

Scenarios of sharing medical care between parents and healthcare professionals for children with profound intellectual and multiple disabilities who live at home: A qualitative study

Journal:	<i>Journal of Child Health Care</i>
Manuscript ID	JCHC-2023-0192
Manuscript Type:	Original Article
Keyword:	Parents' experiences, Child with profound intellectual and multiple disabilities, Shared decision making, Family Centred Care, Humanization of healthcare policy
Abstract:	<p>Background: Many children with profound intellectual and multiple disabilities are cared for by their parents at home. Childcare often includes medical care. Little is known about how parents and healthcare professionals share the medical care for the child. Aims: The study aims to contribute to the understanding of how the provision of medical care and associated interactions between parents and healthcare professionals are achieved and of how healthcare professionals can align care to family needs. Methods: The design was inspired by grounded theory. We analysed in-depth interviews with 25 Dutch parents. Findings: Our analysis identified five dimensions that together reflect experiences with medical care: fragility, planned care, irregularities, the presence of providers, and parents' preferences. We proposed three scenarios in which dimensions interplayed and that characterized different interactions between parents and healthcare professionals: dependent care, dialogical care, and autonomous care. Conclusions and implications: Findings accentuated the need to communicate and evaluate patterns of sharing care with parents. Healthcare professionals may broaden the relational work of shared decision making to concrete scenarios in sharing medical care. Commitment to parents' autonomy implies that healthcare professionals should be attentive to the parents' emotional and relational needs.</p>

SCHOLARONE™
Manuscripts

Title

Scenarios of sharing medical care between parents and healthcare professionals for children with profound intellectual and multiple disabilities who live at home: A qualitative study

Abstract

Background: Many children with profound intellectual and multiple disabilities are cared for by their parents at home. Childcare often includes medical care. Little is known about how parents and healthcare professionals share the medical care for the child. **Aims:** The study aims to contribute to the understanding of how the provision of medical care and associated interactions between parents and healthcare professionals are achieved and of how healthcare professionals can align care to family needs. **Methods:** The design was inspired by grounded theory. We analysed in-depth interviews with 25 Dutch parents. **Findings:** Our analysis identified five dimensions that together reflect experiences with medical care: fragility, planned care, irregularities, the presence of providers, and parents' preferences. We proposed three scenarios in which dimensions interplayed and that characterized different interactions between parents and healthcare professionals: dependent care, dialogical care, and autonomous care. **Conclusions and implications:** Findings accentuated the need to communicate and evaluate patterns of sharing care with parents. Healthcare professionals may broaden the relational work of shared decision making to concrete scenarios in sharing medical care. Commitment to parents' autonomy implies that healthcare professionals should be attentive to the parents' emotional and relational needs.

Keywords: Parents' experiences, child with profound intellectual and multiple disabilities, children with medical complexity, shared decision making, family centred care, humanization of healthcare policy

Introduction

1
2
3 Children with profound intellectual and multiple disabilities (PIMD) have a
4 combination of severe cognitive, neuromotor and sensory impairments (Nakken and
5 Vlaskamp, 2007). Possible underlying conditions are chromosomal anomalies, metabolic
6 diseases, deviated brain formation and acquired brain damage (Jansen, 2015; Nakken and
7 Vlaskamp, 2007). Associated with disabilities, the children often have medical complexities
8 such as gastrointestinal, respiratory and feedings difficulties, epilepsy syndromes, spasticity
9 and scoliosis (Van der Putten et al., 2017). Nonverbal expressions as sounds and muscle
10 tensions are often the only form of communication (Petry and Meas, 2006; Van der Putten et
11 al., 2017). Children with PIMD form a specific group among the larger and heterogenous
12 group of children with medical complexity (CMC)(Cohen et al., 2011; ICPCN, 2022).
13
14
15
16
17
18
19
20
21
22
23
24
25

26
27 In high income countries, many children with PIMD are cared for by their parents and
28 live at home (Breitkreuz et al., 2014; Luijkx et al., 2017). Components of caring for a child
29 with PIMD at home are, for example, continuously monitoring the child's health, managing
30 medication and therapies, and accessing equipment and resources like help with childcare
31 (Luijkx et al., 2019; Whiting, 2014; Woodgate et al., 2015)(Author 2019, 2021). Parents deal
32 with a wide network of healthcare professionals (HCPs) who are involved in monitoring the
33 development and health of the child. Care involves deliberating the child's health, dignity and
34 quality of life and death, and making medical decisions for acute emergencies and advance
35 care planning (Kruithof et al., 2022a; Loeffen et al., 2018; Olsman et al., 2022). Because
36 parents and HCPs are mutually dependent effective relationships between them are key in
37 childcare.
38
39
40
41
42
43
44
45
46
47
48
49
50
51

52
53 Previous studies described that it is often difficult in practice for parents to build
54 relationships with HCPs that are effective and grounded in equal cooperation (Kruithof et al.,
55 2022b; Smith et al., 2015; Zaal-Schuller et al., 2016). Models such as family centred care and
56 shared decision making are widespread in healthcare but implementation in everyday
57
58
59
60

1
2
3 practices is lagging behind (Elwyn, 2021; Smith et al., 2015). At the same time, parents
4
5 addressed they are not properly shouldered by healthcare and many parents felt forced to
6
7 become advocates for child and family (Kirk et al., 2005; Spiers and Beresford, 2017; Wij
8
9 zien je Wel, 2022). Caring duties continue around the clock and many parents take over and
10
11 continue medical procedures typically performed by HCPs (Kirk and Glendinning, 2002;
12
13 Woodgate et al., 2015). Little research has been done on how interactions between parents
14
15 and HCPs take place and on what indicates their burden or success (Jansen et al., 2018;
16
17 McDonald et al., 2016; Whiting, 2019). This study aims to contribute to understanding of how
18
19 the provision of medical care to children with PIMD at home and the associated interaction
20
21 with HCPs is achieved.
22
23
24
25

