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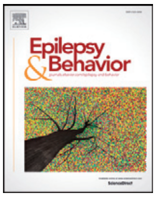

Influence of patient, caregiver, and family factors on unmet healthcare needs in children with drug-resistant epilepsy undergoing epilepsy surgery evaluation

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ABSTRACT

Objective: The aim of this study was to assess whether patient, caregiver, and family factors correlate with unmet healthcare needs in children with drug-resistant epilepsy (DRE) who were evaluated for epilepsy surgery.
Methods: Caregivers of children with DRE completed questions on unmet healthcare needs, including physical and mental needs. Patient (demographic, seizure severity, and comorbidities), caregiver (sociodemographics, depression, and anxiety), and family (family functioning, resources, and demands) factors were evaluated. A series of bivariable regression was conducted to assess the associations with unmet needs (overall, physical, and mental needs), followed by multivariable regression.
Results: Of the 86 children with DRE, 32 (37.2%) reported having overall unmet healthcare needs, 21 (24.4%) have unmet physical, and 13 (15.1%) have unmet mental healthcare needs. Multivariable regression showed that the odds of overall unmet needs in children were higher in patients with higher depressive symptoms (odds ratio [OR] = 1.7, $p = 0.03$), and caregivers who were unemployed (OR = 3.8, $p = 0.008$). The odds of unmet physical needs in children were higher among patients with higher depressive symptoms (OR = 2.0, $p = 0.02$), caregivers who were older (OR = 1.1, $p = 0.02$), and caregivers who were unemployed (OR = 6.5, $p = 0.008$). There were no significant correlates between patient, caregiver, and family factors with unmet mental needs (all $p > 0.05$).
Interpretation: Children with DRE were at risk for not receiving needed healthcare. Patient factors such as patient depressive symptoms, and caregiver factors such as caregivers' age and employment status were associated with unmet needs. Identifying the predictors of unmet healthcare needs is a necessary step in addressing unmet needs in this population with DRE.

1. Introduction

Patients with drug-resistant epilepsy (DRE) have persistent seizures despite treatment on two or more antiepileptic drugs (AEDs) [1]. Children with DRE require multidisciplinary healthcare services to manage not only their seizures but also comorbidities, such as behavioral and cognitive impairments [2–4], and emotional disorders [5,6]. Children with DRE consume more healthcare services [7], and have 2–4 times higher healthcare costs than those with well-controlled epilepsy [8–10]. Despite using more healthcare resources, patients with epilepsy (both adults and youths with epilepsy) were more likely to report that they did not receive the healthcare services that they needed compared to the general population; that is, they have unmet healthcare needs [11,12]. Unmet mental healthcare needs were also more frequent in patients with epilepsy compared with the general population [11,13], likely related to higher behavioral and emotional comorbidities in these patients.

Unmet healthcare needs have been considered as an indicator of access problems [14,15] because of limited availability of healthcare services when and where they are required. However, unmet needs can also arise from individual accessibility problems, such as cost and transportation, which could in turn be linked to household income [16]. A previous systematic review of unmet healthcare needs in patients with epilepsy has identified several reasons for unmet needs, including lack of availability of health services, long wait lists, accessibility issues, uncoordinated care, and lack of health information [12]. Andersen's model of health service utilization states that in addition to the healthcare system (or external environment), there are enabling,

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need, and predisposing factors that could affect health services utilization [17]. Predisposing factors include patient age and gender. Need factors include epilepsy severity, and comorbidities such as mood disorders and cognitive impairment. Enabling factors include caregiver and family factors such as household income, caregiver education, caregiver mental health, and family environment. There is a dearth of literature on the predisposing, need, or enabling factors that could influence unmet healthcare needs in children with DRE. The aim of the study was to assess the correlates of unmet healthcare needs in children with DRE who were evaluated for epilepsy surgery. The hypothesis was that there were predisposing factors such as patient demographics, need factors such as epilepsy severity and comorbidities, and enabling factors such as caregiver psychopathology and family environment that could influence unmet healthcare needs in children with DRE.

