support that is affordable and accessible for families. Results will support future programme planning for services for individuals caring for those with Down syndrome.

Keywords: Down Syndrome, Caregivers, Support Needs, Concept Mapping, Mixed Methods.

Background

Individuals with Down syndrome display a specific pattern of strengths and challenges, or behavioural phenotype, across different domains of development (Fidler et al. 2009). This profile results in both the individual with Down syndrome and their caregivers to present with unique needs for support (Dykens et al. 1994; Nes et al. 2014). Supports are often needed from birth (Canary 2008), and the provision of this care requires physical, emotional, social and financial resources from the caregiver (Silver et al. 1998; Povee et al. 2012). Increased demand for support and care can affect the overall well - being of caregivers (Nes et al. 2014). Studies focusing on mothers of children with Down syndrome (Hodapp 2007) note concerns for physical health problems (Brehaut et al. 2009), psychological distress and adjustment problems, increased risk for clinical depression and lower levels of life satisfaction compared with those of mothers of children without disabilities (Singer 2006; Bailey et al. 2007; Nes et al. 2014). The additional psychosocial needs are a result of the compounding effects of the behavioural characteristics, co - existing medical concerns and the need children have for long - term care (Pelchat et al. 1999). In order to understand and appropriately address concerns for caregiver well - being, the perspective of caregivers must be captured. Of importance is gaining their perspective related to their needs as a caregiver as well as supports and barriers to having those needs met (Resch et al. 2010). By understanding more about caregiver perspectives, service providers will be better informed and can be better equipped to provide services that meet their needs. The current study, therefore, explores the support needs of caregivers of children with Down syndrome using a participatory research approach.

To improve caregiver well - being, a match between caregiver needs and resources is necessary (Resch et al. 2010). Research identifies support needs that are not currently met for caregivers of children with disabilities including access to information and services, financial barriers to obtaining services, school and community inclusion and family support (Resch et al. 2010). Caregivers of individuals with disabilities have also identified a lack of available respite and counselling services to assist in taking a break to attend to the needs of other family members and their own needs (Murphy et al. 2007; Papageorgiou & Kalyva 2010).

Few studies, however, focus on the specific needs of caregivers of children with Down syndrome. Often caregivers of children with Down syndrome function as control groups in studies exploring the support needs of caregivers of children with other disorders, most commonly autism spectrum disorder (Sanders & Morgan 1997; Siklos & Kerns 2006). These studies point to an inaccurate representation of the support needs of caregivers of children with Down syndrome; they are found to have fewer support needs and cope better by comparison (Hodapp 2007). When caregivers of children with Down syndrome are compared with caregivers of typically developing children, however, they experience higher levels of stress and worse coping (Sanders & Morgan 1997; Siklos & Kerns 2006; Hodapp 2007).

There is also evidence that the types of support needs vary across disability groups and some may be more relevant to families with a child with Down syndrome. For example, Siklos and Kerns (2006) investigated the needs of caregivers of children with autism spectrum disorder with caregivers of children with Down syndrome acting as control participants. Caregivers of children with autism spectrum disorder and Down syndrome reported similar numbers of needs and multiple overlapping unmet needs (e.g. financial support, issues related to the social stigma and need for information). Caregivers of children with Down syndrome also reported distinctly

different needs related to educational supports, community programming and friendship opportunities for their child. This research points out potential differences between families with a child with Down syndrome and other disabilities, reinforcing the need for research focused on Down syndrome.

Within the current literature, there are limited studies that have investigated the support needs specific to caregivers of children with Down syndrome (Hodapp 2007). Even fewer capture caregiver perspectives through qualitative approaches (Povee et al. 2012; Marshall et al. 2015). In one study, Marshall et al. (2015) conducted interviews and focus groups with parents of children with Down syndrome and service providers focusing on the supports and services for perinatal, infant and childhood services in supports. Caregivers of children with Down syndrome reported concern for their child's quality of life as their child grows older, specific concerns for educational planning, social and community supports and long-term health care. This study was limited, in that it only focused on services and supports needed by children birth to age 3. In a mixed-method study of 224 caregivers of children with Down syndrome investigating family functioning, Povee et al. (2012) found that caregivers report a need to have time to pursue their own interests and they identified the importance of respite and emotional support from family and friends. While this study did identify some family supports and needs, it only did so as part of a larger focus on family functioning. More research is needed to adequately capture the perspectives of caregivers of individuals with Down syndrome. This study aims to build on the previous research, by exploring the support needs of caregivers of children with Down syndrome using a participatory research approach. Understanding caregivers' perspectives on support needs is an important step to designing systems to improve family outcomes. Caregivers of children with Down syndrome are well positioned to effectively inform organisations of what

their support needs are, yet little research incorporates their voices. Further, we know of no studies that have employed participatory research approaches to address this topic. There are a number of benefits to participatory approaches including increasing the relevance and rigour of research, increasing the uptake of research results and enhancing empowerment (Turnbull et al. 1998). We address this apparent gap in the literature by interviewing parents on their support needs and engaging them in analysis of the data through Trochim's (1989) concept mapping procedures.

Concept mapping is a structured methodology for organising the ideas of a group into a common framework, or concept map, which can be used for planning and evaluation research (Trochim 1989). It is a mixed-method research tool applying quantitative analysis techniques to qualitative data (Trochim 1989; Jackson & Trochim 2002). Concept mapping is also a participatory approach to research. Participants voice their ideas and are invited to categorise the data and engage in the coding and analysis procedures. Researchers avoid imposing their own biases because the content of the maps is entirely determined by the participant group and not an external analyst's interpretation of the data (Kane & Trochim 2007). The concept mapping procedure includes five steps: project preparation, statement generation, statement structuring, data representation and interpretation of data. Concept mapping is recognised as an effective research tool in planning and programme evaluation research (Kane & Trochim 2007) and has been used in a variety of contexts with various participant populations including adults and children and with individuals with disabilities (Nowicki et al. 2014a, 2014b; Brown et al. 2017; Cleversey et al. 2018; Nowicki et al. 2018).

Concept mapping has not yet been employed to explore the support needs of caregivers of children with Down syndrome and is used in the current study to involve caregivers in the

research process and obtain the perspectives of caregivers of children with Down syndrome on support they currently receive and areas where further support is needed. Caregiver perspectives were gained from individual interviews addressing the research question ‘Are caregivers of individuals with Down syndrome supported, why or why not?’ and participants were involved in the analysis of the data through Trochim’s (1989) concept mapping procedures

Method

Participants

Participants were included if they self-identified as a caregiver of a child with Down syndrome. As the focus of the study was on the experiences of caregivers of children with Down syndrome, participants were excluded if they reported their child had a co-morbid diagnosis of autism spectrum disorder. Participants were recruited in two groups: the initial interview (n = 23) and sorting and rating (n = 10). Kane and Trochim (2007) recommended involvement from 10 to 20 participants for a concept mapping study and note that it is not a requirement that the same participants or the same number complete the sorting and rating tasks. A flow chart of participation is presented in Fig. 1. Initially, 52 caregivers were invited to participate in the interview portion. Of the 52 caregivers emailed, 23 consented to telephone interviews. Two participants were excluded, as they did not self-identify as caregivers. All 21 caregivers from the interview phase were invited to participate in the sorting and rating tasks. Three participants did not complete the sorting task, and four additional participants were recruited via email to participate in the sorting task. Four participants sorting data were excluded from analyses owing to improper completion. Six additional participants were recruited via email. Demographic information was provided by 24 of the study participants and is presented in Table 1.