26
27 To contribute to understanding of how parents handle the medical care for the child at
28
29 home and of how providers can align care to family needs, we present a secondary analysis of
30
31 in-depth interviews with Dutch parents. The leading question in this analysis was: “What
32
33 dimensions affect the medical care provided by parents and healthcare professionals caring
34
35 for children with profound intellectual and multiple disabilities at home?”
36
37
38

39 **Methods**

40
41
42 This study is part of a larger research that explores the experiences of Dutch parents
43
44 who care for children with PIMD at home. The research included in-depth interviews with 25
45
46 parents. The original analysis focussed on the essence of the phenomenon “parenting a child
47
48 with PIMD” (Author, 2022). For this study, we reanalysed interviews and concentrated on
49
50 how parents deal with the provision of medical care to the child. We applied a grounded
51
52 theory (GT) inspired approach in the analytic process (Chun Tie et al., 2019; Corbin and
53
54 Strauss, 2014). This approach was chosen because it allowed us to explore the underlying
55
56
57
58
59
60

1
2
3 dynamics of the parents' perspectives and characteristics of healthcare delivery (Corbin and
4
5 Strauss, 2014; Tracy, 2020).
6
7

8 **Selections of participants**

9

10
11 The parents that participated in the study were engaged through our own network and
12
13 social media (N = 10), online community plus support networks (N = 6), specialized day care
14
15 and respite care services (N = 5) and paediatric healthcare professionals (N = 1). During the
16
17 course of the study the parents approached fellow parents they knew (N = 3). All parents had
18
19 cared for the child in the family home for a period of two years and longer in order to reach
20
21 thorough understanding of meaning. Furthermore, we aimed for a wide scope of the clinical
22
23 profiles of the children, diversity in parents' cultural, ideological and socioeconomic
24
25 backgrounds, and nationwide coverage. Bereaved parents were included in the study. We
26
27 started approaching parents in April 2019 and stopped once data saturation was obtained
28
29 (December 2020). We used Table 1 prepared for a previous study to provide information on
30
31 the final sample of 25 parents and their families (Author, 2022).
32
33
34
35

36
37 <TABLE 1>
38
39

40 **Ethics approval and consent to participate**

41

42
43 The research was approved by the Medical Research Ethics Committee. The parents'
44
45 rights as research participants were described in the invitation letter and informed consent
46
47 form. Information was also talked over by phone and again prior to or directly after the
48
49 interview. Parents were informed about the procedure of member reflections, possibility of
50
51 withdrawal and destruction of data during the course of the study, and storage of anonymised
52
53 transcripts at Anonymize following publication. All participants signed informed consent.
54
55
56

57 **Data collection**

58
59
60

1
2
3 Most interviews were at the parents' home (N = 21). Five interviews were with both
4 parents at the same time because this was preferred by them. Two individual parents favoured
5 a work related location for practical reasons. Another two single interviews were conducted
6 online via Microsoft Teams because of quarantine due to the COVID-19 pandemic. All
7 interviews were with the first author and in Dutch language. The interviews had an open and
8 nondirective style. We used a small topic list based on a previously published narrative
9 literature review to fall back on if needed (Author, 2019). Each interview started with an open
10 question: "Would you like to tell me about your family?" The interviews were recorded by a
11 voice recorder and took an average of 84 minutes.
12
13
14
15
16
17
18
19
20
21
22
23

24 **Data-analysis**

25
26
27 In the original study, interviews were transcribed verbatim. To identify the
28 information important for this secondary analysis, two authors (XX and XX) independently
29 reread the transcripts several times. We used ATLAS.ti for the iterative process of coding.
30 Inductive open coding was completed in two rounds: we first coded half of the interviews and
31 then developed a coding scheme and coded subsequent transcripts. The procedure of axial and
32 selective coding was conducted by all authors. We identified five dimensions in the
33 experiences of parents. Through reflection on the narrative storylines in the parents' accounts,
34 we distinguished three scenarios with interrelated dimensions that reflect interactions between
35 parents and HCPs regarding medical care.
36
37
38
39
40
41
42
43
44
45
46
47

48
49 Interpretation of data was discussed with parents through member reflections (Tracy,
50 2020). Findings were supplemented with a Dutch translation to allow participants to review
51 our interpretations and the choice and translation of quotes.
52
53
54
55

56 **Findings**

1
2
3 Central to the parents' experiences was the necessity to deal with complex medical
4 issues and decisions about provision of care. Parents conveyed the child's health played a
5 decisive role in the dynamics of day-to-day family life. This started with continuously
6
7
8
9
10 monitoring the child's wellbeing:

11
12
13 I always had to keep an eye on her. Is she having another (epileptic) attack, is
14 something else going on? When I started cooking I gave her a rattle because I knew
15 that if the rattle stopped I had to check on her, then something was wrong with her.
16
17
18

19
20 (T08)

