

Family functioning as a constituent aspect of a child's chronic illness

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Abstract

This study explored how family functioning may contribute to trace a child's illness trajectory. We conducted semi-structured interviews with 33 parents of children in care at a hospice in northern Italy. We also examined the medical records of the children, and interviewed the physician who cared for them. Data analysis was based on the grounded theory approach. Different illness progressions corresponded to the different ways with which families experienced the illness: possibility, focus on illness, denial, and anger. Clinical interventions should involve the whole family and take into account their role in the construction of illness trajectories.

Key words

Children, chronic illness, family, grounded theory, qualitative methods

Improving medical care has expanded the lifespan of children with progressive, life-threatening illnesses. In Italy, approximately three million children live with chronic health conditions (Italian Pediatric Society, 2012). Many of these children survive for long periods with complex chronic conditions, characterized by the progression of the illness to a point that requires specialized and time-consuming care. The terminal phase, however, may be months or even years away. The continuous care required by these conditions implies an enormous engagement by families and health systems. Hospices were created to assist children and their families, but the recent trend toward home care increases the involvement of the families.

Many studies have focused on the impact of the child's illness on the family (Hopia, Paavilainen, & Paivi, 2005; Katz, 2002; Moola, 2012) and on the adaptation process and coping strategies families use (Garro, 2004; LaMontagne, Hepworth, Salisbury, & Riley, 2003). In contrast, few studies have explored the influence of family on an individual's response to illness (Knafl & Gillis, 2002). In this introduction we present only the literature in this area because the aim of our study was to explore how family functioning may contribute to the development, maintenance, and progression of the child's illness.

The family as producer of health and illness

In the Family Systems Illness Model, Rolland (1987) emphasized the intertwining of two evolutionary threads: the illness and the individual family life cycles. Referring to the three major disease phases (i.e. crisis, chronic, and terminal), he outlined an understanding of the ever-changing needs and requirements of the patient, the couple, and the family system over the course of the family life cycle.

Patterson, McCubbin and Warwick (1990) found that parents' involvement in activities that enhanced self-esteem helped manage psychological tension, and provided

social support benefits for the health of a child with cystic fibrosis. Wong and Heriot (2008) pointed out that the hope and despair parents had for the future of their child and parental coping styles were important predictors of parent and child adjustment. Other studies heightened that greater family cohesion and less family conflict were linked to better adjustment in children with chronic illness (Mitchell et al., 2007; Perrin, Ayoub, & Willet, 1993; Thompson et al., 1999). By contrast, less cohesiveness and expressiveness and more control in families were associated with internalizing behavioral symptoms (e.g., anxiety and depression). Greater family conflict and less encouragement of independence were associated with externalizing behavioral symptoms (e.g., acting out behaviors) (Morris et al., 1997).

In addition, family roles, communication, and effective responsiveness were indicated as involved in predicting child behavioral functioning. Caregivers who did not involve children in family responsibilities and who treated their children as more vulnerable may have led these children to experience more difficulties with internalizing behaviors. Alternatively, these children may have been perceived as less likely to complete responsibilities because of symptoms related to internalizing behaviors. Family communication also influenced internalizing behaviors, but again the direction was not clear: poor family communication may have led children to be more reserved, or vice-versa (Piazza-Waggoner, Adams, Muchant, Wilson, & Hogan, 2006).

The main limitation of these studies was they observed correlations between family factors (usually assessed by the Family Environment Scale) and the child's psychosocial adjustment (variously assessed by different behavior problem checklists or social competence scales). This prevented an understanding of the process leading to this association. Such a comprehension would offer useful indicators to build an effective clinical intervention with these families.

In our study, we explored how family functioning and the way of living with a child's chronic illness may influence the illness trajectory. A trajectory is defined as the course of the illness over time from the perspective of the ill person and the carers (Glaser & Strauss, 1968). By involving both parents of the ill child, we wanted to understand illness trajectories in relation to the ways parents related to each other and to their children, and to the family story and organization. We considered different chronic illnesses instead of focusing exclusively on cancer, because so much research has analyzed the experience of parents of a child with cancer (Bjork, Nordstrom, Wiebe, & Hallstrom, 2011; Earle, Davies, Greenfield, Ross, & Eiser, 2005). By contrast, research on other chronic illnesses has been less expansive or has focused on a single chronic illness, thus preventing a comparison of the experiences of families coping with different illnesses. Patterson, Holm and Gurney (2004) found that most of the strains, resources, and coping behaviors of the parents of a child ill with cancer were similar to those described in studies of families experiencing other childhood chronic conditions, thereby allowing us to overcome this distinction and focus on how family organization may impact on illness trajectory.

Method

Participants

We recruited thirty-three parents (twenty mothers and thirteen fathers, aged between 30 and 55) of children with progressive chronic illnesses at a pediatric hospice in Padua, Italy as study participants. The affected children (twelve males and eight females) ranged in age from three months to eighteen years, and all lived at home. They were diagnosed with different progressive chronic illnesses, and were all indicated for palliative care. Five children had central hypoventilation syndrome (CCHS), one CHARGE syndrome, two Menkes syndrome, one Duchenne muscular dystrophy, one central core disease (CCD), five spinal muscular

atrophy (SMA), two brain tumors, one spinal dysraphism, one transverse myelitis, and one Down syndrome. The final number of participants was not predetermined. Our sampling ended once we considered the theoretical saturation had been reached, that is the point at which gathering more data yields no further theoretical insights about the emerging theory (Charmez 2006).

