

### **What is already known about the topic?**

- There are similarities in children's emotional and spiritual struggles with their illness, their striving for normality and independence as well as physical symptoms and treatment experiences.
- Children are aware of issues related to their illness or prognosis even when information is not openly available to them.

### **What this paper adds**

- There was the dichotomy between some of the children's experiences and their parents' beliefs about their child's experiences.
- Children and their families can act as if they suspect or know about the prognosis and another day they can relapse back in pretending not to know.

### **Implications for practice, theory or policy**

- Parents' and healthcare professionals' understanding of the experiences of children diagnosed with life-threatening and life-limiting illnesses can be deepened by being aware that there are differences in what the child knows and what the parents think their child knows.
- Healthcare professionals can be helped to provide support by communicating with families and their children on any given day in their illness trajectory by assessing what stage they are at.
- Longitudinal studies which look at various phases of children's illnesses from diagnosis and beyond, would add an additional perspective in understanding children's experiences.

## **Introduction**

Serious illnesses affect all aspects of children's lives, such as their sense of well-being, independence, daily activities, as well as their relationships (1). Although the experiences of children diagnosed with a life-threatening or life-limiting illness have been studied over the past century, it remains an area which needs further exploration (2). A life-threatening illness is one where there is a high probability of premature death due to severe illness, but there is also a chance of long-term survival to adulthood such as with cancer (3). Life-limiting illnesses are defined (4) as conditions where 'premature death is usual' and where there is no reasonable expectation of a cure.

Research focuses on the experiences of children diagnosed with life-threatening or life-limiting illnesses (5, 6); others use parents' perspective as a proxy to understand children's experiences (7). There is a clear need for additional research on the experiences of children with life-threatening and life-limiting illnesses receiving paediatric palliative care, from their perspective as well as how their parents think that their children are experiencing their disease (7, 8).

The theories of awareness contexts and children's development are used to guide and inform the research, to offer a framework for the analysis, as well as to generalise from the findings. The theory of awareness contexts was developed in the 1960s by Glaser and Strauss (9) to explain the interactions between dying patients, their relatives and medical personnel. Glaser and Strauss found that awareness contexts have a significant impact on patients' interactions with their families and medical staff (10). They identified four types of awareness contexts: closed awareness, suspected awareness, mutual pretence and open awareness. Bluebond-Langner and Nordquest-Schwallie (11) explain that children function in these four different awareness contexts, depending on how much they know about their illness and prognosis and how their parents and healthcare professionals relate to them. Children intuitively adjust to the way their parents talk about their illness and death, matching the carer's level of disclosure (11, 12). Adults are frequently unwilling to share information with children related to their illness or prognosis, because they want to protect them. However, children are able to read subtle cues or they may talk with other children to learn about their illness when information is not openly available to them (13).

## Research Question and Objectives

‘What are the experiences of children diagnosed with life-threatening or life-limiting illnesses and how do their parents interpret their child’s perspective?’ Gaining insight into both the child’s and their parent’s perspective will allow a comparison between the two groups.

The objectives were:

- (1) to develop an understanding of the illness as experienced by the children;
- (2) to explore the perspective of the parents on how their child is experiencing disease;
- (3) to compare and contrast the results of both groups.

## Methods

### Research philosophy

The underlying research philosophy was from the interpretivist paradigm, which is characterised by using subjective epistemology in which there are diverse interpretations of reality (14). This perspective recognises that reality is socially constructed, subjective, interactive and changing, so the researcher aims to interpret the meanings others have about the world (15).

The theory of awareness context, developed by Glaser and Strauss (9) in 1965, was applied to children in Bluebond-Langner’s (12) study, which showed that children learn gradually about their illness and move from one context to another. The awareness context theory was extremely useful in the present study because it provides the theoretical lens through which we can understand what children know about their condition, as well as parents’ status in the context of their shared interaction (16). The theory explains the patterns of interactions among children and parents and how these interactions are affected because of what they know about the illness and prognosis.

### Setting/Population

Children who had been diagnosed with a life-threatening or life-limiting illness and the same children’s parents were recruited. The study was conducted in Romania. The children attended a palliative care unit which provides psycho-social, medical and spiritual support for children with various severe incurable diseases from diagnosis onwards; thus not all the children were in a terminal phase.