Procedures

Ethics approval for this study was obtained from the university ethics review board. Participants were recruited through convenience, purposive sampling. Study advertisements were sent via the Canadian Down syndrome organisation's email lists, and agencies offering services to individuals with developmental disabilities advertised the study. Interested caregivers contacted the research team via email. Participants were also recruited from the participant pool of an ongoing survey study by providing their email if they were interested in participating in an optional follow-up interview. Interviews took place over a 2-week period, and participants completed the sorting and rating phases over 4 weeks. Participants were provided compensation for participation with an honorarium of a \$20.00 gift card.

The Interview Phase

A graduate student researcher, completing a degree in counselling psychology including coursework on interviewing, contacted all individuals who provided their email address and sent an overview of the study and description of their participation to each participant. Participants were contacted via telephone by the researcher, and verbal consent was obtained from all participants at the beginning of the interview. All responses were audio recorded with participant consent.

During the interview, participants were asked demographic questions. Questions pertained to the participant's relation to an individual with Down syndrome (mother, father and caregiver), their age, level of education, range of household income and the age of their child with Down syndrome. Participants were also asked to indicate if they lived in a rural or urban setting. Following this, the researcher asked participants the focus prompt question: 'Are caregivers of individuals with Down syndrome supported, why or why not?' Participants were instructed to think of as many ideas and statements as they could to answer the question. The

focus prompt was developed via brainstorming between the researchers (Kane & Trochim 2007) and discussed with a local parent Down syndrome advocacy group parent meeting prior to beginning the interviews. To evoke further responses, the researcher asked follow-up questions as needed such as ‘Can you tell me more about that? Do you have any more thoughts on that? Could you provide me some examples of that?’ The length of interviews ranged from 8 min 23 s to 26 min 58 s (M = 13 min 6 s), similar to that of previous single focus prompt concept mapping studies (Nowicki et al. 2014a, 2014b). Following the interviews, participants were thanked for their participation and informed that they would receive an emailed link to perform the follow-up sorting and rating tasks online.

Data Preparation Phase

Participants’ interviews were transcribed verbatim by the researcher. The researcher extracted all participants’ statements from the recorded interviews that answered the focus prompt question. Two hundred seventy-three statements were generated. Following Kane and Trochim’s (2007) concept mapping steps, authors coded the statements as unique or redundant, and unclear responses were edited. Compound ideas were split and broken into statements with one unique idea. Initial agreement between the two authors on uniqueness/redundant was 90.1%, and discrepancies were resolved through discussion between the authors. As a result, 159 statements were coded as redundant and excluded from the set. Statements were then entered into the web-based Concept System® Global Max© software (Concept Systems Inc. 2019). A final list of 114 unique statements was provided to study participants for the sorting phase. Kane and Trochim (2007) noted that it is ideal to obtain a final data set of approximately 100 statements from the interview data for the sorting and rating tasks so as to ensure that a breadth

of the contributed ideas are represented and a manageable number of statements are provided to participants.

Sorting Phase

Participants were emailed a link and login for the online concept mapping tool (Global Max© software). Participants were instructed to sort the statements into categories in a way that made sense to them and to label each category they created. Participants were informed that they could not put all statements in one pile, they could not group one statement into more than one pile and each statement could not be sorted in its own pile. Instructions were given to participants to sort the statements into categories rather than to sort the statements according to priority or value (Kane & Trochim 2007). Participants completed the task by clicking on a statement to select it and dragging and dropping the statement into ‘on screen’ categories on the virtual desktop.

Rating Phase

Participants were instructed to rate each individual statement according to their level of agreement that the statement was a need that caregivers of children with Down syndrome experienced. The Likert scale ranged from 1 to 5, with a rating of 1 indicating strong disagreement and 5 indicating strong agreement. A numeric value corresponding to the Likert scale was provided in a drop-down tab beside each statement on the virtual desktop.

Data Analysis

Concept System® Global Max© software (Concept Systems Inc. 2019) was used for data analyses. The web-based software program is designed to accomplish the multidimensional and hierarchical cluster analysis procedures used within concept mapping (Kane & Trochim 2007).

Multidimensional Scaling

With the use of the software, a matrix is created for each participant's sorted data, where the number of rows and columns equals the number of statements and a value of 1 is entered when statements are sorted into the same pile (Kane & Trochim 2007). Individual matrices are summed to create a group proximity matrix. Multidimensional scaling procedures are employed to create a two-dimensional point map representing the participants' statement sorting results. The analysis locates each statement as a separate point on the map, and the distances between the statements represent the frequency that participants sorted statements together. Points more proximal to one another are statements that were sorted together more frequently (Jackson & Trochim 2002).

The stress index indicates the degree to which the distances between statements on the map are different from the values in the group proximity matrix (Kane & Trochim 2007). The stress index ranges from 0 to 1, with lower values indicating better fit between the data and the point map. Approximately 95% of concept mapping projects yield stress values between 0.205 and 0.365 (Trochim 1989).

Hierarchical Cluster Analysis

Hierarchical cluster analysis partitions participants' statements from the point map into clusters of statements reflecting similar concepts. Each statement begins as its own cluster, and, at each stage of analysis, statements are merged until all statements are merged into one single cluster. At each stage, a cluster map is produced from the analyses and depicts the groupings of related statements overlaying the original multidimensional scaling point map (Kane & Trochim 2007).

A bridging value is calculated for all statements in a cluster solution and ranges between 0 and 1. The value is an indication of whether a statement was sorted with others that are close to

it on the map or whether it was sorted with statements that are farther away on the map. A low bridging value (0–0.30) represents that a statement was rarely sorted with statements in other clusters. A high bridging value (>0.70) indicates that a statement was often sorted with statements in other clusters. A mean bridging value is also calculated for each cluster; clusters with low bridging values are usually more cohesive, are easier to interpret and indicate that the statements within the cluster were not grouped with other concepts frequently (Kane & Trochim 2007).

