

Evaluating mental health in caregivers of patients with Dravet syndrome: a systematic review

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Overview



QUESTION

What are characteristics of the available published literature addressing mental health concerns in parents/caregivers of people with Dravet syndrome?

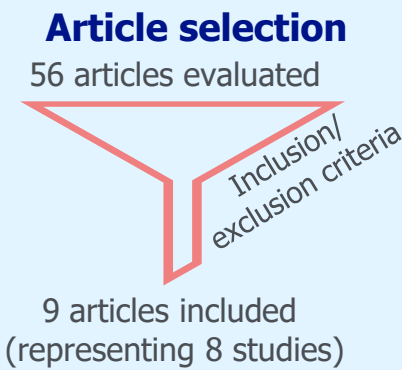


INVESTIGATION

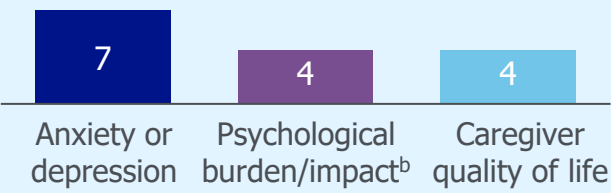
Systematic literature search of PubMed and EMBASE for peer-reviewed articles in English that included findings on mental health in parents/caregivers of people with Dravet syndrome.



RESULTS



Number of articles with mental health findings^a



^aCategories are not mutually exclusive; ^bIncludes aspects of psychological burden/impact other than anxiety or depression.

Major themes identified from article review

- Heterogeneity of study design
- Use of caregiver surveys to measure mental health
- Lack of consistency of tools
- Lack of consistency in mental health terminology



CONCLUSIONS

An unmet medical need exists to properly assess the mental health of parents/caregivers of people with Dravet syndrome.

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Background

- Dravet syndrome (DS) is a rare pharmacotherapy-resistant, developmental and epileptic encephalopathy that is typically diagnosed in early childhood.¹
- DS involves seizures accompanied by severe cognitive, behavioural, and motor impairments.^{2,3}
- Clinical, economic, and humanistic burdens of DS have been characterised in the published literature^{1,3}; these burdens may also affect mental health in caregivers.

Objective

- To characterise the available literature to date addressing the mental health concerns in parents/caregivers of people with DS.

