

ALEXANDRA BURDASS BSc (Hons) MSc

EXPLORING PSYCHOSOCIAL WELLBEING AND QUALITY OF  
LIFE IN YOUNG PEOPLE WITH NARCOLEPSY.

Section A: Psychosocial Wellbeing and Quality of Life in Children  
and Adolescents with Narcolepsy: A Narrative Review

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Perceptions and Psychosocial Outcomes, and the Mediating Role  
of Coping in Young Adults with Narcolepsy

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## **Summary of MRP Portfolio**

### **Section A**

This is a narrative literature review of research investigating the psychosocial wellbeing and quality of life in children and adolescents with narcolepsy. The review summarises and evaluates thirteen studies that found psychosocial difficulties and diminished quality of life in this population. Clinical implications include the need for preventive strategies subsequent to diagnosis, alongside psychological interventions targeting psychosocial wellbeing and quality of life. Research implications include prioritising the exploration of underlying mechanisms that may account for these findings, whilst taking into account the methodological recommendations emphasized.

### **Section B**

This is a mixed methods cross-sectional study of young adults with narcolepsy. It explores their illness perceptions, coping strategies and psychosocial outcomes, and the potential relationships between these variables. Results showed that this population experiences high levels of perceived threat in relation to their condition, poorer quality of life than the general young adult population, and moderate levels of anxiety and depression. They also engage in predominantly problem-focused coping strategies and find sleeping, napping and taking medication the most helpful ways to cope with their condition. Finally, higher levels of illness threat perception were associated with lower physical and mental quality of life. The implications of findings for research and clinical practice with this population are discussed.

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Alexandra Burdass BSc (Hons) MSc

Major Research Project

Section A: Literature Review

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## **Abstract**

**Background:** Evidence suggests that children and adolescents with narcolepsy are at risk of experiencing psychosocial difficulties and poor quality of life. This evidence, however, has not been collectively synthesised to date.

**Method:** A narrative literature review was conducted to identify research investigating the psychological functioning, social functioning and quality of life in children and adolescents with narcolepsy.

**Results:** Thirteen papers were identified, and the majority of these papers were evaluated to be of good quality. This population was found to experience marked levels of anxiety and depression that were significantly higher than healthy controls. Social functioning was impaired in this population in the domains of family relationships, social difficulties and educational difficulties. Finally, the quality of life in children and adolescents with narcolepsy was significantly diminished compared with healthy controls, however the specific domains most impacted remains unclear.

**Conclusions:** This review highlighted the need for preventative strategies subsequent to diagnosis, alongside psychological interventions targeting psychosocial wellbeing and quality of life. In future investigations, it is essential to prioritise the exploration of underlying mechanisms that may account for these findings, while taking into account the methodological recommendations emphasised in this review.

**Keywords:** *narcolepsy, psychosocial wellbeing, quality of life, children, adolescents*

## **Introduction**

### **Narcolepsy in Children and Adolescents**

Narcolepsy is a chronic, neurological condition that affects the brain's ability to control wakefulness and sleep. There are two types of narcolepsy; type 1 narcolepsy and type 2 narcolepsy. Both types are characterised by excessive daytime sleepiness, however, people with type 1 narcolepsy also experience cataplexy, which is sudden muscle weakness triggered by emotion, whereas people with type 2 narcolepsy do not usually experience cataplexy (Slowik, 2021). Whilst narcolepsy was traditionally thought to be a condition experienced by adults, research now shows that the symptoms often first appear in childhood and early adolescence, with more than 50% of people with narcolepsy reporting the onset of their symptoms starting before the age of 18 (Blackwell et al., 2022). As well as experiencing the physical symptoms of their condition, children and adolescents with narcolepsy require strict management regimes such as a regular sleep/wake schedule and daily naps (Chung et al., 2022) which can make it difficult to keep up with school work and cause isolation from their peers (Barlow et al., 2006), all which can have a significant impact on their psychosocial wellbeing.

