Importance of parental involvement in paediatric palliative care in Hong Kong: qualitative case study

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ABSTRACT

Objective To compare and contrast the perceived care needs of children with life-limiting conditions (CLLC) from the perspectives of the children, parents and healthcare providers.

Design A qualitative case study method using semistructured interviews was employed with a within-case and across-case analysis. Themes and subthemes emerging from the cases were compared and contrasted in the across-case analysis to explore the similarities and variations in participant perceptions.

Setting/participants The setting was the paediatric departments of five regional hospitals in Hong Kong. Twenty-five sets of informants (CLLC–parent–healthcare provider) were recruited, with 65 individual interviews conducted.

Results A total of 3784 units of analysis were identified, resulting in three themes with subthemes. 'Living with the disease' (55.8%) occupied the largest proportion, followed by 'information and understanding about the disease' (27.4%), and 'care support and palliative care' (16.8%). Healthcare provider support mainly focused on physical concerns. Family and social support were present, but carer stress created tension between couples. Doctors were the primary source of medical information, but the parents had to seek further information via the internet and support from patient groups. There was a perceived need for better coordination and collaboration of care. The palliative care approach coordinated by nurses was seen as helpful in addressing the care needs of the CLLC.

Conclusions This original study identified the importance of palliative care with active engagement of parents which can address the service gap for CLLC.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ Children with life-limiting condition (CLLC) require complex medical interventions due to their multiple care needs in their relatively short lifespan.

⇒ Parents of CLLC often compromise their own well-being as caring for their children is a lifelong commitment.

⇒ A palliative care approach is recommended to address CLLC's physical and psychosocial needs.

WHAT THIS STUDY ADDS

⇒ This study simultaneously examining perspectives of the CLLC, parents and healthcare providers found that care is fragmented, and the voices of the parents were missing.

⇒ The palliative care approach is a promising way to address the service gap of fragmented care and provide holistic care with coordinated multidisciplinary efforts.

⇒ A nurse coordinator could be instrumental in addressing care fragmentation in an interdisciplinary team with the partnership of the parents and CLLC.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ The triangulated findings in Hong Kong revealed that parental voice was missing in palliative care for CLLC despite the literature consistently highlights its importance.

⇒ A coordinated and proactive care model with a nurse coordinator could potentially save costs by streamlining compartmentalised hospital services.

⇒ The study's findings support the clinical guidelines proposed by international experts and could assist policymakers and healthcare professionals in designing future paediatric palliative care services.

INTRODUCTION

Children with life-limiting condition (CLLC) face many challenges that affect themselves, their families and healthcare providers. Research has shown an increase in the number of CLLC in recent years.[1] Caring for CLLC is a life project for the parents. Despite having a shorter life expectancy than healthy children, CLLC continue to grow up encountering wide-ranging developmental needs.[2] Evidence suggests that parents require both support from people (eg, family, healthcare professionals) and community resources (eg, hospital care, school and social services) when caring for CLLC.[3] A qualitative meta-summary analysis[4] revealed that both parents and healthcare providers emphasised the need for services from the time of diagnosis until the child's death. The provision of psychosocial support, such as counselling and patient group networking, was helpful and while living with a challenging day-to-day reality, a more family-centred approach was preferred.[2 5]

These parents play a key role throughout their child's life journey and have a considerable need for information to help them understand the disease and treatment choices. Provision of informational support can prepare parents for what to expect in terms of the progression of their children's illness and potential treatment.[6] Early and regular engagement in communication between parents and healthcare providers was seen as important.[3] Parents in one study found that decisions relating to advance care planning were made relatively late in the illness.[7] Studies also revealed a discrepancy between the views of children/families and those of healthcare providers in terms of information content and amount,[4] availability of information in easily understandable language[8] and their engagement in shared decision-making.[9]

Evidence suggests that CLLC benefit from a palliative care approach that advocates holistic care supported by an interdisciplinary team. The initiation of paediatric palliative care (PPC) is recommended once a life-limiting diagnosis is confirmed or even prior to diagnosis where appropriate.[10] The implementation of PPC embraces the physical, psychological, social and spiritual well-being of both the children and their families.[2] In Hong Kong, the Hospital Authority issued a Strategic Service Framework in 2017. This framework guides the development of adult and paediatric palliative care services, aiming to support patients with life-limiting conditions by addressing their physical, psychological and spiritual needs and improve their quality of life.[11] However, it was noted in the document that PPC, unlike the adult group, is underdeveloped and not carried out in a structured manner.