The university ethics committee approved the study. The chief of the hospice mediated contact. We met physicians, nurses, and psychologists who provided care to the children and interviewed one of the pediatricians, a 58-year-old woman. Each day, for a period of four months, one of us went to the hospice and collected data on the experiences of the families. There, a nurse or a psychologist of the ward introduced the study and the researcher to the families and obtained written informed consent. One mother among those who were asked refused to participate in the study, as did seven fathers whose wives participated in the study: Four were not interested, two were not available, and the last was unable to agree on a date.

Data Collection

We developed a semi-structured interview guide based on our review of the literature on parenting children with chronic illnesses. After a brief general introduction to the interview, participants usually spontaneously began to tell their experiences; otherwise, the interviewer began with a broad question about the parents' experiences with their child's illness and went on to explore various topics (e.g. when it was diagnosed, how parents reacted, their relationship with health care system, family roles and history).

Most of the interviews were conducted at the pediatric hospice. Our goal was to gather the experiences of both parents individually, but they were not always both present at the hospice, so we conducted a minor part of the interviews at participants' homes.

We also examined the medical records of the chronically ill children, and interviewed the physician who provided care to the children in order to obtain data pertaining to their clinical conditions, illness progression, family situation and relationship with the health care professionals from the physician's point of view. The interviews lasted from forty minutes to two hours. They were audio-taped and transcribed verbatim. We used pseudonyms to indicate children.

Data Analysis

The analysis of the interviews was guided by the principles of grounded theory (Strauss & Corbin, 1998). We conducted a preliminary analysis following each interview and used its results as a guide for the subsequent interviews and to verify the saturation criteria.

Therefore, the data generation and data analysis proceeded simultaneously. We started by reading through the interviews to form a general impression. In the second reading, we identified and categorized central themes. We subsequently revised some of these categories in light of the other interviews, and included notes and comments (open coding). The following step was comparing similarities and differences between themes, and relating categories to their subcategories along the line of their properties in order to develop a shared analytic framework (axial coding). Disagreements between our individual interpretations were resolved by discussion. The final phase of the analysis consisted in integrating the data from parents' interviews with those derived by the physician's interview and clinical records to generate a comprehensive theory of the illness trajectory in relation to family functioning (selective coding).

In accordance with the quality criteria for qualitative research, the aim of this study was to achieve credibility (Lincoln & Guba, 1985). The depth and breadth of the interviews enabled us to claim a comprehensive, authentic understanding of the experiences of the

caregivers. Analysis was tested by maintaining logical consistency and subjective interpretation. To increase the dependability an auditor reviewed and verified the consistency of this research. Finally, transferability was achieved by grounding, using thick quotes from the interviews.

Results

From the combination of the data derived by all study participants, we identified five final categories: illness experience (its meaning within the family and how the family coped with it), illness progression (the ill child's autonomy and severity of illness, referring especially to the physician's evaluation), family roles and history (including couple relationship, presence of siblings, working activities, and social relationships), trust in the health care system (what the parents asked and how they related with the health professionals), and resources (in whom the parents confided for help, including religion). On the basis of these categories, we grouped the families that had similar experiences among them and obtained four groups representative of four illness trajectories. The results will be presented by reporting the characteristics of each group of families in the five categories. We named the four illness trajectories 'possibility,' 'focus on illness,' 'denial,' and 'anger.'

Possibility

Nine families had given a meaning to the child's illness and subsequently reorganized their lives after an initial feeling of loss, as their words testified. 'A bolt from the blue' (Paul's father), 'It is like if you are quietly walking and fall down in a ravine, it upsets you' (Nancy's mother), but 'not everything comes to upset too much' (Paul's mother).

The children of these families were diagnosed with different illnesses: three Ondine syndrome, two spinal muscular atrophy (SMA), one CHARGE syndrome, one Menkes

Syndrome, one spinal dysraphism, and one Duchenne muscular dystrophy. The diagnosis dated back between few months and 13 years before. Sometimes it represented a relief for the parents, compared to the previous uncertainty about what was happening to their child or the fear of something worse. 'We were told he would have died, thus, when, two days later, we were told he had Ondine syndrome, thereby there was a possibility to manage it to live; it was a blessing for us' (Morgan's father). Being parents of a child with specific needs was lived by these parents as an opportunity that allowed them to discover new ways of being in a relationship. In two cases, the child's illness allowed parents to stop running and discover a new way of living like a family.

Before, I worked too much and during my child's first three years, I was here too little, and now I realize this. We stabilized a lot materially and inside ourselves. I am much quieter and my husband as well. Before, we lived too much of a race; we met too little. On the contrary now... sometimes I say that it has been better like that.

(Paul's mother)

These parents did not consider the child's illness an external enemy, but an event that may happen and must be accepted. 'Why [did this happen] to us? This is a question I never posed to myself, rather: Why to someone else? Or why not to me?' (Flora's mother)

More than one person told me: "Ah, but why, right to you?" If you do the math, for the law of big numbers, a jinx must happen to you. Let's call it jinx, i.e. a particular situation, you cannot always have everything: tall, beautiful, and with blue eyes.

Something must happen. (Mya's father)

Parents expressed ownership of the situation. ‘You begin very slowly to become confident with the illness that very slowly is not ‘rare’ anymore, it becomes yours. “Rare” because it is rare in the quantity of people [with the illness], but then you make it yours’ (Mya’s mother).