21
22
23 All parents shared responsibility for the child's health with HCPs. However, the way they
24 shared care with providers differed between parents and frequently also by specific area of
25 care. Our analysis identified five dimensions affecting the medical care provided: (A)
26 "fragility", (B) "planned care", (C) "irregularities", (D) "the presence of providers", and (E)
27 "parents' preferences". We distinguished three different scenarios in which the dimensions
28 interplayed and that characterized different interactions between parents and HCPs:
29 "dependent care", "dialogical care", and "autonomous care". Within areas of care, changes in
30 dimensions (A up to E) led to changes of scenarios of interactions between parents and HCPs.
31
32
33
34
35
36
37
38
39
40

41 42 **Dimensions that established medical care**

43
44
45 The child's fragile health and physical vulnerabilities of the time formed the basis of
46 medical care provided at home (A: "fragility"). The parents in our study cared for children
47 with diverse syndromes and disorders. Health issues they were dealing with were, for
48 example, feeding difficulties, epileptic seizures, coughing attacks, respiratory distress and
49 susceptibility to infections. These conditions were not stable, however, so parents had to
50 manage caring responsibilities accordingly:
51
52
53
54
55
56
57
58
59
60

1
2
3 Now she is doing quite well. Now I can do laundry without the baby monitor, or go
4 upstairs for a short moment. Or go outside, even. When I get back, I take a look right
5 away. If she is not that well, I always have the baby monitor with me, and I do not do
6 laundry. Because then I am, then I might be too late. (T09)
7
8
9
10
11
12

13 Parallel to fragility of the child's health, parents dealt with medical interventions that
14 were necessary and that sometimes became a stable part of day-to-day routines (B: "planned
15 care"). Parents described care routines in which they, for example, gave parenteral nutrition,
16 administered medicines orally or by feeding tube, and did bowel washouts. The use of
17 healthcare technology like oxygen saturation monitors, feeding tubes, and suctioning
18 machines therefore was an integrated component of everyday life:
19
20
21
22
23
24
25
26

27 He has to be catheterized five times a day. We have to give him enemas. That is every
28 other day. And he has a feeding tube. (T04)
29
30
31
32

33 In contrast with the rhythm of many medical interventions and procedures, caring for
34 the child often involved irregularities or emergencies (C: "irregularities"). Parents described
35 complications like, for example, changes in consciousness, breathing stops and problems with
36 feeding tubes such as unwanted removal of the tube or skin lesion. Examples ranged from
37 situations in which they had to act immediately to situations in which they had to await what
38 would happen, such as with acute high fever. The changeability of the child's health meant
39 that situations in which parents had gradually become comfortable could suddenly turn into a
40 situation in which they felt unfamiliar and unsafe:
41
42
43
44
45
46
47
48
49
50

51 You need to be able to make the right estimations. Is this still ok? Do we need to call
52 the emergency services? But in the meantime we have already started acting ourselves.
53
54
55
56 (T01)
57
58
59
60

1
2
3 How parents perceived the presence and role of healthcare services and HCPs
4
5 involved also affected the provision of medical care and the way parents and providers shared
6
7 medical care (D: “the presence of providers”). Parents conveyed that HCPs like paediatricians
8
9 or neurologists had central roles in choices for interventions that became part of day-to-day
10
11 childcare. Sometimes this was done in consultation with parents or cooperatively in
12
13 specialized teams. Many parents were taught how to perform medical interventions by HCPs
14
15 during periods of hospitalization or through specialized homecare. Parents’ accounts about the
16
17 role of HCPs in more stable periods varied. Some parents reported that HCPs provided some
18
19 of the day-to-day childcare including medical interventions at home or at day care or respite
20
21 facilities. In other cases, parents did much themselves. Parents’ satisfaction with interactions
22
23 with HCPs and outcomes of care differed widely. Many parents had good experiences with
24
25 providers. However, there were also many parents who conveyed they no longer entrusted
26
27 their child to HCPs:
28
29
30
31
32

33 In that period he came home (from day care) with aspiration pneumonia all the time.
34
35 The last time was after it took us six weeks to get him better with antibiotics and
36
37 oxygen and nebulization. After six weeks he was well. And within three days he was
38
39 brought home again, all rasping and blue. (...) After that happened I decided, from
40
41 now on he stays at home, period. (T16)
42
43
44
45

46 The parents who had negative experiences with HCPs in childcare indicated the lack of trust
47
48 created a distance between themselves and providers. This distance became increasingly
49
50 difficult to bridge. Parents were outraged that the HCPs involved treated the child in such a
51
52 way that the child’s wellbeing was compromised.
53
54

55 The parents’ preferences regarding their own roles also affected the provision of
56
57 medical care (E: “parents’ preferences”). Most parents had no medical background and doing
58
59 medical interventions was often quite challenging both technically and emotionally.
60

1
2
3 Moreover, providing medical care to the child conflicted with their parenting role. They
4 preferred to only perform medical procedures that they felt at ease with. However, all parents
5 learned at least some medical skills as this was inherent to caring for the child at home.
6
7 Parents actively developed in dealing with medical aspects of childcare. They lived together
8 with the child and therefore learned by trial and error. In order to improve, they used a broad
9 range of resources, such as input from HCPs, academic or professional articles available
10 online, and recommendations of fellow parents. Parents indicated that their abilities and
11 confidence grew because of the experiential knowledge gained, although many parents were
12 sometimes unsure and doubted whether their actions were good enough. In addition to the
13 ambiguities mentioned, broader family circumstances also affected the parents' preferences
14 for larger or smaller roles in medical care. Examples were the presence of other children and
15 the place where parents lived. Parents' role did differ per area of medical care.
16
17
18
19
20
21
22
23
24
25
26
27
28
29