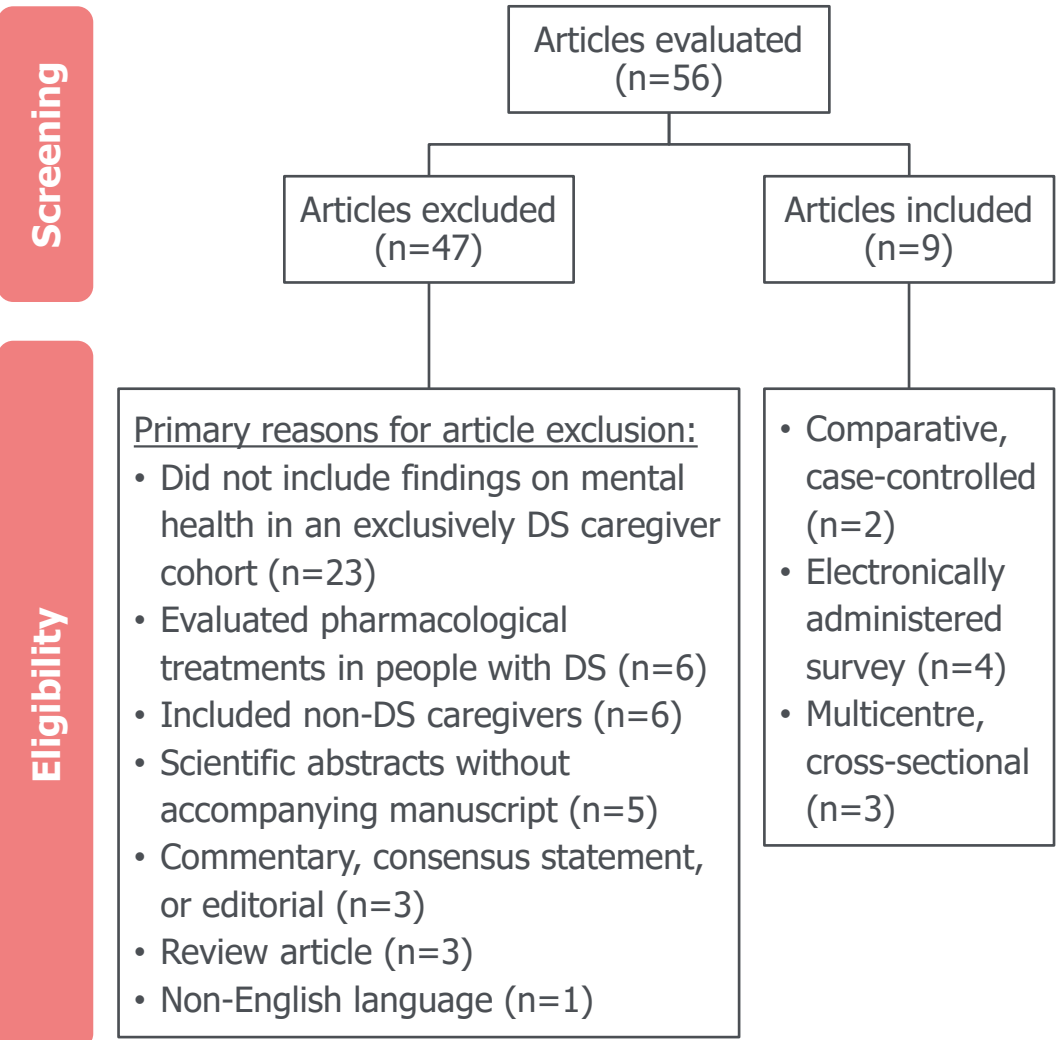
Methods

- Systematic searches of PubMed and EMBASE (beginning of records through November 2023) were conducted using multiple individual search strings.
 - Search terms: (Dravet syndrome OR severe myoclonic epilepsy of infancy OR SMEI) AND (caregiver OR mother OR father) AND (mental health OR depression OR anxiety).
- Inclusion criteria: published manuscript in peer-reviewed journal; mental health in caregivers of people with DS.
- Exclusion criteria: non-English language, case reports, case series, reviews, editorials, studies evaluating pharmacological treatments, or included patients other than those with DS.
- Articles meeting inclusion/exclusion criteria were reviewed for key takeaways/themes.

Results

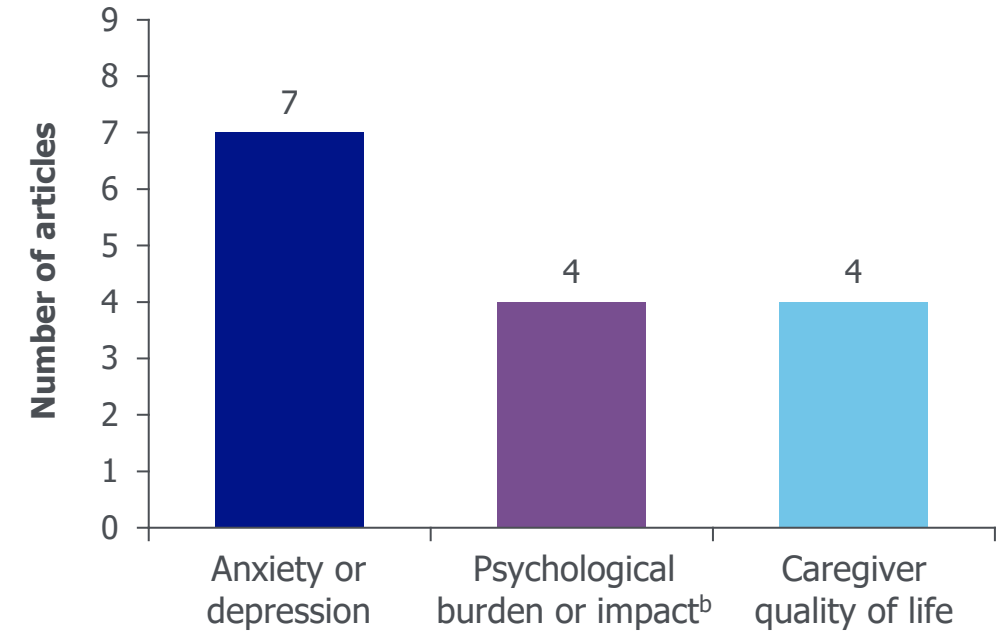
- Of the 56 articles evaluated against inclusion/exclusion criteria, 9 were included in the analysis.
 - Articles were mostly from European countries (n=8) and conducted between 2017 and 2023.
 - The 9 articles represented 8 studies.