2. Methods

2.1. Participants

Children with DRE aged 6 to18 years were identified during their inpatient video-electroencephalography (EEG) evaluation for epilepsy surgery candidacy in the epilepsy monitoring unit at the Hospital for Sick Children. Inclusion criteria included children with focal DRE as assessed by clinical semiology and/or video-EEG, and who were evaluated for focal resective epilepsy surgery. Exclusion criteria included prior resective surgery, prior or planned nonresective surgery (e.g., corpus callosotomy), or vagal nerve stimulator placement as these are palliative procedures, neurometabolic disorders, neurodegenerative disorders, genetic epilepsy syndromes, and epileptic encephalopathies as these disorders frequently precluded epilepsy surgery treatment. For those who met inclusion criteria, the research coordinator approached the participants and their families, and obtained informed consent and assent from caregivers and children, respectively. The questionnaires were completed electronically on the Research Electronic Data Capture (REDCap) system, a secure web application [18], at home. A total of 110 children with DRE aged 6 to 18 years were approached for this study; 24 declined participation, and 86 were enrolled into the study. Some patients in this study were included in prior publications [19–21]. The study has the approval of the institutional research ethics board.

2.2. Study measures

2.2.1. Unmet healthcare needs

Data on overall unmet healthcare needs were obtained by asking the caregivers the question “During the past 12 months, was there ever a time when you felt that your child needed healthcare, but he/she did not receive it”. The caregivers who answered “yes” were subsequently asked “Thinking of the most recent time, what was the type of care that was needed?”; the possible answers included a) treatment of a physical health problem, and b) treatment of an emotional or mental health problem, indicating unmet physical and mental healthcare needs, respectively, and c) other unmet care needs (regular check-up, care of the injury, EEG studies, or other tests). The questions were based on those used in the national health surveys, the Canadian Community Health Survey (CCHS) and the National Survey of Children with Special Healthcare Needs (NS-CSHCN).

2.2.2. Patient, caregiver, and family factors

All questionnaires pertaining to patient, caregiver, and family factors were completed by patients and their caregivers, irrespective of caregivers' responses to the above questions on unmet needs.

Patient factors assessed included age, sex, age at seizure onset, seizure frequency, number of AEDs, intelligence quotient (IQ), anxiety, and depression. Intelligence quotient was assessed using the Wechsler Abbreviated Scale of Intelligence-II [22]. Child depressive symptoms were measured using the Revised Children's Anxiety and Depression Scale (RCADS - major depressive disorder subscale) [23] for participants aged 6 to 11 years, and the Quick Inventory of Depressive Symptomatology (QIDS) Scale [24,25] for participants aged 12 years and above. Child anxiety symptoms were evaluated using the RCADS (generalized anxiety disorder subscale) for participants aged 6 to 11 years, and the Generalized Anxiety Disorder 7-item scale (GAD-7) [26] for participants aged 12 years and above. Revised Children's Anxiety and Depression Scale score of 65 or higher reflects depression or anxiety above clinical threshold; QIDS score greater than 21 indicates severe depression; GAD-7 score of 14 or higher indicates severe anxiety. For both depressive and anxiety symptoms, the scores obtained from different instruments were standardized using z-scores, to place the instruments on the same scale for analysis.

Caregiver depressive symptoms were assessed using the QIDS, and caregiver anxiety symptoms were assessed using GAD-7. Sociodemographics assessed included caregiver age, sex, employment status, education, marital status, and socioeconomic status (SES: household income). Family environment was evaluated using three validated measures. Family relationships were measured using the Family Adaptability, Partnership, Growth, Affection, and Resolve (Family APGAR) scale, with higher scores indicating higher family function [27,28]. Family resources were quantified using the Family Inventory of Resources for Management (FIRM) [29]; two subscales (family mastery and health, and extended family social support), which have been found to be associated with adjustment to childhood epilepsy [30] were used. Financial well-being subscale was not included as part of the larger study. Higher FIRM scores indicate more family resources. Family demands were quantified using the Family Inventory of Life Events and Changes (FILE) [31], with higher score indicating more family demands.

