Parent-Reported Experiences with Health Care for Children with Inherited Metabolic Diseases in Canada: Results from a Multi-Centre Survey

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Study Rationale & Goal

Rationale

- Patient experience with care is a key pillar of high-quality health care systems and is also associated with clinical and safety outcomes[1,2]
- Patient experience is founded on principles of patient-centred care:[3-5]
  - Accessible services, respect and support, clear communication, coordination and continuity of care, involvement of family/friends
- In pediatrics, family-centred care further emphasizes developmental needs and the central role of family[6,7]
  - Strategies to promote family-centred care for children with chronic conditions: co-developed care plans, a 'Medical Home', a designated key provider, provider collaboration, strong family involvement[6-10]
- Our previous qualitative research identified that care for children with inherited metabolic diseases (IMDs) does not always adhere to these principles of family-centered care. Parents reported:[11]
  - Problematic interactions with services and providers outside of the metabolic clinic that were unfamiliar with the child's disease; and
  - Poor care coordination for children requiring health services from multiple providers and systems.

Goal of this study:

- To describe experiences with health care for children with IMD in Canada, from parents' perspectives, based on principles of family-centred care.

Methods & Respondents to Date

Sample

- Parents/guardians of children (<12 years of age) with an IMD enrolled in the Canadian Inherited Metabolic Diseases Research Network (CIMDRN) cohort across 13 participating treatment centres (www.cimdrn.ca) are eligible to participate in a cross-sectional survey (Fig 1)
- Survey currently underway; implemented in participating centres as they receive research ethics board approval

Study Design and Data Collection:

- Eligible families are contacted by mail up to 4 times[12] to invite their participation
- Questionnaire completed by the person in the family most knowledgeable about the child's care and returned by mail

Questionnaire:

- Developed based on a literature review and qualitative study[11]; pilot-tested in 6 parents of children with IMD using cognitive interviews conducted by telephone
- Measures of Process of Care (MPOC-20)[13] instrument to assess family-centred care in the metabolic clinic
- Questions adapted from the Family Experiences with Care Coordination (FECC)[14] instrument to evaluate care coordination, community service needs, and out-of-pocket expenses
- We developed study-specific questions about satisfaction with care in common care settings for IMD and factors considered important in ratings of satisfaction with care

Analysis: Descriptive analysis conducted with data received to date; preliminary findings presented

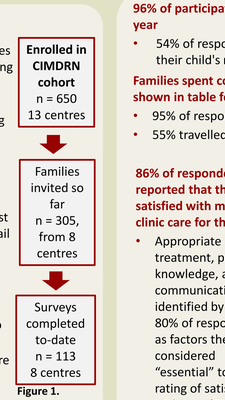

Figure 1. Participant flow chart to date (survey ongoing)

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Results: 1. Participant Characteristics

- Most survey respondents were the biological mothers of children with IMD (Table 1)
- 73% of respondents were aged 35 years or older, 60% worked full or part-time, and 74% had completed some post-secondary education
- 64% were born in Canada and 61% lived in a large city
- A minority of respondents reported having more than one child diagnosed with an IMD (13%); in these families, one child was the "index child" and the respondent was asked to consider this child when reporting experiences with care

Table 1. Respondent characteristics

Respondent (n=113)	n (%)
Female	91 (81%)
Biological parent	106 (94%)
Age	
25-34 years	30 (27%)
35-44 years	59 (52%)
45 years or older	24 (21%)
Working Part/Full-Time	68 (60%)
Education	
High school or less	28 (26%)
Vocational/technical	12 (11%)
College/university	52 (47%)
Graduate School	18 (16%)
Born in Canada	72 (64%)
Community Size	
Large city	67 (61%)
Medium city	24 (22%)
Small community	19 (17%)
> 1 child with IMD	15 (13%)

Table 2. Child characteristics

Index Child (n=113)	n (%)
Female	62 (55%)
Birth Year	
2006-2007	25 (22%)
2008-2009	23 (20%)
2010-2011	19 (17%)
2012-2013	21 (19%)
2014-2015	25 (22%)
IMD (specific disease)	
MCAD deficiency	28 (25%)
Other FAO disorder	12 (11%)
PAH deficiency	21 (19%)
Other AA disorder	8 (7%)
OA disorder	9 (8%)
Other	35 (31%)

AA = amino acid, FAO = fatty acid oxidation, MCAD=medium-chain Acyl-CoA dehydrogenase; OA = organic acid; PAH=phenylalanine hydroxylase

- 55% of index children were female; birth years were from 2006 to 2015 (Table 2)
- MCAD deficiency and PAH deficiency were the most common diagnoses

2. Metabolic Clinic Care

96% of participating children received care at a metabolic clinic in the past year

- 54% of respondents identified a metabolic clinic physician or dietitian as their child's main health care provider (Table 3)