There is no calculation to select the correct number of clusters for the final solution (Kane & Trochim 2007). The selection of the final solution is made by examining the clusters that are combined when merging or reducing the solution from a larger cluster solution to a smaller solution (e.g. merging from 20 clusters down to 19 clusters). This is done sequentially and recorded in a ledger of decisions. The decision of the final number of clusters in the solution was made by the researcher reviewing the ledger of decisions and identifying the merger where the researcher's opinion was that the cluster solution would no longer be applicable. In the current study, the solution was first reviewed by the first author and then with the second author.

The concept mapping software identifies 'closest fit' labels for each cluster based on participants' suggestions. Once the final cluster solution was determined, the researcher reviewed the statements in each cluster as well as participants' suggested labels to determine the cluster labels.

Results

An eight-cluster solution was selected (Fig. 2). The number of statements per cluster ranged from 6 to 25 with mean bridging values ranging between 0.15 and 0.88 (Table 2). Participants generated eight thematic clusters including (1) online and social support; (2) support

system gaps; (3) areas where support is lacking; (4) Down syndrome community support; (5) financial support; (6) advocacy needs; (7) educational support; and (8) concerns for community programming. The final stress index of 0.276 in this study revealed that there was a good fit of the data and the point map and that participants were reliable.

Statements from caregivers of children age 0 to 4 years were most frequently sorted into the support gaps cluster, 12 statements of 49 total statements. Statements from caregivers of children ages 5 to 19 years were most frequently sorted into the financial support needs cluster, four of nine total statements. For caregivers of children 10 to 14, statements were most frequently sorted into the support gaps cluster, 12 of 49 total statements. For the caregivers of children 18+, statements were most frequently sorted into the Down syndrome community supports cluster, four of six total statements.

Rating Analysis

Participants rated their level of agreement that the statements were a support need for caregivers of children with Down syndrome. See Table 2 for individual cluster and statement ratings listed in descending order within a cluster. The mean individual statement ratings ranged from 1.57 to 4.83 ($M = 3.54$, $SD = 0.70$). Based on the mean ratings, the clusters most agreed upon as representing needs of caregivers were (1) advocacy needs ($M = 3.74$, $SD = 1.08$); (2) support system gaps ($M = 3.65$, $SD = 0.67$); (3) concerns for community programming ($M = 3.65$, $SD = 0.52$); and (4) online and social support ($M = 3.64$, $SD = 0.54$). These clusters may be key support needs based on general agreement during rating. It is important to note, however, that given the range of the scales (1–5), the mean ratings are not significantly different; that is, the difference between the highest mean rating (3.74) and the lowest mean rating (3.34) is less

than 1. Thus, all the clusters represent support needs, but some may be less representative than others.

Results by Cluster

Cluster 1: Online and Social Support

Statements in this cluster reflected the belief that social support as well as support received via social media was an accessible and beneficial support for caregivers. Many of the statements reflected the idea that participants currently make use of such support (e.g. “Family and friends have of course been great. Like we are pretty connected that way”). Some statements reflected the belief that more could be done to provide social support and some caregivers indicated social support was not a need they experienced. The low mean bridging value ($M = 0.39$, $SD = 0.10$) of this cluster indicated that participants consistently sorted the statements together and had good inter-sorter reliability. Participants’ agreement ratings of the statements in this cluster reflected a general sense of agreement with a mean cluster rating of 3.64 ($SD = 0.54$) and all ratings above 3.

Cluster 2: Support System Gaps

Statements within this cluster focused on caregiver opinions of the service system including the difficulty of accessing supports for their child due to their geographical location, inadequate notification of services, and the limited number of services available. Statements within the cluster reflected differing opinions of whether services were suitable or adequate, for example, ‘I don't think that everybody is getting a similar service’. The statements had low bridging values, revealing participants consistently grouped the statements together even though they represented differing opinions. This cluster had the lowest mean bridging value ($M = 0.15$, $SD = 0.07$) of all eight clusters indicating good inter-sorter reliability. Participants’ ratings varied

among statements, from strongly disagree (min of 1.57) to strongly agree (max 4.57) with a mean cluster rating of 3.65 ($SD = 0.67$).

Cluster 3: Areas Where Support is Lacking

A lack of supports was identified by participants in the areas of initial support from birth, preparedness of staff to assist caregivers, and specific training for caregivers. Additionally, participants indicated a belief that the services that are offered are inconsistent. This cluster had a low mean bridging value indicating consistent sorting of the statements and good inter-sorter reliability. Participants' ratings reflected a range of opinions from strong agreement (max of 4.54) to disagreement (min of 1.63) that the statements were support needs ($M = 3.42$, $SD = 0.82$).

Cluster 4: Down Syndrome Community Support

Statements in cluster 4 were focused on ideas related to the services provided from Down syndrome associations. Statements suggested improvements to Down syndrome associations were necessary, including the creation of associations in communities that are not serviced (e.g., 'Right here locally there is not much [of a Down syndrome community]'). Some statements expressed that services were available and offered adequate support. The low mean bridging value ($M = 0.15$, $SD = 0.08$) for the cluster indicated statements were consistently sorted together. This cluster had the lowest mean rating value of 3.34 ($SD = 0.49$), reflecting participants' general indecision of their agreement of whether the statements were support needs.

Cluster 5: Financial Support

Statements in cluster 5 focused on participants' thoughts on funding sources and financial concerns. Statements included descriptions of what participants use funding for, the types of financial assistance available, the sufficiency of current funding, and concerns related to the

expense of services (e.g., 'We get [funding] each year through [a government organization] and that's supposed to pay for extra support that [my child] needs, we go through it within three months.'). This cluster had a low mean bridging value ($M = 0.29$, $SD = 0.22$) indicating good inter-sorter reliability between participants and consistent grouping of the statements in the cluster. Rating results of the statements revealed, on average, participants experienced indecision about whether the statements reflected a support concern with a mean cluster rating of 3.47 ($SD = 0.66$).

Cluster 6: Advocacy Needs

The theme of advocacy was clear within the statements of cluster 6, with some statements describing the role of the caregiver as an advocate (e.g. 'I feel as your child gets older, you kind of become an expert. It's really up to the individual parents to advocate themselves,') and a need for advocacy opportunities (e.g., 'There is not a way to advocate,'). This cluster had the highest mean bridging value within the eight-cluster solution indicating inconsistent grouping and participants frequently sorting the statements into other clusters. High individual statement bridging values ($M = 0.88$, $SD = 0.07$) indicated that the statements were not a good conceptual fit for the cluster. The statement ratings in this cluster revealed participants consistently agreed with most statements, having, the highest mean cluster rating of all the clusters in the solution ($M = 3.74$, $SD = 1.08$).