Inclusion and exclusion criteria were used to select research participants based on the research aim and research question. Purposive selection was used to ensure that the participants in the study represented the phenomena under investigation. Some of the children who receive palliative care are unable to talk because they have severe cognitive problems or because they have a brain tumour which severely affects their capacity to verbally express themselves (17). In these circumstances, they were considered unsuitable for inclusion in this research. Children with a severe level of intellectual delay were not included in the study as the research focuses on children who could understand and answer questions. The inclusion and exclusion criteria can be seen in the table below:

**Table 1 – Inclusion and exclusion criteria**

Inclusion criteria	Exclusion criteria
Children:	Children:
(1) aged between 14 and 17 years;	(1) who were unable to speak because their ability to verbally express themselves was affected by their illness;
(2) diagnosed with life-threatening or life-limiting illnesses;	(2) who had a severe level of intellectual delay;
(3) who were willing to be interviewed;	(3) who were in a terminal phase;
(4) who did not have a severe level of cognitive impairment;	(4) who were distressed at the time of the interview;
(5) who received consent from their parents;	(5) who were not Romanian speakers.
(6) who gave their assent to participate in the study;	

---

(7) who agreed to be voice-recorded during the interview;  
(8) who were not actively dying.

---

### Ethical Issues

Ethical approval was obtained from a palliative care service in Romania and subsequently from the Faculty of Health and Medicine Research Ethics Committee at Lancaster University and Lancaster University Research Ethics Committee in 2016.

Special attention was given to the protection of participants' data. Data were managed in accordance with the Romanian National Ethics Council (18) and with the UK Data Protection Act (19). Participants were assured about the confidentiality of their personal information. Each interview was conducted in the Romanian language and recorded using an encrypted audio device. Audio recordings were transferred in a password protected laptop which was also encrypted.

### Recruitment and consent

Participants were identified through the database of a paediatric palliative care unit. The parents were contacted by phone and told about the purpose of the study. They also received information sheet by post about the project. With the parents' approval, the children were approached regarding participation in the study. The research required the parent's consent and the child's assent.

### Data collection

This study employed in-depth semi-structured interviews. An interview topic guide was used and conversation encouraged with cues from the interviewer. All interviews were face-to-face and lasted from 30 to 70 minutes. All participants were allocated pseudonyms. Data were collected by one of the authors of this paper as an insider researcher. She had a prior relationship with the participants in her role as a paediatric palliative care social worker.

### Data analysis

The interview data were openly analysed using thematic analysis by EC under the supervision of NP and a second supervisor (20). This approach allowed for the description and interpretation of the participants' views (21). We followed the six-step process for thematic analysis outlined by Braun and Clarke (22) which includes familiarising oneself with the data, generating initial codes, searching for themes, reviewing themes, defining and naming themes and producing the report. The text from each participant was coded using NVivo (23). The results from the participants' interviews were explored for similarities and differences (24). The final phase produced a scholarly report based on the analysis (22).

## Findings

Twenty individuals were interviewed for the study. Ten of them were children (six boys and four girls) aged between 14 and 17 years (median age 16.5 years) diagnosed with a life-threatening or life-limiting illness (Table 2). Five children were excluded based on the exclusion criteria. Thirteen families were contacted about the study of which three recruited children died before they could be interviewed. The median duration of palliative care services for the whole study population was four years.

**Table 2** - The children's characteristics

Children	Diagnosis
Victoria	Muscular Dystrophy
Melissa	Epidermolysis bullosa
Paula	Autoimmune thrombocytopenia
Patrick	Mixed Connective Tissue Disease
Alisia	Dystrophic Epidermolysis Bullosa
Denis	Duchenne muscular dystrophy

Flaviu	Stg IV Lymphoma
Damian	Abdominal Lymphangioma
Anton	Acute lymphoblastic leukaemia (Relapsed)
David	Non-Hodgkin Lymphoma (Relapsed)

Interviews were also undertaken with the parents of these ten children, therefore ten parents (in all cases, the mothers) participated in the study. The saturation was achieved at the eighth interview and continued until the tenth; the last two interviews did not provide new insights. The data produced six major themes which are presented below.