30 31 **Scenarios of interactions between parents and HCPs**

32
33
34 Interactions between parents and HCPs can be characterized in three scenarios (see
35 Table 2). The scenarios different between and within practices of home caring by parents.
36
37 This meant that parents and HCPs sometimes interacted through different patterns per specific
38 medical issue in time.
39
40
41
42
43

44 <TABLE 2>

45 46 47 ***Scenario of dependent care***

48
49
50 In this scenario parents relied on HCPs while cooperating closely together. This
51 scenario often occurred when medical complexities were high or new (A: "fragility) or where
52 there were high risks of complications (C: "irregularities"). HCPs played an important role in
53 day-to-day caring for the child at home or elsewhere, such as at day care or respite care
54 facilities. This pattern also occurred in more stable situations where medical care was less
55
56
57
58
59
60

1
2
3 complex or at least anticipated (B: “planned care”). In these situations, parents preferred a
4
5 greater role for HCPs or were unable to take on a larger role themselves (E: “parents’
6
7 preferences”). Within this scenario, parents were unable or unwilling to bear the responsibility
8
9 for the medical care concerned. They perceived HCPs as experts who knew what was needed
10
11 when, and felt that child and family were well cared for (D: “the presence of providers”). In
12
13 these situations, parents felt adequacy supported in coping with distress associated with
14
15 medical aspects of childcare:
16
17
18
19

20 At a certain point we could choose between a personal care budget and what we had.
21
22 The care we received went well, so we did not opt for the personal care budget. We
23
24 can only think very positively about that time. (...) They understood things and knew
25
26 what to do. They came to us, so the boys were cared for in their own environment.
27
28
29 (T05)
30
31

32 *Scenario of dialogical care*

33
34

35 The second way of interacting with HCPs meant parents partly provided medical care
36
37 to the child themselves and partly relied on HCPs. This scenario occurred in a variety of
38
39 health situations (A: “fragility”). Within this scenario medical care was reasonably stable and
40
41 parents had already gained some experience in specific care routines. Parents had well defined
42
43 ideas and about their own roles in medical care and corresponding arrangements with
44
45 providers (E: “parents’ preferences”). For example, some parents explicated they had
46
47 professional backup for interventions or procedures that they would rather not do and learn. It
48
49 was characteristic within this scenario that parents and HCPs communicated equally with one
50
51 another about the practicalities of day-to-day medical care (B: “planned care”). This included
52
53 acting and cooperating in worrisome or emergency situations (C: “irregularities”). Parents had
54
55 clear expectations of healthcare contributions and confidence in HCPs involved in childcare
56
57 (D: “the presence of providers”). Dialogues with providers frequently also addressed
58
59
60

1
2
3 longtermly medical issues and their implications for quality of life for the child and family.

4
5 Parents indicated this was important to them:

6
7
8 I really like it when you, as a parent, feel that you can discuss things with someone.

9
10 (...) That providers rely on your knowledge and your experience with your child.

11
12
13 (T20)

14 15 16 *Scenario of autonomous care*

17
18
19 The third way of interacting with HCPs existed of parents arranging care more
20 autonomous and independent from the traditional healthcare system. They had often created
21 their own network of self-employed HCPs and personal connections to organize childcare (E:
22 “parents’ preferences”). Characteristic was that parents had developed clear visions on quality
23 of care, life and death. They felt well attuned to child and family:

24
25
26 We have a personal care budget for her. Care is arranged from that budget. We placed
27 her in the heart of that care. Just as her siblings are central in their own lives. (...) We
28 divided her care over five days, each with its own schedule. In this way the care does
29 not become too heavy for carers. (...) This way we can care for her at home for a long
30 time. (T14)

31
32
33 In this scenario, childcare had to be in line with parents' own ideas and values as much as
34 possible. Parents felt that regular providers were often not flexible or receptive enough (D:
35 “the presence of providers”). Other parents were professionals themselves and explained that
36 an autonomous approach felt more natural to them. Within this scenario, parents performed
37 many medical interventions and procedures independently and taught others involved in
38 childcare how to operate (B: “planned care”). When the medical situation of the child partly
39 changed (A: “fragility”) or when unexpected complications appeared (C: “irregularities”), the
40 parents had to rely on regular healthcare services such as emergency services and specialized
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

1
2
3 care units. However, they regularly encountered customs and values that were dominant there
4
5 but clashed with their own views on quality of care.
6
7

8 **Changeability and mobility of scenarios**

9

10
11 The parents' accounts revealed that changes in the nature of dimensions led to changes
12
13 in the interrelatedness of dimensions and therefore to changes in parents' interactions with
14
15 HCPs. For example, sudden emergencies (C: "irregularities") could lead to deterioration of
16
17 the child's health (A: "fragility") and changes in day-to-day routines (B: "planned care").
18
19 Consequently, parents had to find new ways to share care with providers. Viewed through the
20
21 lens of the scenarios, this meant, for example, that parents had to temporarily switch from
22
23 "dialogical care" or "autonomous care" to "dependent care" for particular areas of medical
24
25 care. Another commonly occurring change of scenario was the move towards interactions
26
27 typical for "autonomous care". This often happened when actions of HCPs were not in line
28
29 with parents' expectations (D: "the presence of providers"). Parents' own modes of dealing
30
31 with medical care changed because they were not satisfied by the care provided (E: "parents'
32
33 preferences"). Parents were also increasingly able to facilitate changes because they knew
34
35 better and better how to implement their own ideas about optimum childcare and wellbeing
36
37 for child and family.
38
39
40
41
42
43