Cluster 7: Educational Supports

The statements in cluster 7 focused on the supports provided by educational institutions and included both positive and negative opinions. Some statements suggested more supports are needed ('I think that educationally there is not enough support for kids with DS'), while others indicated the educational supports that were available ('School wise [our child] has had the

support that they've needed, [our child] went from 3 hours of support last year to 6 hours support.'). The high mean bridging value ($M = 0.08$, $SD = 0.09$) for this cluster revealed that the statements were frequently sorted with statements in other clusters. Rating results for cluster 7 revealed that participants had differing opinions about the statements reflecting caregiver support needs. Some statements, on average, were disagreed with, while others averaged strong agreement ($M = 3.39$, $SD = 0.71$).

Cluster 8: Concerns for Community Programming

The statements in cluster 8 reflected caregivers' concerns for the manner in which community programs are offered to caregivers and included concerns for the lack of services in rural areas, opinions that professionals are not adequately connecting caregivers to services, and concern for the lack of childcare available. The high mean bridging value ($M = 0.67$, $SD = 0.15$) for this cluster indicated that statements were frequently sorted with statements in other clusters. Participants consistently rated statements as undecided with a mean rating value of 3.65 ($SD = 0.52$).

Conclusions

The current study investigated caregivers' perspectives on their support needs related to caring for their child with Down syndrome and demonstrated the usefulness of concept mapping as method for involving participants in both the generation of and the analysis of data. With this study, we were able to generate knowledge about support needs and understand the relationships according to a conceptual framework informed by caregivers. Statements generated regarding caregiver support needs overlap with themes identified in previous literature. For example, statements from cluster 1 (online and social support) align with previous research suggesting that caregivers make use of social support both in person and online (Siklos & Kerns 2006; Derguy et

al. 2015). Our work also identifies specific needs described in previous literature, such as the need for more intensive therapies, information and consistent programme offerings (e.g. clusters 2 and 3; Siklos & Kerns 2006; Marshall et al. 2015). Concerns about financial support (cluster 5), educational opportunities (cluster 7) and appropriate childcare (cluster 8) have also been identified in the literature (Kapell et al. 1998; Siklos & Kerns 2006; Povee et al. 2012; Marshall et al. 2015). Lastly, caregivers' statements questioned the preparedness of professionals (e.g. medical professionals) to discuss care options with families (Nicolaidis et al. 2015). This, and previous work, suggests that stigma from health care providers is a common concern and there is a need for health care professionals who are sympathetic, well-informed and good communicators to ensure that individuals with disabilities are not discriminated against or left untreated (Nicolaidis et al. 2015).

The current study also generated novel caregiver support needs. Caregivers expressed the opinion that advocating for their child is left to them and concern was raised regarding the lack of opportunities for caregivers to advocate for their children. Previous research does not cite caregiver concerns related to the lack of Down syndrome associations in smaller, rural cities and the difficulties that caregivers experience in accessing services based on where they reside. The current research highlights the need for organisations to offer support and programming that is affordable and accessible for families outside of urban centres. Sorting results revealed that participants consistently sorted statements in three clusters, clusters 2 (support system gaps), 3 (areas where support is lacking) and 5 (financial support), indicated by low mean bridging values. From participants' rating results, four clusters were most agreed upon as representing needs of caregivers. These included clusters 1 (online and social support), 2 (support system gaps), 6 (advocacy needs) and 8 (concerns for community programming). These clusters may be

key support needs for caregivers on the basis of participants' general agreement during sorting and rating. These include the need for more services in general, programming for varying ages, peer support networks online and in person, advocacy opportunities, funding to access support and the resolution of inconsistencies in programming.

High mean bridging values were revealed for clusters 6, 7 and 8 and suggest that participants may have struggled with the placement of some statements. The statements possibly did not fit the themes of the clusters well or were consistently placed in multiple categories. Within individual clusters, statements reflected differing opinions (as indicated by the range in agreement ratings) on whether they were needs for caregivers. The differences of opinion are likely a result of caregivers' different experiences and personal characteristics such as age, level of education, household income, access to funding and location (Papageorgiou & Kalyva 2010). Support needs may also be influenced by the specific pattern of strengths and challenges children with Down syndrome express (Fidler et al. 2009)

Limitations

Owing to participants' self-selection and self-identification as a caregiver, the results of the current study may not be generalisable to all caregivers of children with Down syndrome. The perspectives of caregivers who do not self-identify as a caregiver were absent from this study. Further, because participants were recruited through community organisations, the study may have missed caregivers who are not participating or marginalised from these services. Because of the sample sizes, we were also unable to make comparisons on the basis of caregiver age or other demographics. Although the concept mapping focus prompt was reviewed by a group of stakeholders prior to conducted the study, the interview process was not piloted and fidelity data were not collected during the interview. Including these quality indicators in future

research will enhance the believability of the results. Participants' ability to accurately complete the sorting task may have been influenced by the number of statements in the study. Kane and Trochim (2007) recommended the use of 100 statements so as not to overburden participants. Additionally, some of the statements were found to contain more than one idea following the researcher's statement structuring processes, which may have resulted in complex statements that could have hindered participants' sorting and rating processes. Lastly, the researcher's editing of the statements and addition of details to provide context could have confused participants and made the connection to the focus prompt difficult to comprehend. However, the current study also provides evidence that strong bridging values can be obtained even with longer or more complicated statements. Participants also expressed the difficulty they experienced using the web-based software to complete the tasks; in-person card sorting with paper cards may be helpful to minimise such issues in the future.

Further, restricting participants' access to the rating task until completion of the sorting task could have aided in the loss of participant data (Kane & Trochim 2007). The sorting task was found to be more time-consuming, labour intensive and demanding of participants' effort and resulted in nine participants failing to complete it or complete it correctly. Subsequently, their results were removed from analyses.

The rating task assessed participants' level of agreement that a statement reflected a need that caregivers of children with Down syndrome experience. Previous concept mapping studies have instead assessed participants' ratings of importance rather than agreement. Importance ratings may be a better indicator of the priority of support needs, as agreement does not necessarily equate to importance. Similarly, because the goal of concept mapping is to organise participants' ideas into a conceptual framework, some information is lost in achieving this goal.

The removal of redundant statements prior to sorting and rating means that the frequency of similar statements cannot be used as an indicator of importance or consensus.

Future Directions

Future research should continue to represent the voices of caregivers and other stakeholders through participatory research approaches. Future research should consider recruitment strategies that generate a diverse sample to explore the differences in support needs on the basis of caregiver and child characteristics. Research addressing such distinctions may produce different caregiver priorities and provide a more holistic representation of support needs. Within our study, more mothers participated than fathers, which is common within the research on caregivers of persons with disabilities (Hodapp 2007). Research should consider recruitment strategies to increase the participation of fathers as well as investigating the perspectives of siblings and extended family members. This approach could consider for the interactions among family members and to approach support needs from a system's perspective (Canary 2008). Future research should also consider importance ratings over agreement to more clearly gather data related to caregiver's opinions of the priority of support needs, as agreement does not necessarily equate to importance.