### Knowledge about diagnosis and prognosis

Some of the children did not know their diagnosis, while others were aware of it and the long-term consequences regarding life expectancy. The children diagnosed with life-threatening illnesses reported that they learned their diagnosis from the oncologist. Anton said: *'My mum couldn't tell me from the very beginning that I have cancer ... she was too shocked, so I found out from my doctor.'* Children diagnosed with life-limiting illnesses, experienced a different process probably because they were born with their disease, or diagnosed at a young age. Denis was diagnosed with a life-limiting illness at the age of ten and his family tried to hide the diagnosis from him to protect him. Knowing that it would upset his mother, he learned not to show interest in learning more about his illness: *'I don't remember how I got ill ... I know mum avoids discussing my illness, so I leave things as they are.'*

Parents of the children diagnosed with life-threatening illnesses were more likely to hide the diagnosis from the child but knew that their children would eventually find out. Some parents were shocked about their children's diagnosis as Damian's mother stated: *'I couldn't tell him that he has cancer. The doctor told me that his illness is critical and that he doesn't know what will happen ... I did not have the courage to tell him about it.'* At the time of diagnosis Damian's family was at the level of closed awareness context because of the lack of information the patient received about the diagnoses.

### Death and dying

Many of the children's comments evidenced their awareness that they could die from their disease. Several children expressed their desire to die because of the difficulties they faced. Some children discussed how they started to view their future negatively because of their prognosis, as Flaviu expressed it, *'I was terrified that I would die'*. Patrick, diagnosed with a life-limiting illness, claimed that: *'There are times when I have so much pain and I concluded that I cannot stand it anymore. I said to myself that I'd be better off dead so I would not have to suffer anymore. I would not have to stand so much pain. I thought that it is useless. There is no reason to struggle like that! It's not worth it!'*

Parents were not always aware of their children's thoughts about death. For example, dying was a recurrent thought for Melissa, who had a life-limiting illness, but her mother was not aware of it. For her, Melissa was still the *'little girl who cannot think of such serious things'*. Some parents were trying to conceal information about the diagnoses or prognosis from their children. There were also parents and children who focused only on discussions which were positive. Alisia's mother claimed that she started to encourage her daughter *'to be positive at a very young age; I always tell her not to think of the worst. We don't talk about death at all.'* One side of the mutual pretence context is where all the parties would focus on topics which were safe as in the case of Alisia and her mother. Alisia's mother encouraged open discussions with her daughter, but only to the point where the mother felt safe. This means that parents avoided talking about issues such as their child's illness getting worse or that they could die because of the illness.

### The spiritual response to illness

Some of the participants mentioned spirituality as a source of support while others turned their anger on God. Several children tried to find a purpose for being ill. Melissa said, *'I am often sad and upset, wondering why do I have to be like this?'* Victoria came to the conclusion that it did not help to complain and be upset about her illness:

*'I realised that God made me like this with a purpose. And my purpose now is to show everybody that someone can live like this and all you need to do is not to complain, but to persevere. I have to be thankful!'*

Parents shared their children's frustration about their illness as well as being angry with God. Damian's mother said that her son often asked her: *'Why do I have to be so ill? Are the other children better people than me? Am I the worst of them all?'* His mother explained that she would encourage him that he would be healed but she did not believe it either. Operating in the mutual pretence context, some parents like Damian's mother would avoid communicating with their children about their prognosis. They thought their children would give up fighting if they knew how bleak it was.

### The emotional response to illness

Children explained how they felt scared, depressed, upset, angry or anxious about issues related to their illness. Melissa, confessed her internal turmoil: *'I am sad many times because of my illness ... too many times ... I am worrying and troubled inside and I usually cry for about an hour. Then I calm down.'* For Flaviu it was so difficult when he was told about his diagnosis, that he decided to be silent about his thoughts and feelings regarding his illness.