Several studies have investigated the perceived needs of children, families and healthcare providers separately, but no study has been identified that compares the views of children, parents

Table 1 Demographic characteristics of the study participants

	CLLC (n=25)	Parent (n=25)	HCP (n=15)
	n (%)	n (%)	n (%)
Age in years (mean, SD, range)	13.5 (SD=3.4, range=8–19)	45.4 (SD=7.1, range=35–64)	47.7 (SD=6.6, range=31–59)
Gender			
Female	11 (44.0)	17 (68.0)	7 (46.7)
Male	14 (56.0)	8 (32.0)	8 (53.3)
Education			
Primary	9 (36.0)	2 (8.0)	—
Secondary	16 (64.0)	15 (60.0)	—
Tertiary or above	0 (0.0)	8 (32.0)	—
Religion			
Christian	5 (20.0)	5 (20.0)	8 (53.3)
Catholic	0 (0.0)	0 (0.0)	1 (6.7)
Buddhism	2 (8.0)	2 (8.0)	0 (0.0)
Other	2 (8.0)	2 (8.0)	0 (0.0)
No religion	16 (64.0)	16 (64.0)	6 (40.0)
Marital status			
Single	—	0 (0.0)	—
Married	—	20 (80.0)	—
Widowed	—	3 (12.0)	—
Divorced	—	2 (8.0)	—
Number of children			
One	—	5 (20.0)	—
Two	—	15 (60.0)	—
Three or more	—	5 (20.0)	—
Employment			
Full-time employment	—	7 (28.0)	—
Part-time employment	—	3 (12.0)	—
Not in employment	—	15 (60.0)	—
Housing type			
Public housing	—	17 (68.0)	—
Private housing	—	8 (32.0)	—
Occupation			
Physician	—	—	13 (86.7)
Nurse	—	—	2 (15.3)
Experience in paediatric specialty in years (mean, SD, range)	—	—	21.9 (SD=5.5, range=7–31)
Experience in PPC in years (mean, SD, range)	—	—	4.9 (SD=4.5, range=0–17)
Received training in PPC			
Yes	—	—	10 (66.7)
No	—	—	5 (33.3)

CLLC, children with life-limiting condition; HCP, healthcare provider; PPC, paediatric palliative care.

Table 2 Distribution of cases (n=25) by categories and diagnoses

Together for Short Lives categories	Diagnoses	Number (percentage)
I: curative treatment may be feasible but may fail	Bronchiolitis obliterans with chronic respiratory failure and pulmonary hypertension	2 (8)
	Tuberous sclerosis	
II: long periods of intensive treatment needed to prolong life and allow normal activities	Congenital megaconial muscular dystrophy	5 (20)
	Duchenne muscular dystrophy	
	Duchenne muscular dystrophy	
	Pompe disease	
	Spinal muscular atrophy type 2	
III: progressive conditions without curative treatment options and treatment is exclusively palliative	Giant congenital cystic hydroma	8 (32)
	Glut 1 deficiency	
	Glut 1 deficiency	
	Hereditary spastic paraplegia	
	Hereditary spastic paraplegia	
	Jarcho-Levin syndrome	
	Mitochondrial neurodegenerative disease	
	No definitive diagnosis	
IV: irreversible but non-progressive, causing severe disability leading to health complications and premature death	Apert syndrome	10 (40)
	Down’s syndrome with repaired cardiac condition	
	Dyskinetic cerebral palsy	
	Dyskinetic cerebral palsy	
	Dystrophic epidermolysis bullosa	
	Heteropia, intractable epilepsy, right hemiplegia	
	Klippel-Feil syndrome with complex cardiac condition	
	Osteogenesis imperfecta type III	
	Pseudochondroplasia	
	2q37 microdeletion syndrome	

and healthcare providers simultaneously. This study was therefore conducted to compare and contrast the perceived care needs of CLLC from perspectives of these three stakeholders in Hong Kong.