Implications for Practice

The current study adds to literature in the field of Down syndrome and caregiver support needs by obtaining first-hand perspectives from caregivers of children with Down syndrome and by making use of a participatory research approach. Participatory research allows stakeholders a tool to offer insight into the development of effective policies and services. The results of the current study will support future programme planning that effectively meets the needs of caregivers. The study highlighted novel caregiver support needs not yet reported in the literature

related to caregivers' advocacy needs and the lack of opportunities for caregivers to advocate for their children. Additionally, the study identified the need for more affordable supports to be offered in smaller, rural cities to adequately assist caregivers in accessing services. The key areas of support to address included increasing service offerings, more consistent programming for varying ages of children, the creation of peer support networks (online and in person) and an increase in information on funding opportunities

References

- Bailey D. B., Jr., Golden R. N., Roberts J. & Ford A. (2007) Maternal depression and developmental disability: research critique. *Mental Retardation and Developmental Disabilities Research Reviews* 13, 321–9.
- Brehaut J. C., Kohen D. E., Garner R. E., Miller A. R., Lach L. M., Klassen A. F. et al. (2009) Health among caregivers of children with health problems: findings from a Canadian population-based study. *American Journal of Public Health* 99, 1254–62.
- Brown J., Kapasi A., Nowicki E., Cleversey K. & Salahadin A. (2017) Caregivers of youth with a fetal alcohol spectrum disorder: hopes for them as adults. *Advances in Neurodevelopmental Disorders* 1, 230–9.
- Canary H. E. (2008) Creating supportive connections: a decade of research on support for families of children with disabilities. *Health Communication* 23, 413–26.
- Cleversey K., Brown J. & Kapasi A. (2018) Educational services for youth with fetal alcohol spectrum disorder: caregivers' perspectives. *International Journal of Mental Health and Addiction* 16, 1156–73.
- Concept Systems Inc. (2019) SC Global MAX. Available at:
<https://www.conceptsystems.com/gw/software>

- Derguy C., Michel G., M'bailara K., Roux S. & Bouvard M. (2015) Assessing needs in parents of children with autism spectrum disorder: a crucial preliminary step to target relevant issues for support programs. *Journal of Intellectual and Developmental Disability* 40, 156–66.
- Dykens E. M., Hodapp R. M. & Evans D. W. (1994) Profiles and development of adaptive behavior in children with Down syndrome. *American Journal on Mental Retardation* 98, 580–7. Available at: <https://www-lib-uwo-ca.proxy1.lib.uwo.ca/cgi-bin/ezpauthn.cgi?url=http://search.proquest.com.proxy1.lib.uwo.ca/docview/618483314?accountid=15115>
- Fidler D. J., Most D. E. & Philofsky A. D. (2009) The Down syndrome behavioural phenotype: taking a developmental approach. *Down's Syndrome, Research and Practice* 12, 188–95. Available at: <https://www-lib-uwo-ca.proxy1.lib.uwo.ca/cgi-bin/ezpauthn.cgi?url=http://search.proquest.com.proxy1.lib.uwo.ca/docview/893273983?accountid=15115>
- Hodapp R. M. (2007) Families of persons with down syndrome: new perspectives, findings, and research and service needs. *Mental Retardation and Developmental Disabilities Research Reviews* 13, 279–87.
- Jackson K. M. & Trochim W. M. K. (2002) Concept mapping as an alternative approach for the analysis of open-ended survey responses. *Organizational Research Methods* 5, 307–36.
- Kane M. & Trochim W. M. K. (2007) *Concept mapping for planning and evaluation*. Sage Publications LTD, London, UK.

- Kapell D., Nightingale B., Rodriguez A., Lee J. H., Zigman W. B. & Schupf N. (1998)
Prevalence of chronic medical conditions in adults with mental retardation: comparison with the general population. *Mental Retardation* 36, 269–79.
- Marshall J., Tanner J. P., Kozyr Y. A. & Kirby R. S. (2015) Services and supports for young children with Down syndrome: parent and provider perspectives. *Child: Care, Health and Development* 41, 365–73.
- Murphy N. A., Christian B., Caplin D. A. & Young P. C. (2007) The health of caregivers for children with disabilities: caregiver perspectives. *Child: Care, Health and Development* 33, 180–7.
- Nes R. B., Røysamb E., Hauge L. J., Kornstad T., Landolt M. A., Irgens L. M. et al. (2014)
Adaptation to the birth of a child with a congenital anomaly: a prospective longitudinal study of maternal well-being and psychological distress. *Developmental Psychology* 50, 1827–39.
- Nicolaidis C., Raymaker D. M., Ashkenazy E., McDonald K. E., Dern S., Baggs A. E. V. et al. (2015) Respect the way I need to communicate with you: healthcare experiences of adults on the autism spectrum. *Autism* 19, 824–31.
- Nowicki E. A., Brown J. & Stepien M. (2014a) Children's thoughts on the social exclusion of peers with intellectual or learning disabilities. *Journal of Intellectual Disability Research* 58, 346–57.
- Nowicki E. A., Brown J. & Stepien M. (2014b) Children's structured conceptualizations of their beliefs on the causes of learning difficulties. *Journal of Mixed Methods Research* 8, 69–82.

- Nowicki E. A., Brown J. D. & Dare L. (2018) Educators' evaluations of children's ideas on the social exclusion of classmates with intellectual and learning disabilities. *Journal of Applied Research in Intellectual Disabilities* 31, e154–63.
- Papageorgiou V. & Kalyva E. (2010) Self-reported needs and expectations of parents of children with autism spectrum disorders who participate in support groups. *Research in Autism Spectrum Disorders* 4, 653–60.
- Pelchat D., Ricard N., Bouchard J., Perreault M., Saucier J., Berthiaume M. et al. (1999) Adaptation of parents in relation to their 6-month-old infant's type of disability. *Child: Care, Health and Development* 25, 377–97.
- Povee K., Roberts L., Bourke J. & Leonard H. (2012) Family functioning in families with a child with down syndrome: a mixed methods approach. *Journal of Intellectual Disability Research* 56, 961–73.
- Resch J. A., Mireles G., Benz M. R., Grenwelge C., Peterson R. & Zhang D. (2010) Giving parents a voice: a qualitative study of the challenges experienced by parents of children with disabilities. *Rehabilitation Psychology* 55, 139–50.
- Sanders J. L. & Morgan S. B. (1997) Family stress and adjustment as perceived by parents of children with autism or down syndrome: implications for intervention. *Child and Family Behavior Therapy* 19, 15–32.
- Siklos S. & Kerns K. A. (2006) Assessing need for social support in parents of children with autism and down syndrome. *Journal of Autism and Developmental Disorders* 36, 921–3.
- Silver E. J., Westbrook L. E. & Stein R. E. K. (1998) Relationship of parental psychological distress to consequences of chronic health conditions in children. *Journal of Pediatric Psychology* 23, 5–15.