### **Narcolepsy and Psychosocial Wellbeing**

Psychosocial wellbeing encompasses both psychological wellbeing and social functioning as essential components. Psychological wellbeing is evaluated by considering mental health indicators such as symptoms of anxiety and depression. Social functioning, on the other hand, is assessed by examining various aspects including family relationships, educational difficulties, and social difficulties (Eiroa-Orosa, 2020). Research has shown that both children and adolescents with narcolepsy are significantly affected in both of these

domains. For example, a body of evidence suggests that this population experiences a higher prevalence of depression and anxiety disorders compared with the general population (Barker et al., 2020), and also experiences poor school performance and social withdrawal (Chen et al., 2020).

### **Narcolepsy and Quality of Life**

These psychosocial problems experienced by children and adolescents with narcolepsy are thought to have an impact on their quality of life. Quality of life is a multidimensional construct that measures an individual's subjective sense of wellbeing, their perception of the impact of disease and treatment, as well as their satisfaction within the physical, psychological and social domains (Ravens-Sieberer et al., 2022). A study on 117 children and adolescents with narcolepsy found that the condition significantly impacted their quality of life, and that depressive symptoms played a major role in this (Inocente et al., 2014).

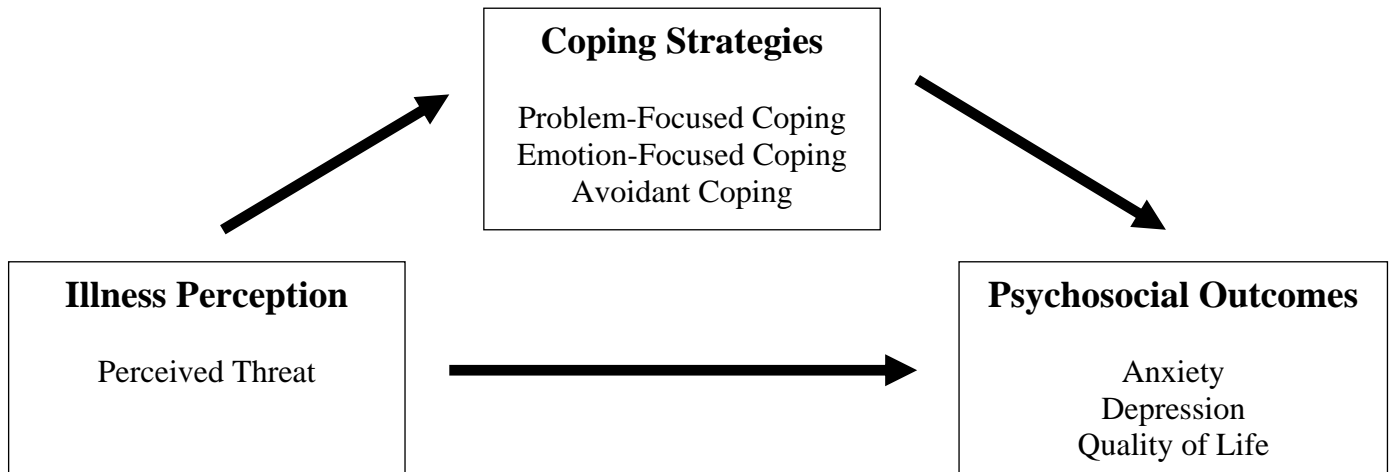
### **The Common Sense Model of Illness Representations**

Whilst it is clear that living with narcolepsy can significantly impact children and adolescents' psychosocial functioning and quality of life (Barker et al., 2020), research has shown that symptom severity alone is not sufficient in explaining this impact. Discrepancies have been found between the underlying pathology of chronic conditions, such as narcolepsy, and the degree of psychosocial dysfunction individuals experience (Verhoof et al., 2014). This has caused an interest into how psychological mechanisms may account for the variation in adjustment and outcomes for people with chronic conditions; a theory that has received substantial attention and theoretical development is Leventhal's Common Sense Model of Illness Representations, known as the Common Sense Model (Leventhal et al., 1980;