Table 3. Child's main care provider[14]

	n (%)
Physician at the metabolic clinic	43 (38%)
Dietitian at the metabolic clinic	18 (16%)
Family physician	32 (29%)
Community pediatrician	11 (10%)
Other	8 (7%)

Families spent considerable time driving to the metabolic clinic (results not shown in table form):

- 95% of respondents reported that they typically drove to the clinic
- 55% travelled 1-2 hours or longer to reach the clinic

86% of respondents reported that they were satisfied with metabolic clinic care for their child

- Appropriate treatment, provider knowledge, and communication were identified by nearly 80% of respondents as factors they considered "essential" to their rating of satisfaction with care (Figure 2)

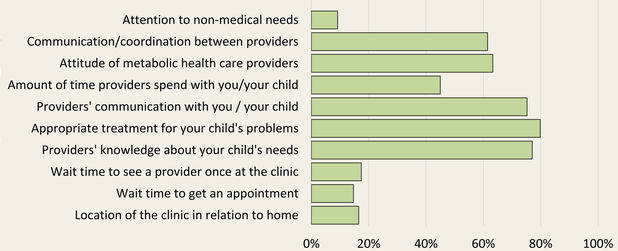

Figure 2. Proportion considering factors "essential" to level of satisfaction with metabolic clinic care (n=109)

Respondents generally reported that their child's care needs were met at the metabolic clinic (MPOC[13] results, Table 4):

- For most MPOC* subscales, respondents reported that needs were met to a fairly great (score of 5) or great (score of 6) extent
- For "Providing General Information", needs were perceived as met to a small (score of 3) or moderate (score of 4) extent
- Respondents who stated they were not satisfied with care in the metabolic clinic reported needs being met to a lesser extent

Table 4. Extent to which needs were met within each domain of family-centred care, Measuring Processes of Care (MPOC)[13]

	Enabling & Partnership		Providing General Information		Providing Specific Information		Coordinated/Comprehensive Care		Respectful & Supportive Care	
	N	Med (IQR)	N	Med (IQR)	N	Med (IQR)	N	Med (IQR)	N	Med (IQR)
Overall	98	5.67 (2.50)	91	3.80 (3.20)	97	5.00 (2.67)	104	6.00 (1.75)	106	6.20 (1.40)
Non-Satisfied	14	4.17 (1.34)	14	2.70 (2.00)	15	4.00 (1.67)	15	4.00 (1.50)	15	4.60 (1.55)
Satisfied	84	6.00 (2.00)	77	4.40 (2.80)	82	5.33 (2.67)	89	6.25 (1.50)	91	6.20 (1.40)
p-value**		<.001		.01		.003		<.001		<.001

*MPOC subscale scores are calculated as a participant's average rating across the items within a subscale, from 1-7 (higher scores reflect needs being met to a greater extent); **Normally approximated p-values for Mann-Whitney U non-parametric test of the difference in scores for those who were non-satisfied vs satisfied with care; IQR = interquartile range, Med = median.

3. Emergency Department & Hospital Care

33% of children were reported to have visited the emergency department (ED) in the past year, while 19% had experienced an overnight hospital stay (Table 5)

73% of parents of children who received ED care reported being satisfied with the ED care their child received

- Appropriate treatment was most commonly rated as essential to satisfaction with ED care (>80%, Figure 3)
- Communication and health care provider attitude were reported as essential to satisfaction by nearly 80% of respondents

Table 5. Child's use of health services, past year

Receipt/frequency of use	n (%)
Emergency Department (n=113)	
Not at all	76 (67%)
1-2 times	25 (22%)
At least 3 times	12 (11%)
Overnight hospital stay (n=113)	
Not at all	91 (81%)
1 time	14 (12%)
At least 2 times	8 (7%)

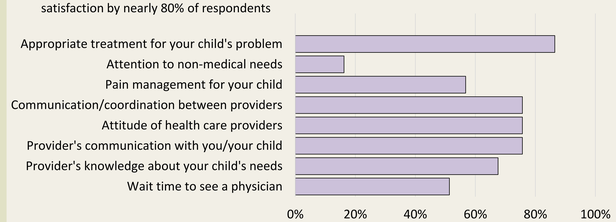

Figure 3. Proportion considering factors to be "essential" to level of satisfaction with ED care (n=37)

4. Blood Laboratory & Pharmacy Care

74% of respondents were satisfied with care provided by the blood laboratory

- 89% of index children had visited the blood lab in the past year; 16% visited 6 or more times
- The only factors rated as essential to care satisfaction at the blood laboratory by a majority of parents were attitude of care providers and communication (Figure 4)

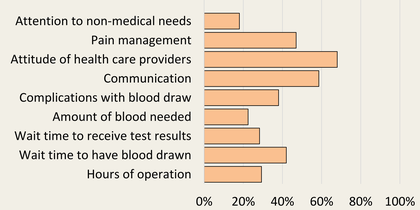

Figure 4. Proportion considering factors to be "essential" to level of satisfaction with care provided by the blood laboratory (n=100)