Singer G. H. S. (2006) Meta-analysis of comparative studies of depression in mothers of children with and without developmental disabilities. *American Journal on Mental Retardation* 111, 155–69.

Trochim W. M. (1989) An introduction to concept mapping for planning and evaluation. *Evaluation and Program Planning* 12, 1–16.

Turnbull A. P., Friesen B. J. & Ramirez C. (1998) Participatory action research as a model for conducting family research. *Research and Practice for Persons with Severe Disabilities* 23, 178–88.

Table 1. Demographics of Study Participants

	n (N = 24)	M	SD
Location			
Rural	4		
Urban	20		
Relation			
Mother	20		
Father	4		
Caregiver Age		45.14	6.99
Caregiver Highest Level of Education Achieved			
High School Diploma	4		
College Diploma	9		
Bachelor's Degree	10		
Master's Degree	1		
Range of Household Income			
25 000- 49 999	1		
50 000- 99 999	11		
100 000- 124 999	5		
125 000- 149 999	0		
150 000+	7		
Age of Child with Down syndrome			
0-4 years	9	9.13	6.52
5-9 years	5		
10-14 years	7		
15-19 years	3		

Table 2: Cluster Statements with Bridging and Rating Values

Cluster and Statement	Bridging Value	Mean Agreement
Cluster 1. Online Social Support	0.39	3.64
102 If there was a formalized network where you could reach out or someone reaches out to you, with someone whose kinda been there done that.	0.38	4.39
107 So, having an online support service I think would be something that would be helpful because then you could come to it where you are	0.38	4.30
3 I feel like some of the challenges we experience [as parents of children with DS compared to typically developing children] might be sort of a step beyond and if there was more targeted DS parent talking to DS parent.	0.53	4.25
35 I would definitely be looking for people to be sounding boards and to chat with.	0.40	4.21
99 It takes time to build those community groups and get people aware and on board with including [my child] in their programs.	0.35	4.09
23 There's a Facebook group that's quite helpful, people post questions and then you can see all the answers.	0.21	3.92
48 It's nice to get people who have been through it to provide their information but if you're going to these [online support groups] always as the person whose been through it providing information, then you don't ever really get ahead of it.	0.40	3.92
1 There is lots of online support, so I was able to join a bunch of online forums through Facebook and was able to connect with [two nearby Down Syndrome Associations].	0.43	3.79
108 The most important thing I found was being part of the [support] group.	0.28	3.78
11 We tend to use the playgroup as our main source of support where we can get together with other parents we can kinda hash out stories and a lot of the kids are the same age and so going through experiences and sort of what works for you and what doesn't. That sort of thing has been really helpful.	0.32	3.67
10 In terms of getting us together, I think it is much harder to build a live community group to gather children together.	0.35	3.63
83 Family and friends have of course been great. Like we are pretty connected that way. Our friends and family have all been fantastic, so that way there is lots of emotional and social support.	0.31	3.52
46 I know [the local DS Association] does things for people with older children as well, I've read a lot of stuff about teenagers or young adults.	0.32	3.50
84 [The local DS Association] have a resource prenatally, to get connected with someone to tell you about their child and their life, living with someone who has DS.	0.35	3.43
67 From our perspective, being relatively educated in the area and knowing how to ask questions we think we are pretty well supported.	0.55	3.39
93 To be honest with you, I found the majority of my support through social media. Things like Instagram. I found so many parents posting what they do on a daily basis, medications you know,	0.21	3.22

	alternative therapies, advice on everything and anything. I got so much invaluable information.		
110	Personally, we don't have any family who live near us or really if they did would be in a position to be helpful where [our child] is concerned [for babysitting].	0.43	3.09
19	[One of my children is gifted and the other is high functioning with DS]. In both worlds I don't really fit into the conversation. It's more difficult to talk about the problems you are having because they seem more insignificant to people who are having more basic issues.	0.55	3.08
9	We don't necessarily see [our child's DS diagnosis] as something that we specifically need help with.	0.59	2.04
Cluster 2. Support System Gaps		0.15	3.65
77	There are gaps [for service and supports] for sure.	0.06	4.57
5	It depends where you are geographically what services and supports you can access easily.	0.11	4.54
39	I don't think that everybody is getting a similar service.	0.08	4.46
21	There are not enough services out there.	0.10	4.29
82	It just seems like an uneven notification of services.	0.05	4.23
81	[Therapy] varies so much and the hands-on therapy that they provide is so limited.	0.15	4.22
64	Like a lot of things with the medical system and support, it takes a long time.	0.15	4.13
104	When you get the services, it's great, but it is so hard to get to the services and that's the issue right now.	0.05	4.13
61	In the more rural communities, you have to go looking for [supports] a little more. It is not as easy to access I would say as I would assume it is like in the cities.	0.27	4.04
78	I feel like [the government agencies] are very hesitant in terms of offering services and they're very selective in terms of who they offer the services to.	0.19	4.04
113	Trying to get speech therapy is a struggle; [our child] gets it a couple times a year for a few weeks.	0.21	3.87
85	I think that we have such a wide spectrum of needs that it probably is difficult to provide support that's going to work for everyone.	0.13	3.70
37	We have only one sort of Children's Centre where kids are assessed as early interventions services and from that we get very little support from them with regards to PT, OT and speech therapy.	0.16	3.67
45	I find that the government run agencies are not extremely helpful.	0.11	3.58
44	I think that our kids aren't served good, they're served systematically. And not personally.	0.12	3.48
111	It's really a hap-hazard slapped together support system.	0.05	3.48
25	We receive supports for specific delayed areas like OT, PT and speech and language that's hosted through [the local] children's centre.	0.30	3.42
49	There [are] a lot of hidden programs.	0.18	3.38
30	For [our] whole region there's currently only one housing navigator offered through the system.	0.17	3.25
47	I think the services that come to us tend to be services originally geared for Autism because that's where the funding is.	0.13	3.25