Flow diagram of SLR article selection



DS, Dravet syndrome; SLR, systematic literature review.

Number of articles focusing on mental health findings^a



^aCategories are not mutually exclusive; ^bIncludes aspects of psychological burden/impact other than anxiety or depression.

- Major themes identified from the analysis were:
 - Heterogeneity of study design
 - Use of caregiver surveys to measure mental health
 - Lack of consistency of the tools (validated and non-validated) to measure mental health in caregivers
 - Lack of consistency in mental health terminology, with some articles calling out certain domains (eg, anxiety or depression) and others only evaluating “mental health.”

Limitations

- Limitations to this systematic review include minimal published research and insufficient data on questions addressing mental health in caregivers of people with DS.

Characteristics of included studies^{4-10,12,13}

ARTICLE AUTHOR, YEAR	STUDY DESIGN	COUNTRY	AVAILABLE DEMOGRAPHIC DATA	RELEVANT ASSESSMENT TOOLS	KEY FINDINGS	THEMES ^a
Campbell, 2018 ⁴	Electronically administered survey	USA	• Primary caregivers (N=30) that provided care for a friend or family member with DS	EQ-5D-5L	• 70% of caregivers had ≥ slight problems of anxiety/depression • 33% of caregivers had ≥ moderate problems of anxiety/depression	1, 2, 3
Domaradzki, 2023 ^{5,6,b}	Electronically administered survey	Poland	• Caregivers of children with DS (N=75): mothers (n=66; 88%); fathers (n=7; 9.3%); other relatives (grandmother or sister) (n=2; 2.7%) • Mean (SD) age, years: 39.7 (6.5) • Age category, years: <30 (n=2; 2.7%); 30-39 (n=32; 42.6%); 40-49 (n=35; 46.7%); ≥50 (n=6; 8%)	Questionnaire constructed based on themes from literature review	• Parents/caregivers reported experiencing: anxiety/fear (68%), sadness/depression (52%), nervousness/impulsivity (50.7%), and mental exhaustion (80%) ⁵ • Family caregivers reported experiencing: fatigue (84%), a deterioration of mental health (60%), and intimacy problems with their spouse/partner (53.4%) ⁶	1, 2
Gil-Nagel, 2023 ⁷	Multicentre, cross-sectional	Spain	• Caregivers of children with DS (N=80)	CarerQoL	• 68.8% of caregivers reported experiencing mental health problems	1, 2, 3, 4
Maltseva, 2023 ⁸	Multicentre, cross-sectional	Germany	• Primary caregivers of children/adults with DS (N=108): mothers (n=100; 92.6%); fathers (n=8; 7.4%) • Mean (SD) age, years: mothers, 44.7 (10.6); fathers, 47.3 (10.6)	HADS	• Caregivers reported experiencing: <ul style="list-style-type: none">Anxiety: normal (0-7): 38.2% (n=39), borderline (8-10): 26.5% (n=27), abnormal: 35.3% (n=36)Overall mean (SD) HADS anxiety score was 9.3 (4.3)Depression: normal (0-7): 49.1% (n=53), borderline (8-10): 27.8% (n=30), abnormal: 23.1% (n=25)Overall mean (SD) HADS depression score was 7.9 (3.7)	1, 2, 3
Salom, 2023 ⁹	Comparative, case-controlled	Spain	• Caregivers of children with DS (N=48): women (n=34); men (n=14) • Mean (SD) age, years: 41.7 (6.2); 77% active workers; 52% lower middle class	CRESIA	• Caregivers had higher CRESIA scores vs the control group (parents of children without any diagnosed disease) across social, general health, psychological, family, and stress caused by the child domains • The impact on the psychological domain directly affected caregivers' emotional state, which differed between caregivers of children with DS and the control group; (t(94)=4.59, p<0.001)	1, 2, 3, 4