METHODS

Study design

This study uses a qualitative case study method, allowing researchers to study complex phenomena in depth within their context while involving these different perspectives and providing rich data.[12]

Setting and participants

The study took place in the paediatric departments of five regional hospitals in Hong Kong from August 2019 to October 2021. Purposive sampling was used with the aim of recruiting 25 sets of participants (CLLC–parent–healthcare provider). Site paediatricians, not involved in the interviews, identified eligible participants and the researchers provided potential participants with information sheets with an explanation of the study. In total, 65 individual interviews were conducted, comprising 25 CLLC, 25 parents and 15 healthcare providers who looked after more than one of the interviewed children.

The inclusion criteria were: (1) families with diagnosed CLLC, as guided by Together for Short Lives[13]; (2) eligible CLLC between 8 and 19 years of age and not in active treatment; (3) paediatricians and nurses with experience of caring for the CLLC; (4) parents aged 18 years or above; and (5) ability to communicate in Chinese or English.

The exclusion criteria included: (1) hospitalised children; (2) newly diagnosed CLLC within the previous 12 months; (3) children in the stage of active dying/facing imminent death; and (4) parents with cognitive impairment.

Data collection

Audio-recorded semistructured interviews lasting 25–60 min were conducted individually in private meeting rooms or online during the pandemic by trained researchers who were not familiar with the participants (JMCH: a female PhD student; TCL: a female research associate). Field notes were taken.

Participant demographic information was collected. The interview guide was constructed based on a literature review reporting the needs of CLLC and validated by an expert panel team composed of three paediatricians and five paediatric nurses. A pilot interview was conducted with a child with spinal muscular atrophy, her mother, and the primary paediatrician to test its clarity and relevance to the participants. The interview guide (see online supplemental file) explored the dimensions of informational, physical–psychological–social needs and healthcare support of the informants. The informants were reassured of confidentiality and anonymity.

Data analysis

The recorded interviews were transcribed verbatim for data analysis with the aid of NVivo V.12. The strategy of within-case and across-case analysis was employed,[12] where cases were the CLLC, parents and healthcare professionals. Data collection continued until saturation was reached. Two researchers independently reviewed the transcripts and segregated each portion of data containing one idea or piece of information into units of analysis.[14] The units of analysis with similar meaning were then organised into themes by two researchers. The initial agreement was 62.6% and improved to 88.7% with discussion. Participants were contacted for clarification of the transcripts where there was uncertainty between the researchers. Deliberation with the involvement of the experienced research team members helped to resolve any discrepancies of the codes. The themes and subthemes were compared and contrasted in the across-case analysis to explore similarities and variations in the perceived care needs of the three groups of participants.[12]

RESULTS

Table 1 shows the participant demographics. The mean ages of children, parents and healthcare professionals were, respectively, 13.5, 45.4 and 47.7 years. The gender split was 44.0% female CLLC, 68.0% mothers and 46.7% female healthcare providers. Twenty-eight per cent of the parents were in full-time employment. Among the 15 healthcare professionals, 86.7% were physicians and 15.3% were nurses; 66.7% of them had received palliative care training.

Table 2 shows the distribution by diagnoses and categories defined by Together for Short Lives.[13] There was a wide range of diagnoses. The distribution of categories were: I—curative treatment feasible (8.0%), II—long periods of intensive treatment needed (20%), III—without curative treatments (32.0%) and IV—irreversible but non-progressive (40.0%).

A total of 3784 units of analysis were analysed, resulting in three themes, namely ‘information and understanding about the

Table 3 Quotes for ‘information and understanding about the disease’