44 **Discussion**

45

46
47 With this study, we aimed to contribute to understanding of how parents deal with
48
49 medical care at home in interaction with providers. The central question guiding the study
50
51 was: "What dimensions affect the medical care provided by parents and healthcare
52
53 professionals caring for children with profound intellectual and multiple disabilities at
54
55 home?". Our findings illuminated that medical care was established by an interplay of
56
57 dimensions that were partly medical in nature and partly related to parents' satisfaction with
58
59
60

1
2
3 interactions with HCPs. When the child's health deteriorated, the medical interventions
4 considered necessary at the time were decisive and the parents had to adapt to what was
5 happening medically. When there was a greater degree of stability and planned care, the
6 parents' interactions with HCPs were decisive for the way medical care was delivered.
7
8 Findings highlighted that the way parents perceived the quality of care provided by HCPs
9 strongly affected how they viewed their own role in medical care, and how they preferred to
10 organize and share care with providers.
11
12

13
14
15
16
17
18
19
20 The study accentuated the importance of enabling parents to communicate their
21 preferred way of interacting with HCPs in particular areas of care. Findings revealed that
22 many parents conformed to the practices of HCPs involved in childcare. This worked well
23 when the care provided matched the parents' preferences. However, various interactions
24 between parents and HCPs seemed to revolve around misalignment and parents experienced
25 gaps between what they thought was important in childcare and what healthcare had to offer.
26 Parents' struggles to fill gaps contributed to their burden of care and distressed parents.
27 Findings presented in "dialogical care" demonstrated that parental distress decreased when
28 parents could communicate about what they considered important given quality of life for
29 child and family and its implications for care delivery. Parents' preferred level of autonomy
30 was then an outcome of continuous dialogue rather than an evolution born of struggle.
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45

46 Deliberation on scenarios of sharing medical care can be related to the work of shared
47 decision making. Findings indicated that deliberation with parents should not only address
48 options in treatment and medical interventions but should also encompass repeatedly
49 discussing scenarios of sharing care with providers. The idea of shared decision making
50 underlines that respect for parents' preferences and priorities, and commitment to parents'
51 autonomy must be integral aspects of care and communications (Elwyn, 2021; Katz, 1994).
52 Findings underlined that parents and HCPs have complementary knowledge when discussing
53
54
55
56
57
58
59
60

1
2
3 options in sharing care. HCPs have expert and specialised knowledge from practicing
4 professionally. Parents often have an intimate bond with the child and therefore develop tacit
5 knowledge with regard to childcare (Kruithof et al., 2020; Polanyi, 2009). Parents' embodied
6 experience includes the day-to-day integration of medical care in broader family life. Our
7 findings accentuated that parents and providers should exchange expertise and should actively
8 listen and talk to arrive at decisions about sharing day-to-day medical care over extended
9 periods of time.
10
11
12
13
14
15
16
17
18
19

20 Findings revealed that when parents reported equal cooperation with providers, they
21 also reported that HCPs engaged in childcare invested in building a relationship with families.
22 Because of that, the parents perceived the providers' presence in childcare as positive and
23 supportive. This pointed to another implication of commitment to the parents' autonomy. As
24 reported in studies on the work of shared decision making, HCPs must be attentive to parents
25 emotional and relational needs (Elwyn, 2021; Gulbrandsen et al., 2016). Through
26 attentiveness for the parents' needs, providers acknowledge parents' existentially challenging
27 position of dependence and vulnerability in parenting their child. Attentiveness should include
28 consideration for parents' feelings about performing medical procedures in caring for the
29 child. Parents' context and distress may compromise agency. When HCPs explore the
30 parents' needs and respond effectively, they thereby contribute to parents' autonomous
31 capacity to make choices that protect the wellbeing of child and family (Gulbrandsen et al.,
32 2016; Mackenzie et al., 2014). In our study, parents also related (restored) autonomy and
33 effective cooperation with HCPs to open and honest communication.
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51

52 This study may add to understanding of how healthcare services and individual HCPs
53 can align their care to family needs. In line with previous studies, our study supports the
54 importance of making integrated medical and palliative care accessible for all families with
55 children with CMC and PIMD (Cassidy et al., 2023; Currie and Szabo, 2019; Vallianatos et
56
57
58
59
60

1
2
3 al., 2021). Consistent with other studies, our findings revealed that careful and early
4
5 discussions about the quality of the child's care and life are not always initiated (Kruithof et
6
7 al., 2022b; Loeffen et al., 2018). Our findings emphasised that such dialogues are crucial for
8
9 parents and that providers should embed these in their interactions with parents. Findings may
10
11 equally point to the need to facilitate providers to respond to family needs. This means HCPs
12
13 should work in organizations that foster relational work with families and where they can
14
15 operate according principles of family centred care, in which decisions are shared in principle.
16
17 Currently, this is often not the case. The parents' accounts supported the value of investing in
18
19 implementation of more democratic and holistic approaches such as family or person centred
20
21 care so that care can be tailored to the unique family or individual situation.
22
23
24
25

26 27 **Study strengths and limitations**

28
29
30 Given the limited number of studies with this particular focus, the in-depth and
31
32 exploratory approach of the study may be considered a strength. The study sample
33
34 represented parents of children with a variety of clinical diagnoses and conditions. All parents
35
36 cared for the child mainly in the family home and parents had different educational and
37
38 socioeconomic backgrounds. However, all parents who participated were caring or had cared
39
40 for the child with PIMD together with a life partner and most parents were of European
41
42 descent. A larger sample with more diversity in cultural backgrounds and family constructions
43
44 can contribute to a better understanding of parents' modes of dealing with medical care in
45
46 future research. More than a third of participants' children had died at the time of the
47
48 interview. This presents a source of potential bias because it meant that parents' experiences
49
50 dated back several years. We considered this critically but found no fundamental differences
51
52 between parents on this point. A strength of the study was that parents actively participated in
53
54 member reflections and thereby contributed to descriptions of findings (Tracy, 2020).
55
56
57
58
59