See Supplementary Table 1 for further details on the above questionnaires.

3. Statistical analysis

Characteristics of patients and their caregivers were analyzed using descriptive statistics, with mean and standard deviation (SD) for continuous data, frequencies and percentages for categorical or binary data. The percentage of missing values ranged from 7.3% to 11.1% for child depressive and anxiety symptoms, respectively, and 22.1% for IQ. Missing data were addressed using multiple imputations with the Markov chain Monte Carlo (MCMC) method [32,33]. Little's missing completely at random (MCAR) test [34] showed that missing values were missing at random ($\chi^2 = 4.11$, $df = 3$, $p = 0.25$). Bivariable logistic regression analyses were employed to determine if patient (age, sex, age at seizure onset, seizure frequency, number of antiseizure medications, IQ, anxiety, and depression), caregiver (age, sex, income, education, marital status, employment status, anxiety, and depression), and family (FILE, FIRM, APGAR) factors predicted unmet healthcare needs (overall, physical, and mental) in children with DRE. Variables with a p-value of 0.05 or less in the bivariable regression were included in a multivariable logistic model. There was no evidence of multicollinearity among the factors as assessed by variance inflation factor ($VIF < 4$ for all variables). All statistical analyses were performed in the Statistical Package for the Social Sciences (SPSS) version 24.0 [35].

4. Results

4.1. Clinical and demographic characteristics

Eighty-six children with DRE were included in the study, with a mean age of 11.87 years ($SD = 3.37$; age range: 6 to 17.58 years) and 50 (58.1%) were males (range from 6 to 17.58 years). Table 1 summarizes the demographic characteristics. Thirty-eight (44.3%) children had daily or weekly seizures. Forty (46.5%) patients had symptoms of moderate to severe depression, and 25 (29.1%) had symptoms of

Table 1
Characteristics of children with drug-resistant epilepsy, caregiver, and family for total sample and those with unmet versus met healthcare needs.

	Total sample (n = 86)	Unmet needs (n = 32)	Met needs (n = 54)
Patient characteristics			
Age (year), mean (SD)	11.87 (3.37)	12.27 (3.90)	11.63 (3.03)
Sex, n (%)			
Male	50 (58.10)	20 (40.0)	30 (60.0)
Female	36 (41.90)	12 (33.30)	24 (66.70)
Mean age (year) at seizure onset (SD)	7.50 (4.08)	7.82 (3.82)	7.32 (4.24)
Seizure frequency, n (%)			
High (daily or weekly)	38 (44.20)	16 (50.0)	22 (57.90)
Low (monthly or year)	48 (55.80)	16 (50.0)	32 (66.70)
Number of AEDs, n (%)			
One	33 (38.40)	13 (39.40)	20 (60.60)
More than 1	53 (61.60)	19 (36.50)	33 (63.50)
Intelligence quotient, mean IQ (SD)	92.76 (18.50)	94.55 (18.43)	91.71 (18.63)
Depressive symptoms, mean raw score (SD)			
RCADS (n = 40)	51.97 (12.45)	–	–
QIDS	6.68 (3.97)	–	–
Mean z score (SD)*	0.01 (0.99)	0.29 (1.09)	−0.15 (0.92)
Anxiety symptoms, mean raw score (SD)			
RCADS (n = 40)	45 (11.34)	–	–
GAD-7	4.71 (3.76)	–	–
Mean z score (SD)	0.00 (0.99)	0.09 (0.92)	−0.05 (1.05)
Caregiver characteristics			
Age (years), mean (SD)	43.09 (7.40)	44.75 (8.74)	42.11 (6.37)
Sex, n (%)			
Male	16 (18.60)	5 (31.20)	11 (68.80)
Female	70 (81.40)	27 (38.60)	43 (61.40)
Marital status, n (%)			
Married	72 (83.70)	28 (38.90)	44 (61.10)
Other (single, widowed, divorced)	14 (16.30)	4 (28.60)	10 (71.40)
Employment status, n (%)*			
Employed	57 (66.30)	16 (28.10)	41 (71.90)
Not employed	29 (33.70)	16 (55.20)	13 (44.80)
Education, n (%)			
College/university	73 (84.90)	27 (37.0)	46 (63.0)
Less than college/university	13 (15.10)	5 (38.50)	8 (61.50)
Annual household income, n (%)			
Prefer not to say	7 (8.10)	2 (28.60)	5 (71.40)
$49,999 or less	13 (15.10)	6 (46.20)	7 (53.80)
$50,000–$99,999	30 (34.90)	11 (36.70)	19 (63.30)
$100,000–149,999	20 (23.30)	6 (30.0)	14 (70.0)
$150,000 or more	16 (18.60)	7 (43.80)	9 (56.30)
Anxiety symptoms, GAD-7 score (SD)	6.80 (6.17)	7.63 (6.50)	6.31 (5.97)
Depressive symptoms, QIDS score (SD)	6.93 (5.55)	6.97 (5.70)	6.91 (5.52)
Family characteristics			
FILE, mean (SD)	371.05 (241.7)	361.31 (255.43)	376.83 (235.56)
FIRM, mean (SD)	49.18 (11.79)	50.09 (11.20)	48.65 (12.20)
APGAR, mean (SD)	7.10 (2.41)	6.97 (2.66)	7.19 (2.28)