	Child with life-limiting condition (C)	Parents (P)	Healthcare professionals (H)	Similarities/differences
1.1 Information and communication process				
1.1.1 Information content and source	The doctor said that my condition is unpredictable. (C19) I browse the internet to learn about my disease, symptoms, how to do exercises. (C6)	The doctor talked to me for an hour, excited about the discovery of such a rare disease … related syndrome, and X-ray investigations … (P16) I searched on Facebook and found a Jarcho-Levin syndrome group, and learned about the difference between spondylothoracic dysplasia and spondylocostal dysostosis. (P16) We learned from other patients of the association of mucopolysaccharidoses … there is an allowance that we can apply for. (P6)	Told (the parent) that the disease is genetic-related, cannot be cured, will affect musculoskeletal function, eating and others, may need oxygen in the long run, will be getting worse, the child may live up to around 20 years. (H5) Nowadays, prefer to prescribe steroids early … told them all the things, well prepared … to take medication, see dietician, prepare for anything that needs to be prevented. (H1)	**Similarities** ▶ Doctor was a primary source for providing information in diagnosis and treatment **Differences** ▶ In addition to the medical information, CLLC and parents sought other sources of information on internet and social media
1.1.2 Process of communication	The doctor mainly talked to my mother when I was small. (C6) My parents would help me write down the areas of concern when consulting the doctor. (C2)	The paediatrician said that my son has many problems, then referred us to orthopaedics, neurology … but the specialists did not explain much to me … they would just focus on the surgery that they were responsible for … the ENT doctor said that the reconstruction surgery could help better breathing … the neurologist said that the surgery was risky … (P20) I feel very helpless. … Why did they suggest surgery in the first instance? It scared me! The doctor said that if the blood test was positive, then the diagnosis would be confirmed. If the blood test was negative, it didn’t mean the problem didn’t exist and surgery needed to be done [to confirm the diagnosis]. I could accept that, but s/he should have told me blood test first, and not surgery. (P14)	To be honest, I seldom talk to the child … I worry that the child is unhappy … I usually communicate with the mother. (H3) The child has physical problem but s/he is capable cognitively, one doctor will focus on cardiac, the other on dental and another on orthopaedics … we hope the parent will integrate the information for the child. (H14) Time is limited … we have consultations with many patients in a day. (H3) We focused on the treatment plan, especially the first few years (of the disease) … it takes time to build a rapport … I rely on the nurse to listen more and address concerns of the parents … (H15)	**Similarities** ▶ The parent was the key person in communicating with the doctor as well as the CLLC ▶ Need for multiple specialty consultations **Differences** ▶ Parents found the multiple consultations confusing ▶ Doctors felt that they mainly focused on treatment and relied on the nurse to listen and address concerns of the parents
1.2 Understanding of child’s diagnosis and treatment process				
1.2.1 Child’s diagnosis		Took a number of years to reach a diagnosis … consulted paediatrics, orthopaedics, neurology … the child’s muscles were weak … they took blood for investigation … didn’t help with genetic investigation … finally diagnosed at age 9–10. (P4)	When the child was born, problems were identified… there were repeated blisters in the arms and legs … we worried that it was a genetic problem… took a skin biopsy and performed genetic testing, and finally reached the diagnosis. (H9) The diagnostic process can be very challenging and take a long time … (H4)	**Similarities** ▶ Problems related to health and development were noted in early stage of life ▶ Confirmation of diagnosis took a long time
1.2.2 Treatment process	I have a problem with sugar, and the medication provides me with an enzyme supplement. (C20) I go for acupuncture on weekends. (C3)	With him growing up and experiencing progressively limited chest expansion … there were more episodes of pneumonia … 35 times this year. (P16) The spinal curvature is getting worse, and the doctor suggested more surgeries; we discussed this in the family and dad did not want to take this big risk … (P13) The hospital integrates western and Chinese medicine, so we learned about the effect of acupuncture … there is research evidence, so later on I sought help with acupuncture and massage myself. (P7)	We worked with neurology and respiratory specialties … the child is slow in walking and needs to use a wheelchair … may need to use BIPAP at night. (H14) The surgery would be palliative to improve the spinal curvature and expand the chest … but the mother didn’t want to take the risk and make the child suffer from the surgery … so the surgery was not done in the end. (H5) I am open to alternative therapy, since the disease is incurable; my principle is that it does no harm … (H4)	**Similarities** ▶ Long-term treatment and medication for symptom management ▶ Repeated surgeries ▶ Parents weighed the risk and benefit of the surgeries **Differences** ▶ Parents and CLLC sought help from complementary therapy ▶ No provision of alternative therapy in the hospital though the doctor did not object to it

BIPAP, bilevel positive airway pressure; CLLC, children with life-limiting condition; ENT, ear, nose and throat.

disease’, ‘living with the disease’, and ‘care support and palliative care’ which, respectively, occupied 27.4%, 55.8% and 16.8% of the units. The parents’ interviews (51.8%) provided the largest number of units of analysis, followed by healthcare professionals (24.7%) and children (23.5%). The richness of the data collected allowed the researchers to gain in-depth insights into the CLLC’s perceived care needs. Tables 3–5 provide anonymised supporting quotes of each of the themes and their subthemes. In each of the tables, the quotes from the three groups of participants are provided side by side for easy cross-reference. Similarities and differences between the care perceptions among the participants are shown at the end column of the table. Below is a description of each theme and subtheme.