60 **Conclusions and implications for practice**

1
2
3 This study contributed to understanding of dimensions that affect the medical care
4 provided by parents who are caring for children with PIMD at home. From in-depth
5 interviews with Dutch parents, we concluded that an interplay of five dimensions established
6 experiences of parents with (sharing) medical care, namely (A) “fragility”, (B) “planned
7 care”, (C) “irregularities”, (D) “the presence of providers”, and (E) “parents’ preferences”.
8
9 We proposed three scenarios that characterized interactions between parents and HCPs:
10 “dependent care”, “dialogical care”, and “autonomous care”. Within the changeable nature of
11 the child’s health and day-to-day family life, changes in dimensions led to changes in
12 scenarios of interaction with providers. Besides, different scenarios simultaneously existed for
13 different areas of medical care. Parents actively improved in dealing with medical care and
14 often evolved in their thinking about quality of care and life of the child and family.
15 Sometimes this was due to parents’ struggles with the care delivered by providers. It is
16 therefore important that HCPs engaged in childcare broaden the relational work of shared
17 decision making to concrete scenarios in sharing medical care with providers.
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35

36 The dimensions and scenarios proposed can be used for developing tools to
37 communicate and evaluate care with parents. This requires follow-up study with a broader
38 group of families with children with PIMD and medical complexities to examine whether and
39 how the dimensions and proposed scenarios also emerge in this larger group. Such tools may
40 help parents and providers become aware of patterns of interactions and empower parents to
41 make conscious choices in sharing care. Providers can then adapt their own role accordingly.
42 Communication with parents should not only address practicalities of providing care but also
43 the delicate issues of quality of care, life and death. Future research may address healthcare
44 design that encompasses opportunities for providers to work according more democratic and
45 holistic approaches. Research may also address the implications of parents performing
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

1
2
3 medical procedures protected by healthcare legislation. This future research may have a
4
5 participatory character so that parents' ideas can be used in future healthcare policies.
6
7
8
9
10
11
12
13
14
15
16
17
18
19
20
21
22
23
24
25
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

For Peer Review

References

Author (2019) [Title omitted for blind review]

Author (2021) [Title omitted for blind review]

Author (2022) [Title omitted for blind review]

Breitkreuz R, Wunderli L, Savage A and McConnell D (2014) Rethinking resilience in families of children with disabilities: A socioecological approach. *Community, Work & Family*, 17(3), 346-365. DOI: 10.1080/13668803.2014.893228

Cassidy L, Quirke MB, Alexander D, Greene J, Hill K, Connolly M and Brenner M (2023) Integrated care for children living with complex care needs: an evolutionary concept analysis. *European Journal of Pediatrics*, Feb 13, 1–16. DOI: 10.1007/s00431-023-04851-2

Chun Tie Y, Birks M and Francis K (2019) Grounded theory research: A design framework for novice researchers. *SAGE Open Medicine*, 7, 2050312118822927. DOI:10.1177/2050312118822927

Cohen E, Kuo DZ, Agrawal R, Berry JG, Bhagat SKM, Simon TD and Srivastava R (2011) Children with Medical Complexity: An Emerging Population for Clinical and Research Initiatives. *Pediatrics*, 127(3), 529-538. DOI: 10.1542/peds.2010-0910

Corbin JM and Strauss AL (2014) *Basics of Qualitative Research. Techniques and Procedures for Developing Grounded Theory*. SAGE.

1
2
3 Currie G and Szabo J (2019) 'It would be much easier if we were just quiet and disappeared':

4
5 Parents silenced in the experience of caring for children with rare diseases. *Health*

6
7 *Expectations*, 22(6), 1251-1259. DOI: 10.1111/hex.12958

8
9
10
11 Elwyn G (2021) Shared decision making: What is the work? *Patient Education and*

12
13 *Counseling*, 104(7), 1591-1595. DOI: 10.1016/j.pec.2020.11.032

14
15
16
17 Gulbrandsen P, Clayman ML, Beach MC, Han PK, Boss EF, Ofstad EH and Elwyn G (2016)

18
19 Shared decision-making as an existential journey: Aiming for restored autonomous

20
21 capacity. *Patient Education and Counseling*, 99(9), 1505-1510. DOI:

22
23 10.1016/j.pec.2016.07.014

24
25
26
27 ICPCN (2022) *International Children's Palliative Care Network (ICPCN)*.

28
29 <https://www.icpcn.org>

30
31
32
33 Jansen S (2015) *Shared responsibility: a load off your mind: Collaboration with parents in*

34
35 *the support of children with Profound Intellectual and Multiple Disabilities*. University

36
37 of Groningen. DOI: 10.1016/j.ridd.2014.03.044

38
39
40
41 Jansen S, Van der Putten A, Post W and Vlaskamp C (2018) Do they agree? How parents and

42
43 professionals perceive the support provided to persons with profound intellectual and

44
45 multiple disabilities. *Journal of Intellectual & Developmental Disability*, 43(4), 441-452.

46
47 DOI: 10.3109/13668250.2017.1287885

48
49
50
51 Katz J (1994) Informed Consent – Must It Remain a Fairy Tale? *Journal of Contemporary*

52
53 *Health Law and Policy*, 10(1), 69-91.