We have always related to her in a normal way. I have always said: “Okay, other mums change the diaper, feed their child with a bottle. I attach a pump and allow her to inhale; in short this is our normalcy” (Flora’s mother).

These parents expressed pleasure in their children’s accomplishments, they trusted and helped their children to do the most they could. This promoted their autonomy and improved their clinical conditions. ‘Despite the severity of her illness, Brooke is a child like any other, who throws a tantrum like all the other children and has a normal life’ (physician). ‘Now she is at an age that she says “I am old enough, I do it on my own”’ (Brooke’s mother). ‘He is very intelligent; he represents a stimulus for us’ (Paul’s mother). ‘He always tries to do the best; he is very tenacious’ (Nancy’s father).

These parents usually continued to work and do other activities. The flexibility of their roles allowed them to exchange roles and keep their own spaces, doing homework, gardening, and sometimes going out with their friends. The couple relationships were already solid and the parents maintained them after the child’s illness diagnosis, as their shared viewpoints on important questions testify:

We are very close to each other, even if sometimes we are more tired and we blurt, but we are lucky because I think that—and later we’ll see what he thinks about this—we live and think in similar ways on important matters, especially in difficult moments, like when she [Flora] is ill and I need to hear from him [the father] what he thinks about it. (Flora’s mother)

Three of these couples wanted to have other children. In the other families of this group, parents with other children recognized their needs and did not forget them. On the contrary, siblings became a resource for the parents and the ill child and, in turn, the ill children were perceived as a resource for their siblings. 'We are lucky because he [Michael's brother] has been a stimulus. If there would have been only Michael, we would have not gone to the sea' (Michael's mother). 'There is a family medical division around Brooke. Her twin says that when they will ride the motorbike, Brooke will bring her inhalator with her, and she will bring the aspirator. They have already divided the tasks' (Brooke's mother).

These families' stories are characterized by previous experiences that led them to face difficult situations in the past and subsequently to reorganize: abortions, migrations, loss of a loved one, the personal experiences of illnesses. They coped with these situations by confiding in themselves, but also confiding in others for help, as the following quotations testify. 'I think that sharing is important, because if you carry one hundred kilograms on your own, you find it difficult. [If you divide it] in two, maybe... cohesion brings you diluted pain' (Michael's father). 'If I need help, I'll ask for it. The first symptom I feel that I cannot face on my own, I'll ask for help' (Paul's mother).

These parents asked for help and searched for comparisons with other families with similar experiences: 'Comparing with others gives us strength' (Nancy's father). They usually also trusted the health care system, but this did not prevent them from searching for the best alternatives for their children and considering health professionals as persons who are not omnipotent.

They expressed faith in different forms, i.e. as faith in research, in one's own partner, in God, but always as one of the possibilities to cope with the situation and not the only one. 'You believe in God, you believe in nature, you believe in fate... I believe a bit in

everything' (Mya's father). 'A strong mother believes in life... I have a kind of faith that is all mine, but my faith helps me' (Manuel's mother).

Focus on Illness

For four families, the child's illness was at the center of their lives and became a mission. It was explained by attributing it to external causation (failed surgery or divinity), but did not happen by chance:

We think it has been the hand of the Lord ... He wanted to test me. "Why us?" I asked myself a lot of times. Because God knew that we could look at her, give her the love that someone else could not give her. (Alison's mother)

If she [Natalie] was not here it would be tough to survive because she fills your life. I was afraid to die before she did. Now I am afraid of the opposite. When she dies, for sure I'll go and help somebody else. It is one of my needs for personal gratification. (Natalie's mother)

These children were the firstborn or the parents had waited for them with great anticipation; sometimes they were also the result of an assisted reproductive procedure. 'Alison is at the center of everything, in the sense that she is the preferred granddaughter. In any case, she is the queen' (Alison's mother).

We are a close family, who has only one grandson: three children and only one grandson... after Maurice dies, it will be tremendous; it will be a huge void, for us, for sure, and also for them [the other relatives]. (Maurice's father)

All the children of this group of families were diagnosed with severe illnesses that require much care for long periods: spinal muscular atrophy (SMA) in three of our cases and a brain

tumor in the other case. Despite the severe clinical conditions that prevent children from having a satisfying life, these parents tried to keep the child alive at all costs.

He [Maurice] was a person who used to smile, now he does nothing, he is ugly to see, the tumor is pushing the eye out of orbit, he is swollen by cortisone, he has undergone fourteen neurosurgical operations . It is dramatic for an adolescent [...] Sometimes he says that he would like [to die], but he cannot for the people around him. When we decided to let him go we received the veto by his parents. They do not admit this possibility because it is in contrast with their religion. (Physician)

The ill child became the center of the mother's life in particular, who usually stopped working and created a 'symbiotic link' (Margaret's mother) with the child, devoting her life to the child's care. In this case, the strong link between the child and his mother kept her away from her husband. The fathers of this group of families usually participated in their ill child's care and changed their habits, but they did not consider this a burden. In three of these families, the couples' relationships were strengthened by the child's illness: 'We are much closer as a couple now' (Alison's mother); 'We tried to make also our marriage rotate around Maurice' (Maurice's father). In one case, the strong link between the child and his mother kept her away from her husband.

In those families with other children, their lives also rotated around the ill child's illness. Their parents presented these siblings of the ill child as responsible and 'too quickly grown up' (Natalie's mother).

He [Margaret's brother] is very responsible; he knows what he must do and say. If we are not in, he is even more responsible. I do not know how he lives this situation.