While Patrick's mother thought the *'hardest thing for him was his struggle with the pain'*, he was more *'troubled by the waves of sadness that came over him'*. Denis was not used to talking about his feelings with his mother; she admitted that she *'did not encourage him to express his emotions regarding his illness'*. She claimed that *'for sure he is not all right with his illness and he is the only one who knows his struggles, only he knows what is deep in his soul.'* However, Denis adjusted his behaviour to his mother's expectations, but he had moments when he would *'burst out'*. Because he was not encouraged to talk about his illness and prognosis, he was reluctant to express his fears as he wanted to protect his mother. In this way he would operate in the suspected awareness context, by searching on the internet for information regarding his illness.

### Striving for normality and independence

A variety of issues described by the children revealed that they were striving for normality and independence. The findings suggest a tension between the desire to be independent and the need for dependency created by the illness. Victoria wished to be independent and resented her mother's continuous presence. However, she also expressed her fear of being left alone and recognised her need of her mother.

Some of the parents' insights overlapped or contrasted with their children's views on their illness. While Paula tried to consider herself a normal child, her mother explained that Paula was trying hard to prove to the other children she was like them:

*'She was trying to redress the balance because she had to go to the hospital so often and in a way, she was trying to retrieve the lost time in the hospital. So she would try to do what other normal children would do ... she would try to demonstrate to her classmates that she can do everything that they were doing.'*

Alisia wanted to make *'an independent decision regarding challenging medical procedures'*, despite knowing that her mother struggled with it. Also, her mother explained how her *'daughter would never be independent and have her own family however much she wished for that'*. She was protecting her daughter by minimising her illness. The way her mother responded indicated her reluctance to talk with Alisia about her illness. In this way Alisia and her mother operated in the mutual pretence context which is initiated when children note that parents are not willing to talk about their condition with them.

### Coping strategies

The range of coping strategies used by children, besides their faith in God, included family, social support, distraction and optimism. In their attempts to cope with their illness, all participants described that they sought comfort from family members or their peers. Patrick reported:

*'Mum is always there for me ... for whatever I need, she's there to help. And I'm so grateful for this. I mean, if I could have a long life to thank her every day for what she does for me, it wouldn't be enough'*

*(...) I would be nothing without her. I don't know if I could go through this if anything ever happened to her.'*

Patrick's mother reinforced his perspective regarding the *'importance of not being alone in his most difficult periods of time'*. Other parents' comments demonstrated a different interpretation of their children's coping strategies. Paula's mother said the way of dealing with her illness was that *'she would try to ignore it'*. She claimed that *'Paula was aware of what her illness meant and about her life expectancy, but she would ignore it altogether'*. Her mother explained that *'ignoring it would mean that she would refuse to talk about it and to deny her fears of the future'*. In this way Paula and her mother were operating in the mutual pretence context, by not openly communicating about Paula's illness.

## Discussion

The thoughts that participants shared about their experiences allowed us to understand what it means for them to bear the burden of a serious diagnosis. Results indicate that children were not only having emotional and spiritual difficulties, but also challenges in fighting for normality and independence.

The experiences of children diagnosed with life-threatening or life-limiting illnesses can be understood in the context of awareness theory. As Bluebond-Langner and Nordquest-Schwallie (11) showed, children manage their awareness of illness prognosis by functioning in one of the four awareness contexts.

One of the most significant findings from the study is that children and their parents do not always stay in the same awareness context. Bluebond-Langner (12) showed that children progressively move from closed to open awareness context. While our study also found this, it also shows that children do not only evolve in one direction from closed to open awareness context, but they might move back and forth from one awareness context to another, depending on the circumstances. For example, one day a patient and their family could act as if they suspect or know more about the prognosis and another day they could relapse back into closed awareness or mutual pretence. For example, David operated in the open awareness context with his mother. They communicated about his diagnosis and prognosis. However, when he realised that his mother did not cope well, he tried to protect her by assuring her that she did not have any reason to worry. He would pretend not to know he could get worse. In this way the family moved to a mutual pretence context, pretending not to know the prognosis.

The dominant mode of interaction between the children and the parents in the study was mutual pretence. These results reflect those of Bluebond-Langner and Nordquest-Schwallie (11) who explain that this practice is the most frequent one because speaking about death is a taboo. Protecting one another usually informed the participants' decision whether or not to talk about their illnesses. As Timmermans (25) explains there is no appropriate emotional response to a terminal condition and no awareness context should be imposed upon children and their families. In this study there was only one family who operated in the open awareness context because the mother openly communicated with her son when he had been diagnosed with leukaemia. She shared all the details given to her by the medical staff about his diagnosis and prognosis.