54
55
56
57 Kirk S and Glendinning C (2002) Supporting 'expert' parents—professional support and

58
59 families caring for a child with complex health care needs in the community.

1
2
3 *International Journal of Nursing Studies*, 39(6), 625-635. DOI: 10.1016/S0020-
4
5 7489(01)00069-4
6
7

8
9 Kirk S, Glendinning C and Callery P (2005) Parent or nurse? The experience of being the
10
11 parent of a technology-dependent child. *Journal of Advanced Nursing*, 51(5), 456-464.
12
13 DOI: 10.1111/j.1365-2648.2005.03522.x
14
15

16
17 Kruithof K, Olsman E, Nieuwenhuijse A and Willems D (2022a) "I hope I'll outlive him": A
18
19 qualitative study of parents' concerns about being outlived by their child with profound
20
21 intellectual and multiple disabilities. *Journal of Intellectual & Developmental Disability*,
22
23 47(2), 107-117. DOI: 10.3109/13668250.2021.1920377
24
25

26
27 Kruithof K, Olsman E, Nieuwenhuijse A and Willems D (2022b) Parents' views on medical
28
29 decisions related to life and death for their ageing child with profound intellectual and
30
31 multiple disabilities: A qualitative study. *Research in Developmental Disabilities*, 121,
32
33 104154. DOI: 10.1016/j.ridd.2021.104154
34
35

36
37 Kruithof K, Willems D, Van Etten-Jamaludin F and Olsman E (2020) Parents' knowledge of
38
39 their child with profound intellectual and multiple disabilities: An interpretative
40
41 synthesis. *Journal of Applied Research in Intellectual Disabilities*, 33(6), 1141-1150.
42
43 DOI: 10.1111/jar.12740
44
45

46
47
48 Loeffen EAH, Tissing WJE, Schuiling-Otten MA, De Kruiff CC, Kremer LCM, Verhagen
49
50 AAE and Pediatric Palliative Care - Individualized Care Plan Working Group (2018)
51
52 Individualised advance care planning in children with life-limiting conditions. *Archives*
53
54 *of Disease in Childhood*, 103(5), 480-485. DOI: 10.1136/archdischild-2017-312731
55
56
57
58
59
60

- 1
2
3 Luijkx, J, Van der Putten AAJ and Vlaskamp C (2017) Time use of parents raising children
4
5 with severe or profound intellectual and multiple disabilities. *Child: Care, Health and*
6
7 *Development*, 43(4), 518-526. DOI: 10.1111/cch.12446
8
9
10
11 Luijkx J, Van der Putten AJ and Vlaskamp C (2019) A valuable burden? The impact of
12
13 children with profound intellectual and multiple disabilities on family life. *Journal of*
14
15 *Intellectual & Developmental Disability*, 44(2), 184-189. DOI:
16
17 10.3109/13668250.2017.1326588
18
19
20
21 Mackenzie C, Rogers W and Dodds S (2014) *Vulnerability: New Essays in Ethics and*
22
23 *Feminist Philosophy*. Oxford University Press.
24
25
26
27 McDonald J, McKinlay E, Keeling S and Levack W (2016) Becoming an expert carer: the
28
29 process of family carers learning to manage technical health procedures at home. *Journal*
30
31 *of Advanced Nursing*, 72(9), 2173-2184. DOI: 10.1111/jan.12984
32
33
34
35 Nakken H and Vlaskamp C (2007) A Need for a Taxonomy for Profound Intellectual and
36
37 Multiple Disabilities. *Journal of Policy and Practice in Intellectual Disabilities*, 4(2), 83-
38
39 87. DOI: 10.1111/j.1741-1130.2007.00104.x
40
41
42
43 Olsman E, Nieuwenhuijse AM and Willems DL (2022) ‘My son has lost his dignity’: dignity
44
45 of persons with profound intellectual and multiple disabilities. *Disability & Society*, 1-18.
46
47 DOI: 10.1080/09687599.2022.2045190
48
49
50
51 Petry K and Maes B (2006) Identifying expressions of pleasure and displeasure by persons
52
53 with profound and multiple disabilities. *Journal of Intellectual & Developmental*
54
55 *Disability*, 31(1), 28-38. DOI: 10.1080/13668250500488678
56
57
58
59 Polanyi M (2009) *The Tacit Dimension*. The University Of Chicago Press.
60

1
2
3 Smith J, Swallow V and Coyne I (2015) Involving parents in managing their child's long-term
4 condition-a concept synthesis of family-centered care and partnership-in-care. *Journal of*
5
6 *Pediatric Nursing*, 30(1), 143-159. DOI: 10.1016/j.pedn.2014.10.014
7
8
9

10
11 Spiers G and Beresford B (2017) "It goes against the grain": A qualitative study of the
12 experiences of parents' administering distressing health-care procedures for their child at
13 home. *Health Expectations*, 20(5), 920-928. DOI: 10.1111/hex.12532
14
15
16
17

18
19 Tracy SJ (2020) *Qualitative Research Methods. Collecting Evidence, Crafting Analysis,*
20
21 *Communicating Impact.* Wiley Blackwell.
22
23

24
25 Vallianatos S, Huizinga CSM, Schuiling-Otten MA, Schouten-Van Meeteren AYN, Kremer
26 LCM and Verhagen AAE (2021) Development of the Dutch Structure for Integrated
27 Children's Palliative Care. *Children*, 8(9), 741. DOI: 10.3390/children8090741
28
29
30
31