SD, standard deviation; AEDs, antiepileptic drugs; GAD-7, Generalized Anxiety Disorder instrument; QIDS, Quick Inventory of Depressive Symptomatology; FILE, Family Inventory of Life Events and Changes; FIRM, Family Inventory of Resources for Management; APGAR, Family Adaptability, Partnership, Growth, Affection, and Resolve.

* $p < 0.05$ using *t*-test for continuous data and chi-square for categorical data.

anxiety. Eighteen (20.9%) caregivers had symptoms of moderate to severe depression, and twenty-four (27.9%) had symptoms of severe anxiety. Thirty-two (37.20%) caregivers reported that their child had overall unmet healthcare needs (Table 1). Twenty-one patients (24.4%) had unmet physical healthcare needs, and 13 (15.1%) had unmet mental healthcare needs. Five patients (5.81%) had both physical and mental unmet healthcare needs, and seventeen patients (19.76%) also had other unmet healthcare needs.

4.2. Unmet overall healthcare needs

Table 2 summarizes the bivariable logistic regression models for the overall unmet healthcare needs. Patients with higher depressive symptoms had higher odds of overall unmet needs than those with lower depressive symptoms (odds ratio [OR] = 1.58, 95% confidence interval [CI]: 1.00, 2.51, $p = 0.05$). Caregivers who were unemployed had higher odds of reporting overall unmet needs for their children than those who were employed (OR = 3.15, 95% CI: 1.24, 8.01, $p = 0.02$). The other patient factors (such as patient age, sex, age at seizure onset, seizure frequency, number of AEDs, IQ, and patient anxiety), caregiver factors (such as caregiver age, sex, depression and anxiety, marital status, education, income), and family factors (such as family relationships, resources, and demands) did not predict overall unmet needs (all $p > 0.05$). Although there was no significant association between income and unmet overall healthcare needs, there was a gradient of unmet overall needs, highest among those who earn $49,999 or less, and lowest among those who earn $100,000–149,999, compared to those who earn $150,000 or more.

Patient depressive symptoms and caregiver employment status were included in the multivariable model (Table 3a). Patients with higher depressive symptoms had higher odds of reporting overall unmet needs in their children that those with lower depressive symptoms (OR = 1.71, 95% CI: 1.05, 2.81, $p = 0.03$), and caregivers who were unemployed had higher odds of reporting overall unmet needs in their children than those who were employed (OR = 3.82, 95% CI: 1.43, 10.20, $p = 0.008$). The model explained 17% of the variance.