Leventhal et al., 1984). This model proposes that people with chronic conditions receive information about their illness from a wide range of sources, such as healthcare professionals, their cultural and social environment, as well as their own experience of their condition (Hagger et al., 2003). This information leads individuals to develop certain beliefs about their condition, which can be categorised into five dimensions: beliefs about the identity of their symptoms and condition, beliefs about the cause of their condition, beliefs about the duration of their condition, the personal consequences of living with their condition and the control they have over their condition (Cameron et al., 2003). These beliefs influence what coping strategies an individual uses to manage their symptoms or 'cure' their condition. Coping strategies include problem-focussed coping which involves planning and informational support, emotion-focused coping which involves humour, acceptance and self-blame, and finally avoidant coping which involves strategies such as self-distraction, substance use and denial (Carver, 1997). If an individual uses more adaptive coping mechanisms then they are more likely to have positive psychosocial outcomes and a better quality of life, whereas if an individual uses maladaptive coping strategies then they are more likely to have negative outcomes in these domains. The Common Sense Model (Figure 1) is therefore a mediation model where coping strategies mediate the influence of a person's illness perceptions on their wellbeing and outcomes (Hagger et al., 2003).

**Figure 1**

*Leventhal's Common Sense Model of Illness Representations (Leventhal et al., 1980)*



### **Applying the Model to Children and Adolescents with Narcolepsy**

A body of research has demonstrated how this model can be applied to a range of chronic conditions, and how illness perceptions and coping are strongly related to health outcomes including psychosocial wellbeing and quality of life (Cheng et al., 2020; Gurkova et al., 2018). Furthermore, these findings have been demonstrated in children and adolescents with chronic conditions (Grey et al, 2007; Verhoof et al., 2014; Stapersma et al., 2019). It could therefore be hypothesised that the Common Sense Model (Leventhal et al., 1980) could also be applied to children and adolescents with Narcolepsy to explain the reported impact on their psychosocial wellbeing and quality of life. However, there is a lack of research on both the patterns of illness perceptions and coping strategies of children and adolescents with narcolepsy, and therefore these areas need to be explored by future research to confirm this.

## **Rationale**

Whilst the body of evidence on the psychosocial outcomes and quality of life in children and adolescents is growing, there have been a limited number of reviews synthesising this information; reviews have predominantly focused on the literature about adults with narcolepsy (Tadrous et al., 2021; Raggi et al., 2019; Schiappa., 2018). Whilst one review did synthesise the literature on children and adolescents with narcolepsy, it focused on their cognitive function alongside psychosocial wellbeing (Blackwell et al., 2017). Therefore, there is a need to update the literature on the psychosocial wellbeing of children and adolescents with narcolepsy and synthesise the literature on the quality of life in children and adolescents with narcolepsy. Doing so will help inform what psychosocial interventions might be useful for this population, as well as provide a platform for future research studies investigating and collating the patterns of illness perceptions and coping strategies in children and adolescents with narcolepsy.

## **Aims**

This paper therefore aims to focus on the following questions:

1. What is the psychological functioning of children and adolescents with narcolepsy, measured by symptoms of mental health e.g. anxiety and depression?
2. What is the social functioning of children and adolescents with narcolepsy measured by elements such family relationships, educational difficulties and social difficulties?
3. What is the quality of life of children and adolescents with narcolepsy?



## **Method**

### **Literature Search**

The search was conducted on 22<sup>nd</sup> July 2022 using the databases PsychINFO, MEDLINE, ASSIA (Applied Social Sciences Index & Abstracts) and CINAHL (Cumulative Index to Nursing & Allied Health Literature). Search terms were selected based on the literature review's aims which generated the following terms: (Narcolepsy OR cataplexy) AND (Adolesc\* OR Child\* OR young) AND (Psycho\* wellbeing OR social functioning OR mental health OR anx\* OR depress\* OR quality of life OR QoL OR quality-of-life). Additional articles were hand searched for in reference sections of the included studies, however, no additional literature was found. Articles were initially screened by title and abstract using the following inclusion and exclusion criteria, and the remaining full texts were read to assess for eligibility.

### **Inclusion/ Exclusion Criteria**

The following inclusion criteria was applied in order to meet the aims of the review:

1. Studies were included if the participants were children or adolescents (between ages 0 and 20) with a diagnosis of narcolepsy (with or without cataplexy).
2. Studies using parent measures were also included as parental outcomes are often used to assess psychosocial wellbeing and quality of life in children.
3. Studies using outcome measures and themes related to psychosocial wellbeing and quality of life were included .
4. Qualitative, quantitative and mixed methods studies were included.
5. Studies had to be published in English, in a peer-reviewed journal to be included.