Information and understanding about the disease

This theme concerns the information delivered regarding the disease or condition and how the information was understood. There are two subthemes, ‘information and communication process’ and ‘understanding the child’s condition and treatment process’. The first subtheme revealed that doctors were the main source of information, and they believed they provided adequate necessary information. Nevertheless, parents often had to seek further information from the internet. Self-initiated connections with patient groups with the same condition were very useful. Medical information from different specialists was reported as being fragmented and some participants did not fully understand the overall management plan. Doctors acknowledged both the limited time for consultations and the fact they usually focused on treatment while relying on nurses for subsequent elaboration and counselling (table 3).

Findings in the second subtheme, ‘understanding the child’s diagnosis and treatment process’, showed that the process of delineating diagnosis was lengthy and often involved extensive investigations. Parents reflected that treatment plans were demanding and complicated. Parents often had to evaluate the treatment options themselves. Some parents sought out alternative therapies, which they believed were more readily acceptable and effective.

Living with the disease

This theme captured how the CLLC and their parents lived with the disease. There are three subthemes: *living with physical concerns*, *living with non-physical concerns*, and *life perspectives*.

Table 4 Quotes for 'living with the disease'

	Child with life-limiting condition (C)	Parents (P)	Healthcare professionals (H)	Similarities/differences
2.1 Living with the physical concerns				
2.1.1 Impacts of symptoms and daily care management	Pain in spine, foot, buttock. (C1) Loss of balance … I need help for bathing until I am over 10 years old … I have to wear a pullover, but for school uniform I need my mother to help me button my shirt. (C19) My mother does chest tapping for me. (C14)	Needs to use ventilator at night … oxygen 24 hours and suctioning. (P16) We keep him company all the time … for places that he cannot walk or use a wheelchair. (P19) The difficult part is suctioning: the procedure involves hand washing, gloving, then letting him breathe in-between and suctioning again. (P16) I learned Qi and massage myself to help (the child). (P7)	There are multiple problems, and we take care of them one by one … for seizures, we prescribe anti-seizure medication … for muscle tension, a relaxant … for eating problems, a speech therapist. (H4) The patient is too big to be lifted and transferred … we now arrange for a wheelchair to reduce the burden of the mother. (H12) We discussed it with the family and referred the patient to an ENT to perform a tracheostomy to reduce the patient's efforts for breathing … now there is no more need for the ventilator … (H8)	**Similarities** ▶ CLLC encountered multiple symptoms ▶ Parents provided home care support in daily living and performed special caretaking tasks **Differences** ▶ Healthcare providers prescribed medication, arranged medical equipment and made referrals for home care. Parents found solutions to adapt to day-to-day living
2.1.2 Parental burden	I think respite care is necessary if available since it's very burdensome for mom and she needs some rest … but I feel that she would only take a day or two since she worries about me. (C6)	My hand is so painful that I cannot do stir fry (for cooking) … it has become a chronic problem. (P6) My child has seizures, and I quit my job 14 years ago to take care of him. (P11) I won't leave my child with someone else even there will be a respite care service because I'm the only one who knows if something is wrong with them. My child is used to being cared for by me and my parents, not others. (P8)	The child has many specialty follow-ups, including neurology, orthopaedics, ENT … and training in school … the mother has no time to attend to her own needs. (H12) I think it would be good to provide parents some space to do their own things, even a break or respite care once a quarter if available… some coordination of care in follow-up appointments can contribute to better quality of life and reduce the stress of caregiving. (H15)	**Similarities** ▶ Parental burden was acknowledged ▶ Parental burden could hamper parents' quality of life and health ▶ CLLC and healthcare providers accepted respite care as a source of relief for parents if available **Differences** ▶ Parents chose to care for the CLLC themselves even if there is a choice of respite care service
2.2 Living with the non-physical concerns				
2.2.1 Psychological and emotional responses	I am afraid of falling … (C6), … surgery.… (C7), death. (C8) I am unhappy, passers-by look at me strangely. (C15)	Fear of (child's) deterioration … (P14) … no one knows how to take care of him. (P13) Now he's in a wheelchair and other people look at him in a special way … he is stigmatized. (P6)	Parents are worried about their child's deterioration … (H15) The child feels like a deviant … the parent feels they are not being respected (by strangers). (H10)	**Similarities** ▶ Presence of fear and worries among CLLC and parents ▶ Stigmatisation
2.2.2 Coping strategies	I treat my disease as my friend. (C6) Do things I like, for example, drawing and computer games. (C14)	Let the child do things s/he likes, for example, listening to music, eating … s/he will be happier. (P2) I bought him a computer for playing video games. (P12)	There are bound to be a lot of emotions (of child and parents) … we only have 5–10 min of consultation … (H3) There is a need for nursing support; the doctor will address medical issues, while the nurse hopefully can do something for the psychosocial aspects. (H4) As a nurse, I am the person who knows the internal and external environment … to fill the communication gap in caretaking (between doctor and parents). (H15)	**Similarities** ▶ CLLC with support of their parents tried to live with the disease by doing things they liked **Differences** ▶ Medical professionals noted the need to support parents to cope with the emotional responses and the nurse would provide psychological support to fill the gap
2.2.3 Family and social support	Everyone speaks very fast, but my dad and mom talk to me slowly … (C7) Family arguments can greatly affect my daily mood. (C14) My classmates are supportive … they listen to my grumbling. (C6) There is sponsorship for the ventilator … the nurse asked the doctor to write a letter … I rely on the ventilator. (C1)	(The child) always asks me to do things for him; his hands can't move. (P1) There is an impact on my relationship with his father … he feels that he needs personal time after work, so he does not stay at home… we have talked about separation and need help from the social worker. (P14) The Christian faith helps with support from the church. (P2)	The carer stress can be tremendous, and sometimes creates conflict between couples. (H5) We have communication with the school so that they can take note of the special needs of the child. (H5) The monthly rent for the ventilator is very high … so we help to apply to the Samaritan fund for the child. (H11)	**Similarities** ▶ Family support existed ▶ Caregiver stress strained couple relationships and affected CLLC ▶ Tangible social support such as financial assistance was made available **Differences** ▶ CLLC need family and social support, and church provides spiritual comfort absent in medical care
2.3 Life perspectives				