Table 2
Bivariable logistic regression of patient, caregivers, and family factors predicting unmet overall, physical, and mental health needs in children with drug-resistant epilepsy.

Predictors	Unmet overall needs		Unmet physical needs		Unmet mental needs	
	OR (95% CI)	*p*	OR (95% CI)	*p*	OR (95% CI)	*p*
Patient characteristics						
Age	1.06 (0.92, 1.21)	0.39	1.09 (0.94, 1.27)	0.24	1.14 (0.94, 1.37)	0.18
Sex (ref: male)	0.75 (0.31, 1.84)	0.53	1.06 (0.39, 2.86)	0.92	0.57 (0.16, 2.02)	0.38
Age at seizure onset	1.03 (0.93, 1.15)	0.58	1.02 (0.90, 1.15)	0.81	1.03 (0.89, 1.20)	0.66
Seizure frequency (ref: high)	0.69 (0.29, 1.66)	0.40	0.83 (0.31, 2.23)	0.72	0.44 (0.13, 1.46)	0.18
Number of AEDs (ref: 1)	0.89 (0.36, 2.17)	0.79	0.80 (0.29, 2.18)	0.66	1.02 (0.30, 3.43)	0.98
IQ	1.01 (0.99, 1.03)	0.49	1.02 (1.00, 1.05)	0.24	0.98 (0.94, 1.01)	0.14
Depressive symptoms	1.58 (1.00, 2.51)	**0.05**	1.73 (1.04, 2.87)	**0.04**	1.25 (0.71, 2.23)	0.44
Anxiety symptoms	1.16 (0.74, 1.80)	0.52	1.33 (0.82, 2.17)	0.25	1.07 (0.60, 1.92)	0.82
Caregiver characteristics						
Age	1.05 (1.00, 1.12)	0.12	1.08 (1.01, 1.16)	**0.02**	1.04 (0.97, 1.13)	0.26
Sex (ref: male)	1.38 (0.43, 4.41)	0.58	1.00 (0.27, 3.38)	0.95	1.31 (0.26, 6.56)	0.75
Marital status (ref: married)	0.63 (0.18, 2.20)	0.47	0.47 (1.00, 2.27)	0.34	0.39 (0.05, 3.22)	0.38
Employment status (ref: employed)	3.15 (1.24, 8.01)	**0.02**	3.77 (1.35, 10.50)	**0.01**	1.28 (0.38, 4.32)	0.70
Education (ref: college/university)	0.94 (0.28, 3.16)	0.92	1.09 (0.27, 4.41)	0.90	2.36 (0.28, 19.90)	0.43
Income (ref: \$150,000–more)						
\$49,999 or less	1.10 (0.25, 4.80)	0.90	0.40 (0.06, 2.52)	0.40	1.30 (0.22, 7.86)	0.78
\$50,000–\$99,999	0.74 (0.22, 2.56)	0.64	0.67 (0.17, 2.59)	0.56	1.08 (0.23, 5.06)	0.92
\$100,000–149,999	0.55 (0.14, 2.18)	0.39	0.73 (0.17, 3.17)	0.67	0.23 (0.02, 2.44)	0.22
GAD-7	1.04 (0.96, 1.11)	0.34	1.02 (0.94, 1.10)	0.68	1.04 (0.95, 1.14)	0.45
QIDS	1.00 (0.93, 1.08)	0.96	1.01 (0.93, 1.10)	0.84	1.00 (0.87, 1.10)	0.70
Family characteristics						
APGAR	0.96 (0.80, 1.16)	0.69	0.90 (0.73, 1.10)	0.29	1.08 (0.84, 1.39)	0.56
FIRM	1.01 (0.97, 1.05)	0.58	1.01 (0.96, 1.05)	0.83	1.00 (0.95, 1.06)	0.89
FILE	1.00 (0.92, 1.08)	0.88	1.00 (0.89, 1.07)	0.64	1.02 (0.92, 1.13)	0.76

Notes: Bolded entries indicate statistical significance at p-value ≤ 0.05. OR = odds ratio, CI = confidence interval for coefficient (95%). Unmet overall needs: depressive symptoms pseudo $r^2 = 0.06$; employment status pseudo $r^2 = 0.09$. Unmet physical needs: depressive symptoms pseudo $r^2 = 0.08$; caregiver age pseudo $r^2 = 0.09$; employment status pseudo $r^2 = 0.11$.