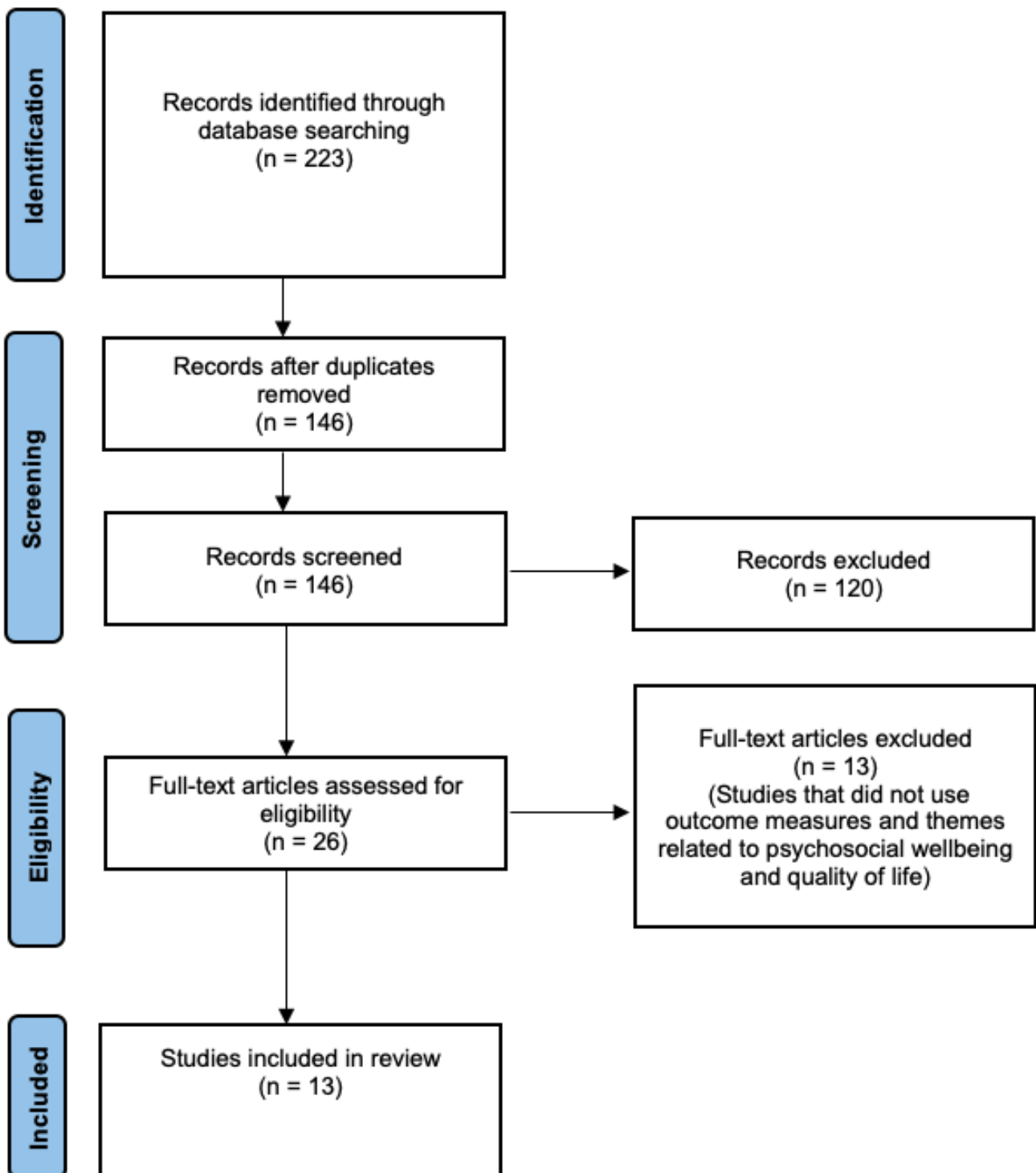
The following exclusion criteria was applied in order to meet the aims of the review:

1. Reviews, metaanalyses and commentaries were excluded.
2. Studies using participants with a condition other than narcolepsy were excluded.
3. Studies using outcome measures and themes that were not related to psychosocial wellbeing and quality of life were excluded.