Continued

Table 4 Continued

	Child with life-limiting condition (C)	Parents (P)	Healthcare professionals (H)	Similarities/differences
2.3.1 Quality of life	(Quality of life is) eating well (C14), playing (C11), dancing. (C24)	Kids like to play, eat out, be happy (P8). It goes by stages … In hospital, the goal was to survive until age 6… In primary school, the goal was to be less sick and eventually to live… Now that the condition is stable, the goal is for my child to live well and participate in extracurricular activities. (P16)	I think cognition is important for quality of life … he can learn and communicate … physical ability affects quality of life, but he is not too bad overall. (H14) The happiest thing for him is to play on the computer, do things he likes online. (H5)	**Similarities** ▶ Quality of life was to maintain normalcy of life, to play and eat and be happy **Differences** ▶ Parents set expectations of life according to the CLLC's stage of condition
2.3.2 Education, growth and employment	Book knowledge is not good enough … you have to gain knowledge and exposure outside book learning. (C16) I want to study accounting. (C13) … I'm learning cooking from my uncle. (C1)	I don't put pressure on him … I don't know how many days he has left. (P12) I was thinking accounting would be good for her, because she can sit and won't need to stand all day. (P6) I told her to work in nurseries, feeding babies … she can do it and she likes babies. (P11)	I wish he could attend school for his growth and social development… like other kids… but his chronic illness limits his exposure. School is essential for his development and social interaction. (H3) Despite having limited fingers, she enjoys drawing. The school's career planning program matches her interests and abilities to appropriate subjects, supporting her pursuit of interests. (H7)	**Similarities** ▶ Needs for growth and development ▶ Had plans for education and career development **Differences** ▶ CLLC and parents set life goals to suit the children's ability and limitations

CLLC, children with life-limiting condition; ENT, ear, nose and throat.