Strzelczyk, 2019 ^{10,c}	Multicentre, cross-sectional	Germany	• Caregivers of children/adults with DS (N=93) • Mean (SD) age, years: mothers (n=93), 42.1 (7.6); fathers (n=93), 45.2 (7.7)	BDI-II, EQ-5D-3L	• Depression (BDI-II scores) symptom severity: none (0-13), 44% (n=41); mild (14-19), 22% (n=20); moderate (20-28), 15% (n=14); severe (29-63), 9% (n=8) • EQ-5D-3L: Caregivers reported higher levels of problems in anxiety/depression compared with German population norms ¹¹ (38.2% vs 4.3%)	1, 2, 3
Strzelczyk, 2019 ^{12,c}	Comparative, case-controlled	Germany	• Caregivers of children/adults with DS (n=93), drug-resistant epilepsy (DRE, n=93), seizure remission (SR, n=93); cohorts were matched based on age and sex	BDI-II, EQ-5D-3L	• Depression (BDI-II scores) symptom severity: none (0-13), 50% (n=37); mild (14-19), 26% (n=19); moderate (20-28), 16% (n=12); severe (29-63), 8% (n=6); mean total BDI-II score: 14.9 <ul style="list-style-type: none">More DS caregivers had moderate to severe depression symptoms compared with caregivers of children with DRE or SR (24% vs 11% or 5%)	1, 2, 3
Villas, 2017 ¹³	Electronically administered survey	USA ^d	• Parents/caregivers of children/adults with DS (N=256); 55% female (n=140)	Survey based on parent-reported surveys and support group discussion boards	• 66% of caregivers reported having suffered from depression; however, only 26% had received some form of family therapy. In open response sections of the survey, 19 caregivers reported anxiety/depression/isolation	1, 2

^aFour themes: (1) heterogeneity of study design, (2) use of caregiver surveys to measure mental health, (3) lack of consistency of the tools (validated/non-validated) to measure mental health in caregivers, (4) lack of consistency in mental health terminology, with some studies calling out certain domains (eg, anxiety or depression) and some studies only evaluating “mental health”; ^bRepresent separate analyses of the same study; ^cRepresent the same DS dataset; ^dCountries of caregivers: 70% USA and 30% United Kingdom, Europe, Australia, Canada, Central/South America, and other regions. BDI-II, Beck Depression Inventory-Second Edition; CarerQoL, Care-related Quality of Life instrument; CRESIA, Childhood Rare Epilepsy Social Impact Assessment; DS, Dravet syndrome; EQ-5D-3L, EuroQoL 5-Dimensions 3-Levels; EQ-5D-5L, EuroQoL 5-Dimensions 5-Levels; HADS, Hospital Anxiety and Depression Scale.

Conclusions

- An unmet medical need exists to properly assess mental health needs of parents/caregivers of people with DS.
- The use of consistent and validated tools, as well as consistent terminology, are needed to properly ascertain the prevalence of mental health concerns in parents/caregivers of people with DS.
- Implementation of a comprehensive mental health support system may provide benefits for caregivers of people with DS.

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