Maybe we take time away from him. (Margaret's mother)

These families found support inside the family and avoided comparing with others, ending up as closed systems: ‘There are friends, but they have their families’ (Margaret’s mother); ‘We are afraid to compare with others; we received help from our families; they pampered us’ (Alison’s mother). They trusted the health care system and relied on health professionals not only for medical care, but also for comfort: ‘They are good people and did even more than necessary’ (Maurice’s mother). But faith was the fundamental recourse for these parents: ‘Faith is helping us. It is a refuge, a comfort to feel pampered by God’ (Alison’s mother); ‘We made a journey with God that changed us and made us see things in a different way’ (Margaret’s mother).

Denial

Two families could not contemplate the illness because the parents already had a child, work, and illness prevented them from having other relationships and having ‘normal’ lives. These parents avoided considering illness and tried to ignore it because their life must go on. In one family, the mother refused to have an amniocentesis even when the results of first trimester screening tests were positive, thereby announcing a possible malformation of the fetus. Sam was born with Down syndrome, the ‘worst progression that a Down syndrome may have,’ as the physician who followed the child told us.

I have two children [apart from Sam]: one older and one younger. The older was affected by Sam’s illness because when Sam was born, I was away seventeen days and when his heart was surgically operated on, I was away for two months in intensive therapy. Eh, so many things! . . . The younger [sibling], who is six years old, now tells me, “Mum, can you not put a battery in Sam’s head? Can you not do anything?” . . . Sam is an angel, because he is good, quiet; but sometimes you think, “If he had been different. . .” I think it is normal that a couple thinks about other

families who do not have a child with a physical illness, who fend for themselves. If they want to go out, they go. Maybe we miss this: independence, not to depend on others. (Sam's mother)

In the second family, the mother described her child's Ondine syndrome as forgetfulness: 'He forgets to breathe' (Tom's mother). In this case, the child's illness has also assumed its worst form, with autistic traits. The physician stated, 'The child does not speak, does not vocalize, has no relation with his mother.'

Both these interviews were conducted in front of the ill children and the mothers told their stories as if their children were not there; nevertheless, they sometimes turned to the children with expressions like, 'You are strong like a little lion, aren't you?' (Sam's mother) or 'He has been very good, my champion!' (Tom's mother). Their narration seemed a dream in which situations followed each other without clear links. They surrendered to the flowing events with a fatalistic mood.

I always had faith, but now I have attached even more, I live it [the child's illness] like a mission. I say to myself, 'It means that God knew that He had to give him to me like this'... In the end, it all depends on fortune and destiny, because you cannot change them. Sometimes you do so much, but if it is predestined that that day must arrive and go in this way, you can jump or do miracles, but you must arrive there.

(Tom's mother)

Migrations and changing reference points led these families to confide mainly in themselves and to experience carelessness on the part of health care systems: 'We have been navigating in the ocean on our own' (Tom's mother). In both families, the mother provided almost all of the care for the child: in one family, the father was engaged in economically sustaining the

family and in the other family he was ‘absent’ and ‘elusive,’ as health care professionals told us. His wife ignored his absence, but we could not interview him because he avoided meeting the interviewer at the set date and place, even though his wife said he would have been very available for an interview.

Anger

Five families saw the illness as an enemy to fight, because it represented an obstacle for the realization of the parents’ life projects. These parents looked for the cause of their child’s illness and were engaged in a perpetual war of resistance. They recognized but did not accept the child’s illness and tried to resist the illness progression, like ‘Japanese who stayed in the trenches for thirty years, waiting for the enemy’ (Bill’s father).

I realize that the problems are very serious, but I live it [the situation] in this way: facing it, trying to curb this rampant sea, and control it as I can... We try to resist evil. I cannot accept giving in to evil, to negativity, to destruction. I never think there is uncertainty. (Alan’s mother)

In two families, the illness (Menkes syndrome in one case and transverse myelitis in the other) worsened aggressively, and the parents perceived this progression as a ‘bombing’ (Bill’s mother). In another family, the constant fight was expressed by the father’s description of his daughter with Ondine syndrome as ‘condemned to sight,’ because she was condemned to have eyes on her for all her life; in the meantime, she condemned those around her to have their eyes on her. Abby was less autonomous than a child with her illness might be, as the physician who took care of her told us.

These parents believed their ill child might do more than he or she could because they did not accept the illness (‘despite coming out bad’ Raul’s father) and its constraints:

‘He is in a chair and tends to lean too much when he could do it on his own’ (Raul’s mother).
 Illness interrupted their projects and passions, and in so doing meant the end of their lives: ‘It is devastating; we have no life any more’ (Alan’s mother). ‘You vanish as an identity’ (Alan’s father).

These families did not find a reorganization after illness. In one family, the disorganization was embodied in the destruction of the house order: ‘The domestic situation is completely distorted: I sleep with Alan and the father with his twin’ (Alan’s mother). The child’s room had become like ‘a room of an hospital’ (Alan’s father), and the drawing room was the other sleeping room, where books, beds, and toys were stacked, as health professionals told us, and where the father played with his three-year-old child with a computer, playing games that were too difficult for the child’s age. He was a university professor and never adjusted to the new life that illness implied.

In these families, the couples’ relationships were destroyed and the only communication between partners dealt with illness. ‘[in our condition] ninety-eight percent of couples break up. We are still here, but it is like if we were not here. We are here for him. If he were not here, we probably would not be here’ (Bill’s father).

Ah, ah, ah. Forgive my sincerity, but I really do not mind [the couple relationship].