4.3. Unmet physical healthcare needs

Bivariable regression analyses showed that patients with higher depressive symptoms had higher odds of unmet physical needs that those with lower depressive symptoms (OR = 1.73, 95% CI: 1.04, 2.87, p = 0.04). Caregivers who were older had higher odds of reporting unmet physical needs in their children than those who were younger (OR = 1.08, 95% CI: 1.01, 1.16, p = 0.02), that is, a one-year increase in caregiver age was associated with 1.08 higher odds of reporting unmet physical needs in their children. Caregivers who were unemployed had higher odds of reporting unmet physical needs in their children than those who were employed (OR = 3.77, 95% CI: 1.35, 10.50, p = 0.01) (Table 2). The other patient factors (such as patient age, sex, age at seizure onset, seizure frequency, number of AEDs, IQ, and patient anxiety), caregiver factors (such as caregiver sex, depression and anxiety, marital status, education, income), and family factors (such as family relationships, resources, and demands) did not predict unmet physical needs (all $p > 0.05$).

Patient depressive symptoms, caregiver age, and employment status were included in the multivariable logistic regression analysis (Table 3b). Patients with higher depressive symptoms had higher odds of unmet physical needs than those with lower depressive symptoms (OR = 2.03, 95% CI: 1.13, 3.66, p = 0.02); caregivers who were older had higher odds of reporting unmet physical needs in their children (OR = 1.10, 95% CI: 1.01, 1.19, p = 0.02); and caregivers who were unemployed had higher odds of reporting unmet physical needs in their children than those who were employed (OR = 6.53, 95% CI: 1.92, 22.23, p = 0.003). The model explained 32% of the variance.

Table 3
Multivariable logistic regression of patient depressive symptoms, caregiver age, and employment status on unmet a) overall and b) physical healthcare needs in children with drug-resistant epilepsy.

a)

Predictors	Overall unmet needs	
	OR (95% CI)	p-Value
Patient depressive symptoms	1.71 (1.05, 2.81)	**0.03**
Employment status (ref: married)	3.82 (1.43, 10.20)	**0.008**

Notes: Bolded entries indicate statistical significance at p-value ≤ 0.05. Model pseudo $r^2 = 0.17$.

b)

Predictors	Unmet physical needs	
	OR (95% CI)	p-Value
Patient depressive symptoms	2.03 (1.13, 3.66)	**0.02**
Caregiver's age	1.10 (1.01, 1.19)	**0.02**
Employment status (ref: employed)	6.53 (1.92, 22.23)	**0.003**

Notes: Bolded entries indicate statistical significance at p-value ≤ 0.05. Model pseudo $r^2 = 0.32$.

4.4. Unmet mental healthcare needs

In the bivariable regression, patient, caregiver, and family factors did not significantly predict unmet mental needs (all $p > 0.05$) (Table 2), therefore, multivariable regression was not conducted.

5. Discussion

Previous studies have reported unmet healthcare needs in children with current or active epilepsy, utilizing data from national health surveys. A prior US study had assessed unmet healthcare needs in children with current epilepsy using the National Survey of Children's Health and showed that the relative risk of unmet medical care and mental healthcare needs was 4.91 and 7.24, respectively, relative to children who were never diagnosed with epilepsy [36]. Reid et al. [11] have evaluated the prevalence of unmet healthcare needs in adults and children with active epilepsy from the CCHS, and found that the prevalence of overall unmet needs was 17.9%. In this study, unmet overall healthcare needs were reported in more than a third of children (37.20%), unmet physical needs in 24.4%, and unmet mental healthcare needs in 15.1%.

