

# Parents' perceptions of health care networks of children with inherited metabolic diseases (IMDs): a mixed methods study

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## Rationale and Objective

- Children with IMDs and their families face challenges in receiving high quality, family-centred health services.<sup>1</sup>
- To inform the development of interventions to improve their health care experiences, we need to better understand care needs from families' perspectives.

**Objective:** We sought to understand parents'/caregivers' (henceforth "parents") perceptions of the health care networks of children with inherited metabolic diseases (IMD).

## Methods

**Design:** an explanatory sequential mixed-methods study embedded in a prospective cohort study

**Participants:** parents of children ≤12 years with an IMD

**Recruitment:** 11 participating Canadian metabolic clinics

**Data collection (Nov 2020-Apr 2022):**

- Parent participants created a 'care map' depicting their perceptions of their child's network of providers and connections between providers (see Results, Fig 1).
- Parents also completed a tailored survey about care coordination and relational continuity for "key" providers (those parents perceived as most important to the child's care).
- A subset of parents participated in a semi-structured interview about their care map.

**Analysis:**

- From parent-drawn care maps, we used egocentric social network analysis<sup>2</sup> to describe, for each child's care network:
  - Size: number of providers included on the care map
  - Density: ratio of parents' perceived connections between pairs of providers / number of possible connections (0-1)
  - Centralization: % of pairwise connections that centre around one provider (the most connected provider) (0-100)
- We also reported social network analysis metrics at the level of provider "type" (e.g., dietitian, metabolic doctor, neurologist)
  - Degree centrality: number of connections a provider has
  - Share: proportion of all connections involving a provider
- We used descriptive statistics to analyze parents' ratings of care coordination and relational continuity for key providers.
- We analyzed interviews thematically and integrated quantitative and qualitative results narratively.

## References

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## Results

### 1. Characteristics of participants, children, and children's care networks

Table 1. Participant, child, and household characteristics (n=60)

	n (%)	n (%)	
Participant relation to child: mother	53 (88.3)	Child diagnosis: Amino acid disorder	15 (25.0)
Child age, years: 0-3	26 (43.3)	Urea cycle disorder	6 (10.0)
4-6	16 (26.7)	Organic acid disorder	3 (5.0)
7-9	14 (23.3)	Fatty acid oxidation disorder	19 (31.7)
10-12	4 (6.7)	Other IMD	17 (28.3)
Child sex assigned at birth: female	32 (53.3)	Travel time to metabolic clinic: 1 hour or less	35 (58.3)
		>1-3 hours	12 (20.0)
		>3 hours	13 (21.7)

- 60 parents provided care maps; 10 of the 60 parents participated in interviews.
- Children's care networks were variable in size (Fig. 1) with a median of 14.5 providers.
- Of 89 provider types, the most common were: metabolic doctors (n=55 children), lab technicians (n=55), dietitians (n=45).

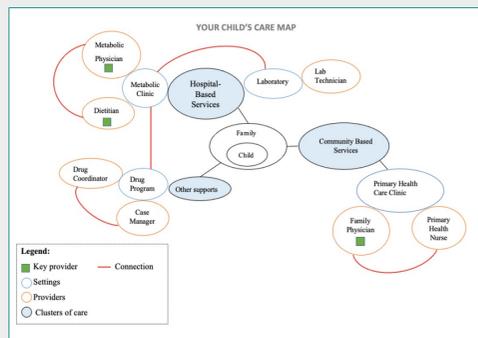


Figure 1a. Example #1 of parent-drawn care maps.

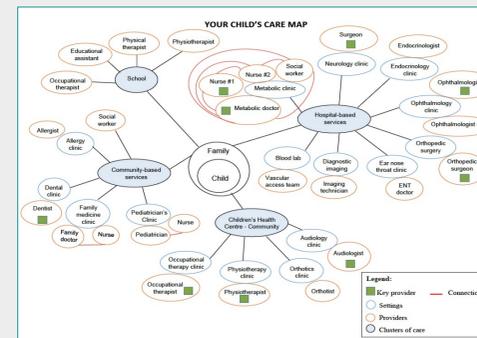


Figure 1b. Example #2 of parent-drawn care maps.

- Parents we interviewed often described their role as informal 'managers', responsible for establishing, maintaining, and coordinating their child's care.
- This was sometimes experienced as overwhelming.