86	We had [our child] on the list for PT, OT and social worker, [our child] had her own individual worker.	0.26	3.22
58	[The local government agency] has community outreach people to come to your home and talk to you and they do a lot of assessing. I don't feel like that amounts to a whole lot more than time spent.	0.20	3.17
18	[The services and support] tend to be serviced at the lower functioning level.	0.09	2.92
97	Right now [the support groups] are all for the older ages, there's none sort of that interest us directed toward [our child's] age group right now.	0.12	2.61
94	We don't feel that there is a gap [in services].	0.23	1.57
Cluster 3. Areas Where Support is Lacking		0.24	3.42
20	I think it's dependent on parenting experience knowing when to ask for more because it's not necessarily offered.	0.34	4.54
50	There is a waiting list in the schools for speech and language, there's waiting list for PT , there's waiting list for OT.	0.29	4.30
105	What I noticed is that it seems like the support is a bit inconsistent.	0.14	4.26
73	I would have liked to have some more support out the gate.	0.19	3.87
52	I feel as though children with DS are neglected in the sense that they do not get the services that they need, that they require to grow and develop.	0.18	3.75
15	Sometimes there's that feeling where you wish that there were more programs that were just for kids with disabilities so that it wouldn't matter [that they were different].	0.30	3.71
103	If you're at the right place, at the right time, and you get the right information in time, then maybe you'll be okay, if you don't, well that's too bad.	0.18	3.65
6	When we went to meet with the Social Worker we were actually better prepared than they were, because my wife and I had done the research and reached out and talked to people.	0.33	3.50
26	I find that most of the supports you get disappear as soon as [children] start school.	0.21	3.25
12	Anything outside of [DS groups] like the hospitals and Social workers we didn't get very much [support].	0.23	3.22
7	Support for toilet training, I could really use another support group around that particular topic.	0.19	2.79
65	I have accessed lots of psychologist support and things like that, social workers.	0.20	2.09
43	We were lucky enough to go to a daycare centre that had a special needs program with a resource teacher. It just so happened that at that daycare we were at, all the special needs children had DS.	0.42	1.63
Cluster 4. DS Community Support		0.34	3.34
22	I find you build your supports.	0.40	4.21
32	Medically, I found I've been supported.	0.35	3.87
75	I find there's more support early on.	0.23	3.70
34	We have the DS Association which is supportive and offers a lot of events and information, and networking things.	0.42	3.67
38	You have to go to different organizations that aren't just Down Syndrome.	0.36	3.63

63	A lot of the programs are not convenient timewise	0.25	3.43
14	I did feel like it was hard to get support.	0.15	3.38
40	I think there is a lot [of supports] out there now more than ever.	0.30	3.38
71	I think that we have lots of organizations and programs being run and they definitely work for a number of people.	0.40	3.30
92	The worker [our child] had was amazing with providing information, support, what to utilize in the community, that kind of thing.	0.30	3.13
29	So if you look at [our local town] for an example, there's no DS chapter here. All of the programs are run in [the nearby city].	0.45	3.08
51	We've had [...] good community support, our neighbourhood association our church have been pretty good supports.	0.37	3.00
24	Right here locally there is not much [of a DS community].	0.42	2.79
66	[Our child] was showing some behaviour issues so we got in touch with a behavioural therapist. They were very helpful.	0.38	2.13
	Cluster 5. Financial Support	0.29	3.47
114	We are left in the hands of having to outsource these services on our own, privately and have to pay for them out of pocket.	0.38	4.18
13	If there was more funding put towards hiring more therapists that would be really beneficial.	0.31	4.17
36	[Funding] it does help, it gets [my child their] camps and some extra activities and stuff during the year so that's part of the yes of being supported.	0.30	4.17
8	We get Disability tax credits so we're supported that way.	0.36	4.04
28	[What is lacking is] being able to access where extra funding is.	0.03	4.04
60	If there was more funding put towards opening up more spaces in daycares getting more people in there to assist with people that would be really beneficial.	0.29	4.00
95	If you have money there are many more supports available to you.	0.41	3.86
2	There really isn't [funding] that's just not based on income. It would be nice to just get that little break regardless of your income.	0.00	3.71
112	I do think the funds get misspent or it takes too long to get a hold of them.	0.23	3.70
76	There is a lot of supports if you have private insurance.	0.52	3.57
79	We get [funding] each year through [a government organization] and that's supposed to pay for extra support that [my child] needs, we go through it within three months.	0.03	3.52
57	You do have to reroute your money from your other children to your child in need, unfortunately but it's true.	0.16	3.50
98	I think that the respite funding that we get from the government is super helpful, as far as it helps us pay for [my child's] horseback riding.	0.21	3.35
62	[A local government agency] gets lots of funding, well they used to get a lot of funding for the Autism program, and I felt like maybe 80% of their offering was for Autism children only. For all the other children with different disabilities they only got 20% of the offering.	0.28	3.17
59	I find it really frustrating that the government has changed the rules about special needs people working they have to get minimum wage now	0.76	2.88
42	We've utilized big funding for a support person.	0.03	2.42

17	[The local DS Association] programs are expensive.	0.78	2.17
70	We have come up to barriers against [getting our child the disability tax credit].	0.16	2.09
Cluster 6. Advocacy Needs		0.88	3.74
54	We need him to have a community and a life and to be able to do things each day.	0.87	4.83
68	It's really up to the individual parents to advocate themselves.	0.85	4.70
4	It really comes down to the fierceness of the parents.	0.92	4.33
88	I feel as your child gets older, you kind of become an expert.	0.84	4.30
101	Our experience is we've had to go out and make those connections and be very very very proactive. In utilizing and finding what's available.	0.75	4.30
55	Because DS has been around, and its been researched, there is sort of an understanding of the level of disability.	0.92	3.13
89	There is not a way to advocate.	0.93	2.10
96	I think having a [Down Syndrome] association, you're kind of singling out DS kids. I want [my child] to be in a world where [they] are going to function along with every other person in the world, not just DS people.	1.00	1.96
Cluster 7. Educational Supports		0.67	3.39
91	I especially worry about when [my son] graduates from high school.	0.86	4.36
27	Where education is concerned, a lot of it falls on us as the parent.	0.69	4.25
33	I think that educationally there is not enough support for kids with DS.	0.64	4.25
106	What I have experienced, not one teacher in the [local school board] has been specifically taught how to teach kids with DS.	0.58	3.91
100	I wish [the school board] would listen to us a little bit more and give [our child] a little bit more chance to do things their way.	0.57	3.68
31	I'm not really sure how things will look once we go to school. I don't know how supported kids are in school.	0.70	3.46
16	If we have issues with [my child] at school [a local organization] will come and advocate with us.	0.80	2.96
53	[Our worker] is helping us with transitioning to elementary school.	0.67	2.96
90	I think the expectations [at our school] are a little bit high that [our child] needs to follow along with everybody else.	0.72	2.74
74	We didn't really feel like we had the option of an inclusive classroom because [our child] wouldn't have the support [our child] needed if we sent [them] there.	0.55	2.43
72	School wise [our child] has had the support that they've needed, [our child] went from 3 hours of support last year to 6 hours support.	0.66	2.35
Cluster 8. Concerns for Community Programming		0.67	3.65
41	We are expected to be not only the parent of the child but also their therapist for all of those specialty therapies as well.	0.56	4.33
87	I don't think that the medical community pushes enough awareness of DS when mothers are first diagnosed, or babies are first diagnosed.	0.94	4.04
109	From my experience with children with DS, it feels like sometimes they kinda just fit in the middle, so they're not disabled enough to go	0.61	3.70

	into certain classes and things like that, but then they're not necessarily able to be a part of a regular things.		
80	My big peeve about a lot of stuff is that babysitting should be part of the package when you are going to information stuff to make it easier for parents to go.	0.78	3.65
69	[Pediatricians and doctors] are not connecting you the way they need to.	0.50	3.52
56	We go to a small community rural school. There's just not a lot of programs in there, I think, that they let [our child] sort of explore.	0.65	2.67