The higher proportion of unmet physical needs compared to unmet mental needs in our cohort may be related to the greater need to address intractable seizures in this cohort with DRE than the chronic mental health comorbidities. The higher proportion of unmet needs in our cohort relative to the study by Reid et al. [11] could be due to a variety of factors. The study population in Reid et al. [11] was derived from a national survey, who may have less severe epilepsy, and consisted of both adults and children. In contrast, our study sample consisted of children who were evaluated for epilepsy surgery candidacy in a tertiary pediatric institution, who have poorly controlled epilepsy, and would likely require more healthcare services. Ott et al. [37] have found that 61% of children with complex partial seizures and primary generalized seizures have behavioral disorders, and about two-thirds of these patients did not receive mental health treatment, i.e., they had unmet mental healthcare needs.

Mental health comorbidities are pervasive in children with epilepsy, with up to 60% having depression, anxiety, behavioral, social, or cognitive issues [36,38–40]. In our study, 39 (45.3%) children had symptoms of depression, and 24 (27.9%) had symptoms of anxiety. Despite the high prevalence of depressive and anxiety symptoms in children with DRE, we did not find an association between depressive and anxiety symptoms and unmet mental healthcare needs. Instead, caregivers were more likely to report unmet physical healthcare needs than mental healthcare needs. It is possible that these children present with subclinical symptoms that were impacting on their lives [41], but these symptoms may not be sufficiently severe to be recognized as mental health issues. Further study is required to determine whether caregivers recognize or acknowledge children's mental health issues, their need for mental health services, and potential barriers for accessing mental healthcare, including stigma associated with mental health problems and mental healthcare [42,43].

Children with special healthcare needs with more severe or unstable conditions, and those with emotional, developmental, or behavioral comorbidities, were more likely to have unmet healthcare needs than those with stable conditions or without comorbidities [16]. Children with current epilepsy/seizure disorders also had a higher risk of unmet medical care and mental healthcare need than children with former epilepsy/seizure disorders [36]. In this study, measures of epilepsy severity such as seizure frequency and number of AEDs did not affect unmet healthcare needs. Our study sample included children who were evaluated for epilepsy surgery candidacy, and therefore, our cohort consisted of children with more severe epilepsy. If we had included a broader spectrum of disease severity by including children with well-controlled epilepsy in addition to DRE, we may have identified differences in the likelihood of reporting unmet healthcare needs based on disease severity.

Reporting of perceived unmet healthcare needs was based on caregivers' subjective assessment that their child did not receive the health services that they needed to address their health problems. Hence, we postulated that beyond availability and accessibility issues, patient, caregiver, and family factors could contribute to caregivers' perception of unmet healthcare needs. Previous studies have limited the assessment of caregiver factor to sociodemographic characteristics. Ott et al. [37] found that sociodemographic characteristic such as parental education influenced whether children with epilepsy and behavioral disorders received mental health treatment. Ganz et al. [16] demonstrated that children with special healthcare needs from lower family income, who were uninsured, or had mothers with lower education were more likely to have parental report of unmet mental healthcare needs. These findings support the notion that caregiver factor such as sociodemographic characteristics may have a bearing on the perception of unmet needs. Our study adds to the literature by evaluating caregiver and family characteristics beyond sociodemographic variables that could contribute to the perception of unmet healthcare needs.

We found that older age in caregivers and unemployment increased the odds of reporting unmet physical healthcare needs in children. A prior study on unmet needs in children with autism found that older mothers were more likely to report greater unmet needs in their children [44]. Higher expectations among older mothers may have contributed to greater perceived unmet needs in their children. Unemployment among caregivers also increased the odds of reporting overall unmet needs in children. Sloper and Turner [45] found that caregiver unemployment was a risk factor for unmet healthcare needs in children with severe physical disability. Parents of children with chronic illness reported lower employment rates and less time for leisure activities [46]. Unemployment may represent an additional stressor to the caregivers, which compounds the burden of caring for a child with a chronic neurological disorder. As well, unemployment may reflect a lack of economic resources that could be mobilized to access healthcare services, and contribute to perceived unmet needs.