32
33 Van der Putten A, Vlaskamp C, Luijkx J and Poppes P (2017) *Position Paper Research*
34 *Centre on Profound and Multiple Disabilities.* University of Groningen: Research Centre
35
36 PMD.
37
38
39

40
41 Whiting M (2014) Children with disability and complex health needs: the impact on family
42 life. *Nursing Children and Young People*, 26(3), 26-30. DOI:
43
44 10.7748/ncyp2014.04.26.3.26.e388
45
46
47

48
49 Whiting M (2019) Caring for children – '24-7': The experience of WellChild Nurses and the
50 families for whom they are providing care and support. *Journal of Child Health Care*,
51
52 23(1), 35-44. DOI: 10.1177/1367493518777149
53
54
55
56
57
58
59
60

1
2
3 Wij zien je Wel (2022) *Werkgroep Wij zien je Wel. Passende zorg en ondersteuning van*
4
5 *mensen met ZEVMB en hun gezinnen. Focus op gezinsleven met #ZEVMB.*
6
7 <https://www.wijzienjewel.nl/>
8
9

10
11 Woodgate RL, Edwards M, Ripat JD, Borton B and Rempel G (2015) Intense parenting: a
12
13 qualitative study detailing the experiences of parenting children with complex care needs.
14
15 *BMC Pediatrics*, 15, 197-5. DOI: 10.1186/s12887-015-0514-5
16
17

18
19 Zaal-Schuller I, Willems DL, Ewals FVPM, Van Goudoever JB and De Vos MA (2016) How
20
21 parents and physicians experience end-of-life decision-making for children with
22
23 profound intellectual and multiple disabilities. *Research in Developmental Disabilities*,
24
25 59, 283-293. DOI: 10.1016/j.ridd.2016.09.012
26
27
28
29
30
31
32
33
34
35
36
37
38
39
40
41
42
43
44
45
46
47
48
49
50
51
52
53
54
55
56
57
58
59
60

Table 1: Sociodemographic characteristics of the participants and their children (N = 25; children: N = 22)(Author, 2022)

Sociodemographic variable	% (N =)
Age range (years)	
30 – 40	24 (6)
41 – 50	28 (7)
51 – 60	24 (6)
61 – 70	16 (4)
Missing data	8 (2)
Gender	
Female	76 (19)
Male	24 (6)
Relationship to children	
Biological parent	92 (23)
Foster parent	8 (2)
Partner status	
Living together	100 (25)
Living alone (separated or widowed)	–
Employment	
Fulltime	4 (1)
Parttime	44 (11)
Retired	8 (2)
Self-employed	24 (6)
Not employed (fulltime caretaker)	20 (5)
Family size (including the parents)	
A family of 3	12 (3)
A family of 4	24 (6)
A family of 5	44 (11)
A family of 6	20 (5)
Age range living children (N = 14 out of 22; median age 15 years)	
0 – 10	22 (5)
11 – 20	36 (8)
21 – 30	5 (1)
Age range deceased children (N = 8 out of 22; median age at passing 11 years)	
0 – 10	18 (4)
11 – 20	5 (1)
21 – 30	9 (2)
41 – 50	5 (1)
The child's main clinical diagnosis (N = 22)	
Brain anomalies	9 (2)

Known genetic disorder (e.g. Cornelia de Lange syndrome, Moebius syndrome, Rett syndrome, Sturge-Weber syndrome, tuberous sclerosis, Wolf-Hirschhorn syndrome)	50 (11)
Metabolic disorder (e.g. metachromatic leukodystrophy, Niemann-Pick disease type C)	18 (4)
Unknown genetic disorder	23 (5)

Table 2: Dimensions in the provision of medical care and scenarios of interactions with characteristic quotes

<i>Dimensions</i>	A	B	C	D	E	Parents' descriptions of interactions with HCPs within scenarios
Scenarios						
Dependent care	+	+/-	+	+	-	<p>We had a lot of people here, at home. They were always nice. They really supported us. (T10)</p> <p>Well, day care is very important. That is number one, actually. It is important that there is a place where he can go every day and where he is well cared for, and where he gets his therapies. That really is the most important thing. (T15)</p>
Dialogical care	+/-	+	+/-	+	+	<p>We know much more about her and can make better estimates than they do. Even the paediatrician who has been involved with her for almost eighteen years or the neurologist who has also been involved for years. When we ask a question, they almost always ask us back what is different and what we think is going on. They have no expertise on her. We are very much on equal footing there. (T01)</p> <p>When they started to talk about all those things with us, for example, how are the other children doing, how are you doing. Then we finally felt relaxation and relief (...) Then we really started to cooperate, with the medical world, so to speak. (...) Something that I, that we, had longed for years, but</p>

which had not yet been sufficiently achieved. (T19)

Autonomous care

+/- + +/- - +

The healthcare system only works provider oriented and not demand oriented. They did not listen to what we asked and what we wanted. (...) He came back from that day care with a high fever every time and that is why we did not want that anymore. But there was no other day care. (...) That meant he came home, and I took care of him at home. (T03)

I am Michael's voice. I see what is happening to him and I have to fight for him. When we came to the intensive care unit the intensivists immediately asked if we wanted Michael to be treated. Yes, why would we not want that? The child enjoys life. At a certain point we actually hung pictures of him at the wall showing him bursting with joy. I think as a parent, if you do not stand up for your child, nobody will. Well, of course there can be situations where we say, we cannot do this anymore, he should not be treated anymore. But at that point, we did not feel like, this is over. (T17)