"And then, we've had a huge turnover in our [community services] team regarding people that are in those particular positions, and that's difficult because of the rare disease aspect of it...So, I am finding myself doing a lot of education right now, and I am managing a lot of the [community services] on my own again right now." (Participant #8, Mother)

Network connectedness:

- Median density of 0.08 (only 8% of possible pairwise connections were perceived by parents)
- Median centralization of 0.23 (the most connected provider was involved in approximately 23% of the pairwise connections).

### 3. Care coordination and providers as part of a dynamic network

Among common key providers:

- Degree centrality (# connections to other providers):
  - Metabolic doctors, dietitians, nurses: median of 3 each.
  - Pediatricians: median of 1.
  - Family doctors: median of 1.

- Most participants rated key metabolic doctors or dietitians as coordinating care fairly or very well (85% & 91%, respectively) (Fig 3).

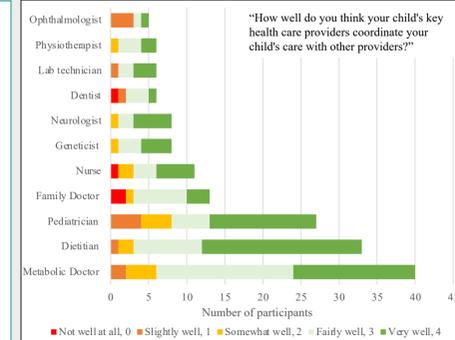


Figure 3. Key health care providers identified by at least 5 participants, and their care coordination ratings.

- Parents we interviewed viewed care coordination as: providers actively working as a team; referrals or consultation; or sharing information.
- Parents generally did not perceive that providers were well connected, particularly non-metabolic specialists, including primary care providers.
- When care coordination was perceived as inadequate, parents described being the 'middle person', relaying information between providers.
- Several parents described identifying a "go-to" provider to help coordinate care: a provider who trusted the family and could directly respond to the child's needs or facilitate access to other providers and services.
 

"I would phone or, she would phone me, and if I had a question, she would find it out for me. If she didn't know she'd phone the doctor that did, or she would have someone contact me, it seems. They have been very helpful that way. Or, when I have needed the appointment, she was who I was letting know I was coming that day, so then she made sure that everybody else kind of had got a hold of me to schedule an appointment." (Participant #5, Mother)

### 2. Key providers and relational continuity

- Parents identified 55 types of "key" providers: these were up to 10 care providers per child, who parents designated as most important to their children's care.
- Metabolic doctors (n=40), dietitians (n=33 children), and pediatricians (n=27) were most commonly named as key providers (Fig. 2).
  - Parents we interviewed explained that they designated providers as "key" because of their expertise, central role, availability, and relationship with the family.
- A majority of participants (80%) reported having at least one health care provider in the network who knew their child and family fairly well or very well (Fig. 2).
  - Parents we interviewed valued this relational continuity, which included the concepts of emotional connection as well as familiarity with the child and family.

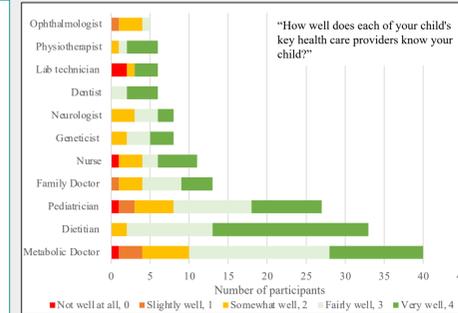


Figure 2. Key health care providers identified by at least 5 participants, and their relational continuity ratings.

- Parents valued providers who demonstrated that they trusted parents to know the child's needs.

"The outcome of that phone call was that I knew [son] best, and if I felt like, over a duration of time, I'm not seeing any of those symptoms subside, to give him a call, that he would call in the increase." (Participant #1, Mother)

## Discussion

**Summary:** Children with IMDs had variable but frequently large care networks that were sparsely coordinated. Parents assumed responsibility for many aspects of care on an ongoing basis and this was often overwhelming. Parents valued providers who trusted them and who had a lasting relationship with the family.

**Strengths:** A mixed-methods design allowed a nuanced understanding of health care networks for children with IMDs.

**Limitation:** We studied parental perceptions about children's health care, which might be different from provider perceptions.

**Conclusion:** Strategies that recognize families' expertise while supporting them with improved coordination are needed. Metabolic specialist providers may be well-positioned to support such strategies in a co-management role with primary care.<sup>3</sup>