In our study, SES (as measured by household income) was not associated with unmet healthcare needs. Others have found that household income was inversely associated with unmet healthcare needs related to accessibility problems, but not for unmet needs related to service availability in the setting of universal healthcare system in Canada [47]. We have not distinguished unmet needs because of accessibility or availability issues, which may account for the discordance in findings. Further, a large proportion (76.7%) of our cohort were from medium to high income households. The fact that many of our study participants were from higher household incomes is not unique to this study. Other studies have also demonstrated that those from lower household incomes were less likely to participate in research studies than those from higher household incomes [48,49]. The SES of the study participants are not necessarily a reflection of the SES of children who are referred for epilepsy surgery evaluation. Although there was no significant association between household income and unmet healthcare needs in our cohort, there was a gradient of unmet overall needs, highest among those who earn $49,999 or less, and lowest among those who earn $100,000–149,999, compared to those who earn $150,000 or more. The lack of statistical association could be related to the relatively small sample size in each category of income. In focus groups of caregivers of children with epilepsy, Wu et al. [50] found that there were differences in caregivers' concerns about which types of needs were met or unmet based on their SES. Caregivers from high SES were more concerned about inadequate educational services, but less concerned about mental health and medical service use than caregivers from lower SES. Ott et al. [37] found that lower parental education was associated with unmet mental healthcare needs in children with epilepsy. However, in our study, caregiver education was not associated with unmet needs. A large proportion (83.9%) of the caregivers in our cohort had college or university education, whilst in the study by Ott et al., 46% of their caregivers had less than high school education.

There are several limitations of this study. Since this study was conducted in a sample of children with DRE recruited from the tertiary epilepsy center, the findings may not be generalizable to children with less severe epilepsy or those who were seen in the community practice and have never been referred to a tertiary care epilepsy center. We have used caregiver report rather than patient report of unmet healthcare needs since younger children may not have the cognitive capacity to respond to questions on unmet needs. Caregiver reporting bias could affect study validity. Children with epilepsy may have different perceptions relating to needs for and access to healthcare services compared to their caregivers. A prior study on unmet healthcare needs of adult survivors of childhood tumors showed poor to satisfactory agreement (kappa of 0.26 to 0.46) between survivors and caregiver proxy report of unmet needs, with caregivers reporting more unmet needs in the survivors than did survivors themselves [51]. Another limitation includes the instrument used to measure unmet needs, with binary responses to the question on unmet needs. The questionnaire did not explore the frequency or details of healthcare services that were unmet. Further, the question on unmet physical healthcare needs did not probe into whether the unmet needs were related to epilepsy itself or other factors, long wait times for specialist appointments, or access to antiseizure medications due to costs. Medications are usually not reimbursed by the Ministry of Health and Long-Term Care in Canada, and are either paid for through private insurance or out-of-pocket costs. Costs of medications may have contributed to unmet physical needs in some children. Despite the limitation, the questionnaire on unmet needs was based on Canadian and U.S. national health surveys, CCHS and CSHCN, which have been administered to large samples of the population for over 10 years.

6. Conclusion

More than a third of children in our cohort had unmet healthcare needs. Patients with depressive symptoms, and those with young or unemployed caregivers were at increased risk. Identifying the predictors of unmet healthcare needs is a necessary step in addressing unmet needs in this population with DRE. Future study on unmet healthcare needs would benefit from a multi-informant approach that incorporate patients', caregivers', and providers' perception of unmet needs, as well as deep dive into the frequency or types of healthcare services that were unmet, to arrive at a more comprehensive evaluation on unmet healthcare needs. More importantly, further work is required to identify the remedial steps to address these unmet needs, targeting the modifiable factors. To address these unmet needs, a system-based approach and the involvement of broad stakeholders including families and patients, such as in Project Access [52], are required to improve access to comprehensive and coordinated healthcare.

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Declaration of Competing Interest

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