The study also found that there was a dichotomy between some of the children's experiences and their parents' beliefs about their child's experiences. Although parents thought they knew almost everything about their children, results suggest that many of them did not know as much as they believed. Anderzén-Carlsson et al.'s study (26) came to the conclusion that parents were aware of their child's concerns, but they were also aware that they were not always able to know their child's mind. Other studies (27, 28) also show that children may think differently from their parents in terms of how they experience their illness. (29).

The study also suggests that both children's development and experiences contribute to the perception of their illness. Children's behaviour can alternate between accelerated maturity and infantile regression according to changing circumstances. As a result of the interviews with both children and their parents, it can be concluded that some of the children can appear to be more mature than their age because of what they have had to deal with; also these children sometimes behaved like much younger ones when it came to their wishes and desires. For example, they could understand the implications of their illness, but they would sometimes behave in an infantile



way when going to the hospital for treatment. Paula had a mature understanding about her diagnosis and prognosis. However, she behaved like a child, as her mother revealed, when she had to go to the hospital for treatment. This is an example of the fragile hold on maturity at this life stage when regression into childlike behaviours can result from fear and anger at the unfairness of their situation.

Acknowledging that children can move back and forth from one context to another could help healthcare professionals to provide support by communicating with families and their children on any given day in their illness trajectory by assessing where they are at.

Although the study adds to the knowledge about the experiences of children diagnosed with a serious illness, future research is warranted especially with younger children because most studies concentrate on adolescents or young people. Also, for a greater understanding of children's experiences, longitudinal studies which look at various phases of children's illnesses from diagnosis and beyond, would add an additional perspective in understanding children's experiences.

### Limitations and strengths

The sample size of ten children and ten parents included in the study is justified as the interviews conducted with children were in-depth and in parallel to interviews with parents, which created a richer context of their experiences. Thus, this study offers meaningful insights into the experiences of seriously ill children and is strengthened by the inclusion of the perspectives of both the children and their parents.

Because the research was conducted in a single institutional setting, and a limited number of children with serious illnesses were included, the results might be specific to certain illnesses. Even so, the discussion was not specific to their illness, but covered more general topics, such as self-esteem and independence.

The study will have a significant contribution in the field of paediatric palliative care in Romania but findings are relevant to other countries. Some experiences of children diagnosed with life-threatening and life-limiting illnesses are universal. There are some culturally-situated experiences which might be more in keeping with countries similar to Romania.

### Conclusion

Children in the study often knew more about their condition than their parents realised. The children therefore managed these feelings alone as they did not have open dialogue with their parents. The findings of this study can be used to deepen healthcare professionals' understanding of the dialogue between children diagnosed with life-threatening and life-limiting illnesses and their family. Using this understanding, healthcare professionals can advise parents and children about how to communicate with one another which would enable the children to give voice to their thoughts, emotions and experiences.

### References

1. Kane JR, Barber RG, Jordan M, Tichenor KT, Camp K. Supportive/palliative care of children suffering from life-threatening and terminal illness. *Am J Hosp Palliat Care*. 2000;17(3):165-72.
2. Gibson F, Richardson A, Hey S, Horstman M, O'Leary C. *Listening to children and young people with cancer*. London, UK: Macmillan Cancer Relief; 2005.
3. ACT Staff, Chambers L, Association for Children's Palliative Care. *A Guide to the Development of Children's Palliative Care Services*. ACT; 2009. Report No.: 9781898447092.
4. Steering Committee of the EAPC task force on palliative care for children and adolescents. Steering Committee of the EAPC task force on palliative care for children and adolescents IMPaCCT: Standards for Paediatric Palliative Care in Europe. *European Journal of Palliative Care*,. 2007;14(3):109-14.