Let’s say that the problem of the couple is the last of my problems. We do not have a personal life, so we do not have a couple life... of course, this problem may have exasperated the diversity we already experienced. (Alan’s mother)

In these families, the siblings of the ill children had problems and the siblings received different caregiving than that of the ill child. Alan’s twin had some linguistic problems. Bill’s twenty-year-old brother had serious behavioral problems. Abby’s newborn brother kept the mother far away from her ill child.

These parents' rage against the illness often became mistrust of the health care system, as for Abby's parents, who complained about the insufficient material support even if they received so much economic aid to be considered privileged by the health care staff, or Alan's father who also questioned the physicians' competence:

Sometimes you hit a wall, I would not say of insensibility, I would rather say of incapability; you hit an incompetence, sometimes incompetence, other times arrogance... In pediatrics, I had to explain how the thing [the illness] was. I mean that I do not think that they have so many cases, but you can go on the Internet and try to understand! This is your job. Lose a quarter of an hour of your time... Here in pediatrics, they cannot cure sane children either! (Alan's father)

These parents felt they must confide only in themselves and they must fight on their own. They were used to fighting, because they previously experienced frustration, loss, and conflict. 'I have always fought. I did not have so many friends. I have always fought to arrive where I had to arrive' (Raul's father). 'You lose your friends, because they all run away. This is for sure... They give you up, because life is like that: if you are fine, ok; otherwise, you lose' (Abby's father).

I am a fighter, and if I die, I die standing up... I know that there is me and all the rest is relative. I must confide in my strengths, and I know that I am available at one hundred percent. About others, you do not know, and I need certainty, so I must confide in myself. It is expensive, but it is my only certainty. (Bill's father)

The last family of this group was a special case, because the illness progressed fast soon after the child's birth. Brian was a three-month-old child dying from an aggressive brain tumor.

The anger against the health care system that did not recognize the illness quickly was the family's only perceived solution to this sudden and unexplained event.

The physician was in front of his computer and my child had a huge head. Then, when we got angry, [the doctor] looked up and measured his [our son's] head. The doctor's face bleached, and he sent me to the emergency, and everything began. (Brian's mother)

Discussion

From the analysis of the interviews, we found four illness trajectories that corresponded to different ways of dealing with the child's illness and different typologies of family systems. We have not found an association between these trajectories and the length or type of illness, thus suggesting that the illness trajectory is more traced by the family structure than by intrinsic aspects of the illness. For instance, three children with Ondine syndrome and almost of the same age, belonged to three different illness trajectories and they had very different illness progressions.

The four illness trajectories have been subsequently defined, not so much on a dimension of time as is usual in the grounded theory literature (Glaser & Strauss, 1968; Corbin & Strauss, 1991), but rather on the basis of two dimensions: the promotion of a child's autonomy and illness acceptance (Table 1). The theory generated by our data proposes that these dimensions may influence illness progression. Illness acceptance and the promotion of a child's autonomy allow recovery progression. Acceptance and the attempt to maintain dependency may contribute to the continuation of the illness. If illness is refused, families may try to ignore it or fight against it. In the first case, parents do not promote autonomy, thus preventing the child from finding effective ways to cope with illness. In the second case, parents' expectations of an excessive autonomy on the part of the ill children (if

compared with the possibilities allowed by their illness) may contrast the progression of illness, but may lead the children to feel guilty because they constantly feel that they should do better. Each trajectory is thoroughly discussed below.

Table 1. The description of the four illness trajectories on the basis of the two dimensions of illness acceptance and child's autonomy promotion

	Illness acceptance	Illness refusal
Promote autonomy	possibility	anger
Promote dependency	focus on illness	denial

Possibility

The first group of families was upset by the illness, but accepted it and was able to re-organize and use the situation as an opportunity to change. Their family roles were differentiated and flexible, and family life did not center exclusively on the illness: parents went on with their activities and cared for siblings' needs. They trusted the health care system. They paid attention and looked for care for the ill child but did not feel fury in a search for an impossible solution. These parents usually searched for comparisons with others and confided in themselves but also in others for help. They had differentiated social networks and found further resources in faith, research, and irony. They re-constructed the new and unexpected event (the child's illness) to find new solutions. In this context, the child's illness progression was the best one possible: children had the most autonomy their illness allows, and parents might accept the eventuality of death if it was the best possibility for their child.

In line with previous studies (Kratz, Uding, Trahms, Villareale, & Kieckheffer, 2009; McNeill, 2007) our first group of families succeeded in adapting to the changes that followed the child's illness when they found some positive meaning that helped them cope with the extra burden that the illness added to their daily lives, such as gaining appreciation for the things that really matter and personal growth. Our results also suggest that when parents care for themselves, their child does better (Patterson et al., 1990).

Focus on illness

The second group of families was centered and based on illness. The illness was not only accepted, but represented a way to keep the family going. The ill child was usually the first and only child, highly anticipated, sometimes not only by the nuclear family but also by the extended family. The illness kept the couple close and in those families with other children, they were left aside, in accordance with the frequent observation reported in literature that siblings are often forgotten family members when a child has chronic illness (Murray, 2002). The family searched for help inside itself more than outside, and usually trusted health care professionals, establishing with them a personal relationship and asking for comfort more than a cure. All of parents' lives, especially those of the mothers who may have had a very strong relationship with the ill child, were dedicated to caring for the ill child. However, they did not promote autonomy. They seemed to have neither trust nor hope for improvement, but often had faith that God might help them. The child's death or increase of autonomy would have threatened the maintenance of the family relationships, because the child represented the "fabric of hope" (Samson et al., 2009). If the child or the child's illness was not there, hope would end. As a consequence, despite the large amount of care provided, rather than facing their child's death, these parents accepted and maintained extreme clinical conditions and their children sacrificed themselves to this choice.