The PRISMA flow diagram (Figure 2) illustrates the number of papers retrieved at each stage of the screening process and presents the database search's results. Following the screening procedure for eligibility, thirteen papers were included in the review; see Table 1 for study-specific information.

**Figure 2**

*PRISMA Diagram of Literature Search and Study Selection*



## **Quality Assessment Tools**

Due to the differences in study design, sample size, outcome measures and statistical analysis a meta-analysis was deemed inappropriate to meet the review's aims. Consequently, a narrative literature review was utilised instead. Due to the different types of study included in the review, a range of quality appraisal tools were used to assess their quality. The Critical Appraisal Skills Programme (CASP) checklist for case control studies (Appendix A), the Critical Appraisal Skills Programme (CASP) checklist for cohort studies (Appendix B) and the Appraisal tool for Cross-Sectional Studies (AXIS) (Appendix C) were selected. These tools use checklists and questions to guide the researcher's assessment of quality for each section of the study. Consequently, the user's quality assessments are subjective and differences may be found between researchers. The studies in Table 1 have been colour coded to demonstrate the researcher's subjective assessments of quality; studies highlighted in green have been assessed to have good quality, studies highlighted in orange have been assessed to have medium quality, and studies highlighted in red have been assessed to have poor quality. A summary of the quality assessment using these tools is included below.

**Table 1***Summary of Reviewed Studies*

Authors	Population	Study Design	Outcome Measures	Main Findings
Guilleminault et al., 1998	51 children with narcolepsy  Mean age of 7.9 years and a range of 2.1 to 11.8 years.  Participants recruited in America.	Longitudinal cohort design  Quantitative	A short questionnaire used by one of the authors during his training in child psychiatry that covers overall mental health status.  School reports	90% of children with narcolepsy were ashamed of their symptoms  80% of children with narcolepsy reported feelings of helplessness with their symptoms  83% of children with narcolepsy considered their illness a significant handicap to acceptance in school, athletics, and social activities  80% of children with narcolepsy indicated being depressed by their inability to be the same as their peers despite not having any physical signs  90% of children with narcolepsy expressed a desire to hide their illness from peers in school  20% of children with narcolepsy were thought to have symptoms of reactive depression with loss of appetite, withdrawal from social interaction, crying spells, loss of interest in activities, or loss of self-esteem  31% of children with narcolepsy were disciplined by teachers for recurrently falling asleep  62% of children with narcolepsy had negative reports related to poor academic performances, with "poor attention span, poor concentration, and memory problems"
Hovi et al., 2022	94 children with narcolepsy  Age of participants not clear  Participants recruited in Finland	Cross-sectional design  Quantitative	Child Behaviour Checklist (CBCL)  Youth Self-Report (YSR)  Children's Depression Inventory (CDI)  Parental Resources Questionnaire  Parental Quality of Life Questionnaire	25% of children had CDI scores that suggested depression  41% had total CBCL problem scores above the clinically significant limit  48% of children were anxious and withdrawn  Sleep latency was weakly associated with the CBCL total problem score  Half of the children needed psychiatric interventions  Parental stress was common
Inocente et al., 2014a	88 children with narcolepsy  Median age of 12 years and a range of 5 to 17.5 years  Participants recruited in France	Cross-sectional design  Quantitative	Children's Depression Inventory (CDI)	25% of the children had clinically significant depressed feelings  Depressive patients were older at diagnosis than non-depressive children  When considering age, the depression scores were significantly higher in girls than in boys, only in the children older than the age of 10 years  In the multivariate model adjusted for gender and age, only fatigue explained depression score variability.
Inocente et al., 2014b	117 children with narcolepsy  Mean age of 11.6 years and a range of 5 to 17 years	Case control design  Quantitative	The Children's Depression Inventory (CDI)  Conners Parents Rating Scale-Revised (CPRS-R)  Adapted VSP-A	25% of the patients versus 15.6% of the control subjects had clinically significant depressive feelings  41% of the patients versus 7.5% of the controls reported school difficulties (p=0.002)  28% of the patients versus 7.5% of the controls did not pass a grade and repeated it prior to narcolepsy diagnosis (p<0.001)  30% of the patients versus 8.9% of the controls had absenteeism