5. Holge-Hazelton B, Timm HU, Graugaard C, Boisen KA, Sperling CD. "Perhaps I will die young." Fears and worries regarding disease and death among Danish adolescents and young adults with cancer. A mixed method study. *Supportive care in cancer : official journal of the Multinational Association of Supportive Care in Cancer*. 2016;24(11):4727-37.
6. Carnevale FA, Gaudreault J. The experience of critically ill children: A phenomenological study of discomfort and comfort. *Dynamics (Pembroke, Ont)*. 2013;24(1):19-27.
7. Ciobanu E. *The experiences of children diagnosed with a life-threatening or life-limiting illness*: Lancaster University; 2019.
8. Ullrich C, Morrison RS. Pediatric palliative care research comes of age: what we stand to learn from children with life-threatening illness. *J Palliat Med*. 2013;16(4):334-6.
9. Glaser BG, Strauss AL. *Awareness of dying*. London,: Weidenfeld & Nicolson; 1965. xiii, 305 p. p.
10. Tom A, Alvita N. Awareness of Dying Remains Relevant after Fifty Years. *Grounded Theory Review: An International Journal*. 2015;14(2).
11. Bluebond-Langner M, Nordquest-Schwallie M. "It's Back": Children with Cancer Talking about Their Illness When Cure Is Not Likely. In: Comacchio C, Golden J, Weisz G, editors. *Healing the World's Children: Interdisciplinary Perspectives on Child Health in the Twentieth Century*. Montreal: McGill-Queen's University Press; 2008.
12. Bluebond-Langner M. *The private worlds of dying children*. Princeton, N. J.: Princeton University Press; 1978. xv, 282 s. p.
13. Bearison DJ. "They never want to tell you": children talk about cancer. Cambridge, Mass.: Harvard University Press; 1991. xiv, 194 p. p.
14. Bunniss S, Kelly DR. Research paradigms in medical education research. *Med Educ*. 2010;44(4):358-66.
15. Creswell JW. *Qualitative Inquiry and Research Design: Choosing Among Five Approaches*: SAGE Publications; 2012.
16. Lindesmith AR, Strauss A, Denzin NK. *Social Psychology*: SAGE Publications; 1999.
17. Muckaden M, Dighe M, Balaji P, Dhiliwal S, Tilve P, Jadhav S, et al. Paediatric palliative care: theory to practice. *Indian journal of palliative care*. 2011;17(Suppl):S52-60.
18. Romanian national ethics council. [Available from: <http://cne.ancs.ro/> (2004, accessed 11 November 2019).
19. UK Data Protection Act. 1998 [Available from: <http://www.legislation.gov.uk/ukpga/1998/29/data.pdf> (accessed 7 December 2019).
20. Braun V, Clarke V. *Successful qualitative research : a practical guide for beginners*. London: SAGE; 2013.
21. Smith J, Firth J. Qualitative data analysis: the framework approach. *Nurse Researcher*. 2011;18(2):52.
22. Braun V, Clarke V. Using thematic analysis in psychology. *Qualitative Research in Psychology*. 2006;3(2):77-101.
23. Mason J. *Qualitative Researching*: SAGE Publications; 2002.
24. Barbour RS. *Introducing qualitative research : a student's guide to the craft of doing qualitative research*. London: SAGE; 2008.
25. Timmermans S. Dying of awareness: the theory of awareness contexts revisited. *Sociology of Health and Illness*. 1994;16(3).
26. Anderzén-Carlsson A, Kihlgren M, Svantesson M, Sorlie V. Children's Fear as Experienced by the Parents of Children With Cancer. *Journal of Pediatric Nursing*. 2007;22(3):233-44.
27. Montoya-Juarez R, Garcia-Caro MP, Schmidt-Rio-Valle J, Campos-Calderon C, Sorroche-Navarro C, Sanchez-Garcia R, et al. Suffering indicators in terminally ill children from the parental perspective. *Eur J Oncol Nurs*. 2013;17(6):720-5.



28. Cataudella DA, Zelcer S. Psychological experiences of children with brain tumors at end of life: parental perspectives. *J Palliat Med.* 2012;15(11):1191-7.
29. Grinyer A. Contrasting parental perspectives with those of teenagers and young adults with cancer: comparing the findings from two qualitative studies. *Eur J Oncol Nurs.* 2009;13(3):200-6.