These families' strategy of adjusting to the child's illness may be read in terms of normalization, defined as the attempt to normalize the child's life, equating the chronic state of illness with the norm (Gravelle, 1997; Knafl & Deatrick, 2002). This search for normalcy is potentially damaging if it continues to elude parents (Earle, Clarke, Eiser, & Sheppard, 2007) and allows them to adopt an overprotective stance (Anderson, 1981; Hill, Higgins, Dempster, & McCarthy, 2009).

Denial

The third group of families did not accept their child's illness, and did not even recognize it. They avoided those procedures that might lead to its diagnosis and ignored or minimized it. This may also lead to avoiding choices that may foster recovery progression. The more the parents tried to deny the child's illness, the more illness worsened. The children of this group of families had the worst clinical conditions their pathology might have implied. The parents of this group usually rely on themselves for help, but can also have a fatalistic view and find in destiny or God an explanation for the situation. They often have other children and believe their family lives must go on, independent of the illness of the child. As shown in previous studies, denial is an attitude toward health and illness that may be due to a difficulty to accept illness (Cipolletta, Beccarello, & Galan, 2012; Cipolletta, Consolaro, & Horvath, 2014; Telford, Kralik, & Koch, 2006).

Anger

The parents of the fourth group never accepted the illness and saw it as something they must fight. The child's illness disrupted parents' personal projects, couple relationship and family life. Illness had a sudden insurgence (as in the case of transverse myelitis) or is rare (e.g. Menkes Syndrome). These parents did not give in because they were used to fighting against

adversity, because they had faced a multitude of problematic situations in their lives, usually confiding mainly in themselves. They could hardly rely on others for help and often mistrusted the health care system. A sense of duty governed their lives.

These parents expected themselves and others, including their ill child, to do more and more. This might have made their children feel guilty and subsequently internalize their feelings in order to avoid disappointing their parents, as Piazza-Waggoner et al. (2006) has also pointed out. In the case of those parents waiting for their child's death, paralysis and waiting, accompanied by anger, were predominant. Their condition is similar to the anger phase of the five stages of grief described by Kübler-Ross (1969). Anger does not help to face death because it keeps alive the need to fight and leaves the family in a void of meaning when there is no longer an enemy to fight.

Implications for nursing practice

As indicated in the previous studies on caregivers of chronically ill patients (Cipolletta & Amicucci, 2014; Cipolletta, Pasi, & Avesani, 2014; Cipolletta, Shams, Tonello, & Pruneddu, 2013), different directions of intervention may be traced on the basis of the four trajectories identified in our study. The families of the first group (possibility) may be actively involved in a collaborative relationship and may be supported by giving them all the information that may allow them to better care for their loved ones and, later, cope with their child's death. The parents of the second group (focus on illness) may be helped to engage themselves in activities other than caring and consider other aspects of their lives. This intervention might decrease the threat derived by an increase of the ill child's autonomy and may help parents to accept the eventuality of their child's death. The third group (denial) needs to be helped to adopt an exploratory attitude, which would allow the family to recognize signs of a worsening of the illness, search for help, and find new ways to cope with

the child's illness and impending death. Moreover, nurses might help these families to recall and to undertake the clinical examinations and treatments that may be forgotten. Finally, the families of the fourth group (anger) should not be contradicted because this would increase their anger. By taking care of the ill child with acceptance, nurses might offer a model of behavior that might allow these parents to consider the possibility to give up their fight against the illness. If their anger decreases, they can start to appreciate their children's accomplishments and support their attempts to autonomously move. At the same time, parents can prepare for the impending death because they no longer need an enemy to fight.

In a previous study (Cipolletta & Oprandi, 2014) we found that nurses are the healthcare professionals who usually have the task and the competence to communicate with the family caregivers of a dying person. Nurses may support families in the concrete management of the situation and by listening and being close to them.

Limitations and future research

We must consider the contributions of the present findings within its limitations. First, we utilized a small sample because we conducted a qualitative in-depth analysis that was very complex and time consuming, primarily due to the large amount of data we had. We need additional research with different samples. Second, as is typical of many studies in the pediatric field, we based our investigation mainly on parents' narrations and health professionals' perspectives rather than on direct observation of the interaction between family and child. It is difficult and sometimes inappropriate to involve children in these studies. Future research will likely benefit from direct observations of families, so interactions may be assessed more directly. Finally, the present investigation precluded an evaluation over time of the relationship between family functioning and illness progression. Unfortunately, it might

be explored only through longitudinal studies involving families before and after the illness insurgence, and that would be difficult to achieve.

Conclusions

Our study is in line with those studies that highlighted that people's attitudes, belief systems, lifestyles, and experiences with health, and how ultimately they used health providers, contributed to their proneness for illness and speed of recovery (Cipolletta, Beccarello, & Galan, 2012; Cipolletta, Consolaro, & Horvath, 2014; Sarafino, 1990) and were construed within the social systems (family, work, and community) to which they belonged (Cipolletta, 2011; Faccio, Bordin, & Cipolletta, 2013; Shapiro, 2002). Our results confirmed a family system perspective that assumed interdependence among family members (what happened to one member affected the other members) and added more detailed and articulated hypotheses on the relationship between some family aspects and illness progression to previous studies (Mitchell et al., 2007; Morris et al., 1997; Patterson et al., 1990; Perrin et al., 1993; Piazza-Waggoner et al. 2006; Rolland, 1987, 2004; Thompson et al., 1999).