	69 control children with a mean age of 13.5 years and a range of 7 to 17 years			(p=0.002)	Compared with the control children the narcoleptic children had significantly lower HRQL (p=0.001) with lower vitality (p=0.003), general well-being (p=0.002), poorer self-image (p=0.03) and tended to have less contact with their parents (p=0.06) and lower school performances (p 0.06)
	Participants recruited in France				The narcoleptic adolescents had a lower quality of life index (p=0.008), lower physical well-being (p<0.001), fewer friends (p=0.001), and leisure activities (p=0.006) than the control adolescents
Parmar et al., 2019a	30 adolescents with narcolepsy	Cross-sectional design Quantitative	Children's Depression Inventory (CDI)		23.3% of participants had CDI total scores in the elevated range and 10% reported suicidal ideation.  Greater CDI scores were associated with worse self-reported sleep quality, greater excessive daytime sleepiness, and lower self-reported physical activity levels
	Mean age of 13.8 ± 2.2				
	Participants recruited in France				
Parmar et al., 2019b	30 adolescents with narcolepsy	Case control design Quantitative	PedsQL family impact module		All scores in the narcolepsy group were similar to the chronic pain group.  Family functioning in adolescents with narcolepsy is impaired and similar to adolescents with chronic pain.  Scores in the worry domain were the lowest of all domains, which asks about worries related to medical treatments efficacy, treatment side-effects, others reaction to their child's illness, illness affecting other family members, and the child's overall future (non significant).
	30 controls				
	Mean age of 13.8 with a range of 10 to 18 years (aged 10 to 18 years)				
	Participants recruited in Canada				
Qu et al., 2022	267 children and adolescents with narcolepsy	Cross sectional design Quantitative	Chinese version of the Depression Self-Rating Scale for Children (DSRS-C)		Narcolepsy patients with ADHD symptoms tended to have more anxiety symptoms, more impulsive behaviour and lower quality compared with narcolepsy patients without ADHD symptoms (p<0.05)
	Children were >12 years and adolescents were 23-18 years		Screen for Child Anxiety Related Emotional Disorders (SCARED)		
	Participants recruited in China		Behaviour Rating Inventory of Executive Function (BRIEF)		
			Inventory of Subjective Life Quality for Chinese children and adolescents (ISLQ)		
Quaedak et al., 2019	53 children and adolescents with narcolepsy	Case control design Quantitative	Social Responsiveness Scale		Children with narcolepsy scored significantly higher on the total score of the SRS, compared to the healthy control group (p<0.001)
	Mean age of 12.1 years and a range of 7-16 years		The Child Behaviour Checklist (CBCL)		Children with narcolepsy scored significantly higher on the sum of the CBCL subscales indicative of social functioning compared to controls (p<0.001)
	64 healthy controls				45.3% of children with narcolepsy reported at least mild to moderate difficulties in social functioning compared to 10.9% of healthy controls (p<0.001)
					20.8% of children with narcolepsy reported severely impacted social functioning compared to 1.6% of healthy controls (p<0.001)