Table 5 Quotes for 'care support and palliative care'

	Child with life-limiting condition (C)	Parents (P)	Healthcare professionals (H)	Similarities/differences
3.1 Care support and coordination	Eye, paediatric … dental, ENT … orthopaedics … I remember there are 7 specialties [I need to attend] … half of the time of a certain month is attending medical follow-ups. (C16) … talked about medication, physical condition … little discussion about daily living [consultation]. (C5)	We wait for a long time and are only seen for 2 minutes … and then on to the next appointment … it takes much effort for the follow-up every time … we have to book a car … it takes one whole day for a follow-up. (P14) For the spinal curvature, we go to Hospital A … for neurological surgery we go to Hospital B, for child brain check-up we go to Hospital C … then to Hospital D for physiotherapy. (P3) The doctors in the public hospitals usually just asked if there was anything special, any seizure, any drug side-effects, then continued the prescription, that's it. (P25)	We should think of supporting the child for normal living … we (try to) connect with specialties in our own cluster … but our request may not be entertained. (H13) There should be better collaboration between paediatricians and adult specialty physicians … the transition from child to adult care is a big problem for the chronically ill. (H11)	**Similarities** ▶ Needed better coordination and collaboration among multiple specialty consultations ▶ Healthcare mainly focused on physical care **Differences** ▶ The poor transitional care from paediatric to adult care was noted by the healthcare professionals
3.2 Understanding of palliative care	(Palliative care) is for those who are severely ill, like cancer patients … (C6) I can communicate with the home palliative care nurse to arrange faster admission to the hospital. (C13) It would be helpful to have a team that knew everything about us … there would be no need to have multiple appointments. (C19)	It seems to be an infirmary place for older people … (P15) If the doctor says the child's life expectancy will be short, I feel that the support should not wait until the end. (P10) I am grateful to the palliative care team. (P12) Currently there is nurse Y who takes care of our case, a bridge in communicating with the doctor … it is much more convenient, because we just need to seek help from Nurse Y. (P14)	(Palliative care) is physical-psycho-social-spiritual care … family-centred … requires support from many team members … There is a contact point in the hospital … and many of the services needed by the child are in the community. (H2) Specialized paediatric nurses can provide support to mothers at home, for example, tracheostomy care … it is challenging for mothers to take care of children with complex needs at home … (H11) I believe once diagnosis is made, we should start involving the palliative care team … co-walk with the parents … it'll also help the family to accept the death (of the child). (H4) We are trying very hard to educate [the younger doctors] to sign the advanced care planning form, it's just at the beginning stage. (H11)	**Similarities** ▶ Palliative care services facilitated the provision of holistic care ▶ Nurse role was crucial in care coordination in a multidisciplinary team **Differences** ▶ Varied understanding of palliative care concept among CLLC and parents ▶ Palliative care service was not provided in all settings and just at the beginning stage

CLLC, children with life-limiting condition; ENT, ear, nose and throat.

The subtheme ‘living with physical concerns’ revealed that CLLC faced various distressing symptoms. Parents had to perform special nursing care at home, with prior training provided by healthcare professionals. The parental caring burden was worsened with the lack of community resources such as limited availability of respite care. Caring for the CLLC significantly impacted parents’ quality of life (table 4).

The subcategory ‘living with non-physical concerns’ made up the largest portion of difficulty living with the disease. CLLC and parents experienced fear, worries and stigma. Healthcare professionals recognised the children’s and parents’ emotions, but time constraints in the clinic meant doctors had limited time to address their psychosocial concerns. Nurses frequently filled the gap providing emotional support. Carers’ stress led to tension and conflicts between couples. Participants said that social support and financial assistance provided by social workers could ease their stress. Some parents also found spiritual support from church helpful.

The subtheme ‘life perspectives’ revealed that the children interpreted quality of life as being able to eat, play and be happy, whereas parents were already grateful for seeing the CLLC able to study in school. Healthcare professionals were generally supportive in facilitating the CLLC and their parents to pursue their wishes. The CLLC and parents reported grasping every opportunity to achieve a reasonable quality of life and maximise normalcy during the children’s limited lifespan.