Finally, our study represents an attempt to extend the methodology derived from grounded theory to an understanding of family functioning. The concept of illness trajectory has been redefined in order to understand the complex role a family plays in tracing a child's illness trajectory. The clinical implications of our study underscored the need for a family-focused approach when working with chronically ill children. Health care professionals interested in understanding the relationships between the illness progression and different risk and resilience factors should make special efforts to involve families in the care of the child. Listening to the individual and frequently different perceptions of the child's illness and understanding the complex role this illness plays within the family will help predict the child's illness progression and promote better development.

References

- Anderson, J. M. (1981). The social construction of illness experience: Families with chronically-ill child. *Journal of Advanced Nursing*, 6, 427-434.
- Bjork, M., Nordstrom, B., Wiebe, T., & Hallstrom, I. (2011). Returning to a changed ordinary life-families' lived experience after completing a child's cancer treatment. *European Journal of Cancer Care*, 20, 163-169. doi:10.1111/j.1365-2354.2009.01159.x
- Charmaz, K. (2006). *Constructing Grounded Theory. A Practical Guide Through Qualitative Analysis*. London: SAGE Publications Ltd.
- Cipolletta, S. (2011). Self construction and interpersonal distances of juveniles living in residential communities. *Journal of constructivist psychology*, 24, 122-143. doi: 10.1080/10720537.2011.548218.
- Cipolletta, S., & Amicucci, L. (2014). The family experience of living with a person with amyotrophic lateral sclerosis: A qualitative study. *International Journal of Psychology*. doi: 10.1002/ijop.12085
- Cipolletta, S., Beccarello, S., & Galan, A. (2012). A psychological perspective of eye floaters. *Qualitative health research*, 22, 1547-1558. doi: 10.1177/104973231
- Cipolletta, S., Consolaro, F., & Horvath, P. (2014). When health is an attitudinal matter. A qualitative research. *Journal of Humanistic Psychology*, 54, 391-413. doi: 10.1177/0022167813507630.
- Cipolletta, S., & Oprandi, N. (2014). What is a good death? Health professionals' narrations on end-of-life care. *Death Studies*, 38, 20-27. doi:10.1080/07481187.2012.707166
- Cipolletta, S., Pasi, M., & Avesani, R. (2014). Vita tua, mors mea: the experience of family caregivers of patients in a vegetative state. *Journal of Health Psychology*. doi: 10.1177/1359105314550348

- Cipolletta, S., Shams, M., Tonello, F., & Pruneddu, A. (2013). Caregivers of patients with cancer: anxiety, depression and distribution of dependences. *Psycho-oncology*, 22, 133–139. doi: 10.1002/pon.2081.
- Corbin, J., & Strauss, A. (1991) A nursing model for chronic illness management based upon the trajectory framework. *Scholarly Inquiry for Nursing Practice*, 5, 155-174.
- Earle, E. A., Clarke, S. A., Eiser, C., & Sheppard, L. (2007). ‘Building a new normality’: Mothers’ experiences of caring for a child with acute lymphoblastic leukaemia. *Child: Care, Health and Development*, 33, 155–160.
doi:10.1111/j.1365-2214.2006.00638.x
- Earle, E. A., Davies, H., Greenfield, D., Ross, R. & Eiser, C. (2005). Follow-up care for childhood cancer survivors: A focus group analysis. *European Journal of Cancer*, 41, 2882-2886. doi:10.1016/j.ejca.2005.08.028
- Faccio, E., Bordin, E., & Cipolletta, S. (2013). Transsexual parenthood and new role assumptions. *Culture, Health and Sexuality*, 15, 1055-1070. doi: 10.1080/13691058.2013.806676.
- Garro, A. (2004). Coping Patterns in mothers/caregivers of children with chronic feeding problems. *Journal of Pediatric Health Care*, 18, 138-144.
doi:10.1016/j.pedhc.2003.08.001
- Glaser, B. G., & Strauss, A. L. (1968). *Time for dying*. New Jersey: Aldine Transaction.
- Gravelle, A. M. (1997). Caring for a child with a progressive illness during the complex chronic phase: Parents’ experience of facing adversity. *Journal of Advanced Nursing*, 25, 738-745. doi:10.1046/j.1365-2648.1997.1997025738.x
- Hill, K., Higgins, A., Dempster, M., & McCarthy, A. (2009). Fathers' views and understanding of their roles in families with a child with acute lymphoblastic