79% of respondents were satisfied with the services provided by the pharmacy

- 75% of index children had used the pharmacy in the past year
- Receiving the correct products and dose, availability of products, and timely access were considered essential to satisfaction by >70% of parents (Figure 5)

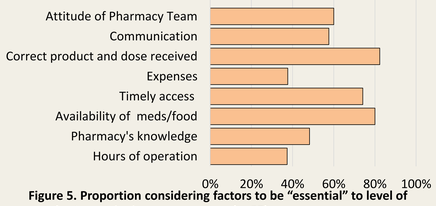

Figure 5. Proportion considering factors to be "essential" to level of satisfaction with pharmacy services (n=85)

5. Care Coordination & Out-of-pocket Expenses

Care coordination (Table 6):

- Of the 57% of respondents whose children used multiple health services, 56% received help in coordinating care
- For this group, the main provider's office was involved in coordination for 68%
- Main providers infrequently provided written summaries and shared care plans

Community Services (Table 6):

- 18% of parents reported a need for community services like home health care and community-based intervention programs
- Half of those who required community services reported barriers to access (e.g., financial barriers, wait times)

Out-of-pocket expenses (Table 7):

- 52% of respondents reported out-of-pocket expenses related to their child's care
- The most common out-of-pocket expenses were medications and medical foods or supplements

Table 6. Care coordination & community services, past year[14]

Service queried and denominator*	n (%)
Child used multiple health services, past year (n=111)	63 (57%)
Among those who used multiple services, family received support in coordinating those services (n=62)	35 (56%)
Among those who received care coordination support, source of this support (n=35):	
Main provider's office	12 (34%)
Outside main provider's office	11 (32%)
Both within and outside main provider's office	12 (34%)
The main provider…	
…provided written summary after clinic visits (n=111)	30 (27%)
…created a shared care plan (n=112)	15 (13%)
…created an emergency care plan (n=111)	54 (49%)
Family needed or used community services (n=111)	20 (18%)
Of those who needed community services, had trouble accessing needed services (n = 20)	10 (50%)

*Care coordination questions were adapted from the Family Experiences with Care Coordination (FECC) instrument[14]

Table 7. Care expenses paid out-of-pocket by family, past year

	n (%)
Household paid from personal or family income to care for index child (n = 113)	59 (52%)
Of those who paid, items paid for (n=59):	
Community-based early intervention programs	5 (8%)
Medical equipment	7 (12%)
Special transportation accommodations	0 (0%)
Medications	25 (42%)
Medical foods or supplements	22 (37%)
Other (vitamins, oils, travel, psychosocial services)	21 (36%)

Discussion

The role of the metabolic clinic as a potential 'medical home' for pediatric IMD

- 54% of parents identified a physician or dietitian at the metabolic clinic as their child's main provider (Table 3), suggesting that some parents may perceive the metabolic clinic as the child's medical home.
- However, while 86% of parents were satisfied with care at the metabolic clinic and needs for family-centred care were perceived as being met in most areas, scores for the "Providing General Information" subscale of the MPOC were relatively low (Table 4), indicating unmet needs regarding connections to community services and other resources. Further, a minority of respondents indicated that the main provider developed written visit summaries (27%) or shared care plans (13%; Table 6).
- Among those children requiring multiple health services (57%), 56% received care coordination support, most often (68%) from the main provider's office (Table 6).
- These findings indicate that parents saw the metabolic clinic as central to their child's care and generally perceived care there as family-centred. There is potential for the metabolic clinic to play a greater role as a medical home with respect to care coordination and developing shared care plans. Access is a potentially important barrier: 55% of respondents travelled at least 1-2 hours to the clinic.

Other health care services, factors essential to satisfaction, and out-of-pocket expenses

- A majority of respondents (>70%) reported that they were satisfied with care within each service outside of the metabolic clinic, including ED care, the pharmacy, and the blood laboratory.
- Across all services, receiving appropriate care and high quality communication from providers were most often rated as essential to satisfaction, while timely access and attention to non-medical needs were less often rated as essential (Figures 2-5). More research is needed to determine the most important factors contributing to experiences with care at the blood laboratory (Figure 4).
- More than half of families reported out-of-pocket expenses related to their child's care (Table 7).

Conclusion and next steps

- Data collection and analysis is ongoing. We will further investigate variation in care experiences across Canadian centres, and gaps in care reported by parents whose reported experiences were less positive.
- Parents of children with IMD in Canada reported satisfaction with their children's care. They most valued appropriate treatment and high quality communication with providers. Gaps in family-centred care relate to care coordination and absence of shared care plans. Out-of-pocket expenses for families, and their impact, are potentially important concerns that require further investigation. Many parents reported driving long distances to care; expenses associated with this travel also warrant further study.

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