Figures

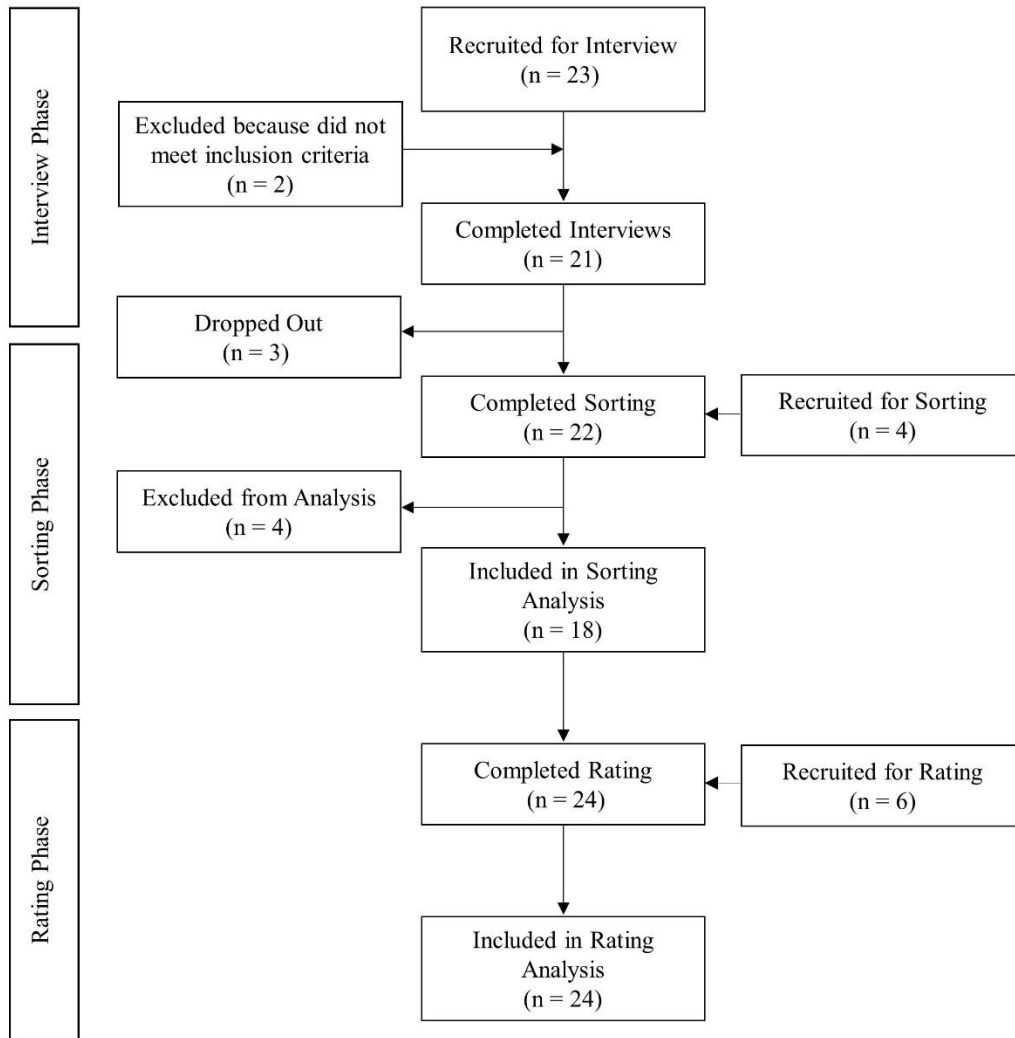


Figure 1. Participant flow chart.

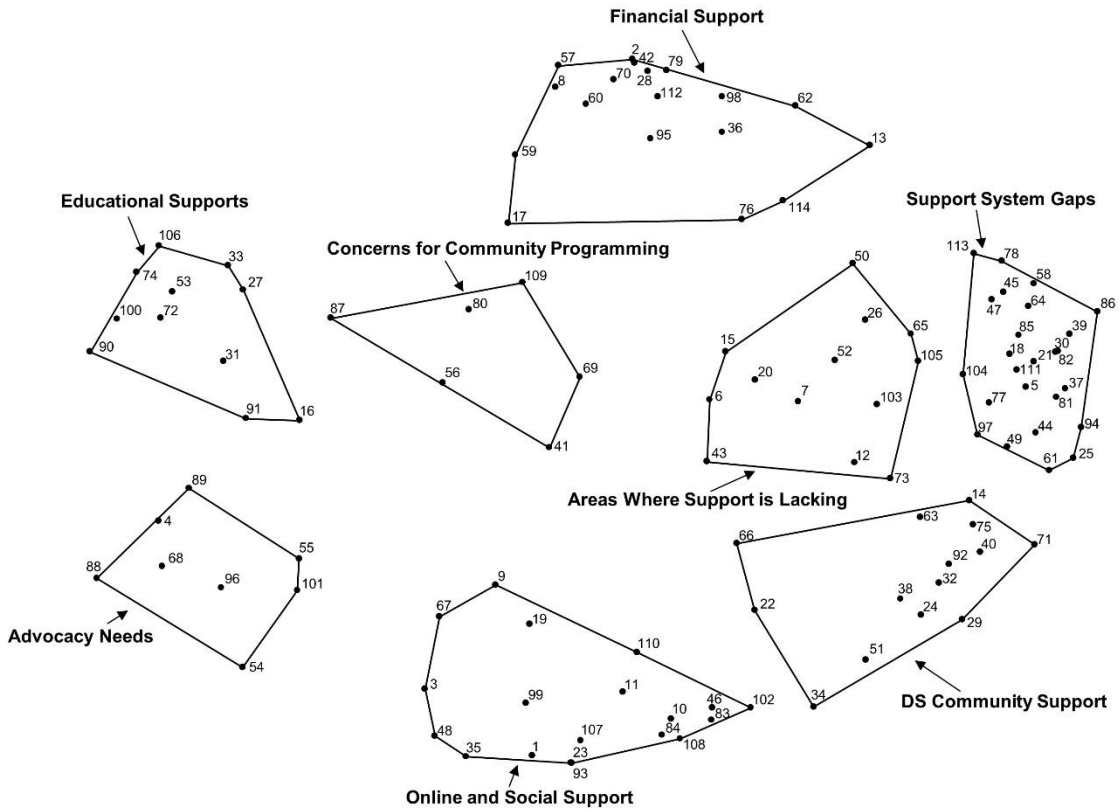


Figure 2. Concept map of statements in response to “Are caregivers of individuals with Down Syndrome supported, why or why not?”