- leukaemia: An interpretative phenomenological analysis. *Journal of Health Psychology, 14*, 1268-1280. doi:10.1177/1359105309342291.
- Hopia, H., Paavilainen, E., & Paivi, A. K. (2005). The diversity of family health: Constituent systems and resources. *Scandinavian Journal of Caring Sciences, 19*, 186-195. doi:10.1111/j.1471-6712.2005.00340.x
- Italian Pediatric society (2012). *Chronic and complex illnesses. Increasing data*. Retrieved from <http://sip.it/notizie/malattie-croniche-e-complesse-dato-in-aumento>
- Katz, S. (2002). When the child's illness is life threatening: Impact on the parents. *Pediatric Nursing, 28*, 453-463.
- Knafl, K., & Deatrck, J. A. (2002). The challenge of normalization for families of children with chronic conditions. *Pediatric Nursing, 28*, 49-56.
- Knafl, K., & Gilliss, L. (2002). Families and chronic illness: A synthesis of current research. *Journal of Family Nursing, 8*, 178-198.
- Kralik, D., Koch, T., Price, K., & Howard, N. (2004). Chronic illness self-management: Taking action to create order. *Journal of Clinical Nursing, 13*, 259-267. doi:10.1046/j.1365-2702.2003.00826.x
- Kratz, L., Uding, N., Trahms, C., Villareale, N., & Kieckheffer, G. (2009). Managing childhood chronic illness: Parents perspectives and implications for parent- provider relationships. *Families, Systems, & Health, 27*, 303-313. doi:10.1037/a0018114
- Kubler-Ross, E. (1969). *On death and dying*. London: Tavistock,
- LaMontagne, L. L., Hepworth, J. T., Salisbury, M. H., & Riley, L. P. (2003). Optimism, anxiety, and coping in parents of children hospitalized for spinal surgery. *Applied Nursing Research, 16*, 228-235. doi:10.1053/S0897-1897(03)00051-X
- Lincoln, Y. S., & Guba, E. G. (1985). *Naturalistic Inquiry*. Newbury Park, CA: Sage.

- McNeill, T. (2007). Fathers of children with a chronic health condition: Beyond gender stereotypes. *Men and Masculinities, 9*, 409-424. doi:10.1177/1097184X05284220
- Mitchell, S. J., See, H. M., Tarkow, A. K. H., Cabrera, N., McFadden, K. E., & Shannon, J. D. (2007). Conducting studies with Fathers: Challenges and opportunities. *Applied Development Science, 11*, 239–244
- Moola, F. J. (2012). This is the best fatal illness that you can have: Contrasting and comparing the experiences of parenting youth with cystic fibrosis and congenital heart disease. *Qualitative Health Research, 22*, 212–225. doi:10.1177/1049732311421486
- Morris, J. A. B., Blount, R. L., Cohen, L. C., Frank, N. C., Madan-Swain, A., & Brown, R. T. (1997). Family functioning and behavioral adjustment in children with leukemia and their healthy peers. *Children's Health Care, 26*, 61-75.
doi:10.1207/s15326888chc2602_1
- Murray, J. S. A. (2002). Qualitative exploration of psychosocial support for siblings of children with cancer. *Journal of Pediatric Nursing, 17*, 327-337.
- Patterson, J., McCubbin, H., & Warwick, W. (1990). The impact of family functioning on health changes in children with cystic fibrosis. *Social Science & Medicine, 31*, 159-164
- Patterson, J. M., Holm, K. E., & Gurney, J. G. (2004). The impact of childhood cancer on the family: A qualitative analysis of strains, resources, and coping behaviors. *Psycho-Oncology, 13*, 390-407. doi:10.1002/pon.761
- Perrin, E., Ayoub, C., & Willet, J. (1993). In the eyes of the beholder: Family and maternal influences on perceptions of adjustment of children with a chronic illness. *Journal of Developmental and Behavioral Pediatrics, 14*, 94-105.
- Piazza-Waggoner, C., Adams, C. D., Muchant, D., Wilson, N., & Hogan, M. B. (2006). Children with primary immunodeficiency disorders: Family functioning, coping

- strategies, and behavioral functioning. *Children's Health Care*, 35, 191-208.
doi:10.1207/s15326888chc3503_1
- Rolland, J. S. (1987). Chronic illness and the life cycle: A conceptual framework. *Family Process*, 26, 203-221. doi:10.1111/j.1545-5300.1987.00203.x
- Rolland, J. S. (2004). Helping families with anticipatory loss terminal illness. In F. Walsh & M. McGoldrick (Eds.), *Living beyond loss* (2nd ed., pp.213-237). New York: Norton.
- Samson, A., Tomiak, E., Dimillo, J., Lavigne, R., Miles, S., Choquette, M., ...Jacob, P. (2009). The lived experience of hope among parents of a child with Duchenne muscular dystrophy: Perceiving the human being beyond the illness. *Chronic Illness*, 5, 103.
doi:10.1177/1742395309104343
- Sarafino, E. P. (1990). *Health psychology: Biopsychosocial interactions* (5th ed). New York: Jhon Wiley & sons.
- Shapiro, E. R. (2002), Chronic illness as a family process: A social-developmental approach to promoting resilience. *Journal of Clinical Psychology*, 58, 1375–1384.
doi: 10.1002/jclp.10085
- Strauss A., & Corbin J. (1998). *Basics of qualitative research: Grounded theory procedures and techniques* (2nd ed.). Newbury Park: Sage
- Telford, K., Kralik, D., & Koch, T. (2006). Acceptance and denial: Implications for people adapting to chronic illness: Literature review. *Journal of Advanced Nursing*, 55, 457-464. doi:10.1111/j.1365-2648.2006.03942.x
- Thompson, R., Armstrong, F., Kronenberger, W., Scott, D., McCabe, M., Smith, B., ...Wright, E. (1999). Family functioning, neurocognitive functioning, and behavior problems in children with sickle cell disease. *Journal of Pediatric Psychology*, 24, 491-498.

Wong, M. G., & Heriot, S. A. (2008). Parents of children with cystic fibrosis: How they hope, cope and despair. *Child: Care, Health and Development*, 34, 344-54.

doi:10.1111/j.1365-2214.2007.00804.x