Care support and palliative care

This theme explored the availability of care support and the participants’ understanding of palliative care. The subtheme ‘care support and coordination’ revealed that both the CLLC and parents opined hospital support focused primarily on the physical aspects of care. However, opinions from different professional consultations were often divided and even contradictory. There was a perceived need for better coordination and collaboration among multiple specialties. Poor transitional care from paediatric to adult service was noted by healthcare professionals (table 5).

The second subtheme was ‘understanding of palliative care’. Informants said that palliative care could be helpful in addressing the CLLC’s needs in a holistic manner but some parents misunderstood palliative care as only applying to patients with cancer or the elderly. Palliative care services were not available in all settings, but those who received palliative care valued it highly. Some healthcare professionals said that in an ideal world, services and palliative care coordinated by a key worker preferably provided by a primary nurse were crucial to facilitate interdisciplinary communication.

DISCUSSION

By simultaneously investigating the perspectives of these three important informant groups, this innovative study using within-case and across-case analysis has identified an important service gap in addressing the care needs of the CLLC. Previous studies were limited by focusing on only one or two groups. Although the CLLC, parents and healthcare providers identified similar care needs for the CLLC, they varied in how they interpreted them. Perhaps unsurprisingly given their roles, healthcare providers viewed the care concerns from medical perspectives, while the CLLC and parents contextualised the needs in overcoming challenges in their daily living environments. These differing perceptions of care problems result in a significant service gap, and parents and children expressed confusion about the fragmentation of care. Narrowing this gap requires a strategy that integrates multidisciplinary team efforts, particularly including the voices of parents and CLLC with shared decision-making in formulating the care plan.

The palliative care approach is a promising way to address the service gap. This approach advocates improving the quality of life for those with life-threatening illnesses through early identification and management of problems in a holistic manner with coordinated multidisciplinary efforts.[15] Dewan and Cohen[16] have proposed engagement of a key worker as a single point of contact to assist care coordination. A narrative review suggests that the nurse is most appropriate to assume the care coordination role.[17] The nurse adopts a multifaceted role in PPC, acting as a direct care provider, counsellor, and advocate for the patient and family while functioning as a coordinator within the healthcare team. Care coordination by the nurse facilitates effective communication among multiple specialists, and between the care team and the clients.[18] Informants of this study who experienced the advantages of a nurse coordinator attested to this.

Early integration of PPC can support the CLLC and family in making sound and realistic decisions,[6] symptom management facilitated by better care coordination.[19] It is advocated that PPC should be introduced at the time of diagnosis of the life-limiting condition, or prior to diagnosis where it can be challenging for rare conditions.[10] The Strategic Service Framework for palliative care introduced in Hong Kong in 2017 helped promote PPC, but the service remains underdeveloped. Not all hospitals have designated teams to provide PPC. Only two out of the five study hospitals had designated PPC team, and other hospitals provided PPC services alongside day-to-day services.

Parents often act as surrogate decision-makers, with the children as passive recipients.[20] The CLLC here wanted to live a normal life like other children, enjoy friends and schooling and make plans for the future.[2] There is an urgent appeal to involve parents as essential members of the caring team.[21] Formation of true partnerships requires healthcare experts to flatten the hierarchical relationships, to enhance reciprocal information exchange[22] and involve the families as co-creators in the care coordination processes with shared decision-making.[23] Caretaking of CLLC bears impacts on the quality of life of families.[20] Studies have frequently reported long hours in caregiving lead to changes in employment status, income[24] and quality of life[25] of parents. As the present findings imply, the outcomes could be poor family functioning[6] and increased carer burden.[26] Goldhagen *et al*[27] suggested reinforcement of community support and community-based PPC is essential to help attain the health-related quality of life of caretakers.

CONCLUSIONS

This original study examining the perspectives of the CLLC, parents and healthcare providers disclosed an important service gap in addressing the care needs of the CLLC. The palliative care approach with active engagement of the parents is a promising way to address the gap. The results showed that all three stakeholders agreed that the most pressing need was support for living with the disease. Parents are identified as the main caregivers, and their involvement in care plan formulation was deemed essential. This study recommends improved coordination of care with a designated nurse and advocates introduction of PPC at an early stage even before the diagnosis can be confirmed. The findings have provided evidence for policymakers to consider allocating appropriate resources to fill the service gap for this vulnerable group.

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