	Mean age of 11.2 years and a range 7 to 16			Social problems were significantly more frequent in girls with narcolepsy compared to boys ( $p < 0.001$ )
	Participants recruited in the Netherlands			
Rocca et al., 2016	29 children and adolescents with narcolepsy with an age range of 7 to 16 years	Case control design Quantitative	The Child Behaviour Checklist (CBCL) Paediatric Quality of Life Inventory (PedsQL)	Patients with narcolepsy had more behavioural problems than IE patients and healthy controls A higher incidence of anxious/depressed, withdrawn/depressed, social, thought, attention problems, somatic complaints, and aggressive behaviours were recorded.
	39 children and adolescents with idiopathic epilepsies (IE) (parallel patient group) with an age range of 7 to 15 years			
	39 health controls with an age range of 6 to 18 years			
	Participants recruited in Italy			
Stores et al., 2006	41 children with narcolepsy with a mean age of 12.46 years	Case control design Quantitative	The Strengths and Difficulties Questionnaire The Child Depression Inventory (CDI)	Children with narcolepsy and children with EDS showed significantly higher rates of behavioural problems and depression than the healthy controls. Children with narcolepsy and children with EDS had significantly poorer quality of life than the healthy controls
	18 children with excessive daytime sleepiness (EDS) with a mean age of 14.26 years		The 50-item Child Health Questionnaire Educational Assessment	Children with narcolepsy and children with EDS had significantly more education problems than the healthy controls There was no significant difference between the children with narcolepsy and the children with EDS for these outcomes
	23 healthy controls with a mean age of 11.30			
	Participants recruited in UK, US, Europe and Australia			
Szakacs et al, 2015	38 children and adolescents with	Cross-sectional design Quantitative	The Development and Well-Being Assessment (DAWBA)	43% of the patients in the PHV narcolepsy group fulfilled the DSM-IV criteria for at least one psychiatric disorder 20% of the patients in the PHV narcolepsy group had major

	<p>narcolepsy with a median age of 15.3 years</p> <p>31 participants had post-H1N1 vaccination (PHV) narcolepsy and 7 participants had non-post-H1N1 vaccination (nPHV) narcolepsy</p> <p>Participants recruited in Sweden</p>		<p>Spectrum Screening Questionnaire (ASSQ)</p>	<p>depression</p> <p>10% of the patients in the PHV narcolepsy group had general anxiety disorder</p>
<p>Szakacs et al., 2019</p>	<p>37 children and adolescents with narcolepsy Mean age of 13.9 years and a range of 8 to 20 years</p> <p>40 controls with an age range of 5 to 20 years</p> <p>31 participants had post-H1N1 vaccination (PHV) narcolepsy and 6 participants had non-post-H1N1 vaccination (nPHV) narcolepsy</p> <p>Participants recruited in Sweden</p>	<p>Case control design</p> <p>Quantitative</p>	<p>KIDSCREEN-10</p> <p>NARQoL-21</p> <p>Adaptive Behaviour Assessment System (ABAS) was used</p> <p>Parenting Stress Index (PSI/SF)</p> <p>The Development and Well-Being Assessment (DAWBA)</p>	<p>Health related Quality of Life was significantly diminished in all domains in the PHV group (<math>p = 0.001</math>) compared to controls</p> <p>Health related Quality of Life was significantly diminished in the School/Concentration domain in the nPHV group (<math>p = 0.004</math>) compared to controls</p> <p>Parents rated significantly higher Total stress, Parent-child dysfunctional interaction, and Difficult child scores compared with parents of controls (<math>p = 0.001</math>, <math>p = 0.005</math>, <math>p &lt; 0.001</math>)</p>
<p>Zhang et al., 2020</p>	<p>46 children with narcolepsy</p> <p>Mean age of 12 years and a range of 5 to 17 years</p> <p>Participants recruited in France</p>	<p>Cross sectional design</p> <p>Quantitative</p>	<p>The Children's Depression Inventory(CDI)</p> <p>VSP-A</p>	<p>Depression was the factor that most affected the quality of life for children with narcolepsy (44%), followed by insomnia (30%) and ADHD symptoms (17%).</p>



## Critique

### Aim and Design

All of the studies clearly stated their aims and provided detailed summaries of the relevant literature. The majority of the studies assessed psychological functioning in children and adolescents with narcolepsy (Guilleminault et al., 1998; Hovi et al., 2022; Inocente et al., 2014a; Inocente et al., 2014b; Parmar et al., 2019a; Qu et al., 2022; Rocca et al., 2016; Stores et al., 2006; Szakacs et al., 2015; Szakacs et al., 2019; Zhang et al., 2020). Eight of the studies assessed social functioning in children and adolescents with narcolepsy (Guilleminault et al., 1998; Hovi et al., 2022; Inocente et al., 2014b; Parmar et al., 2019b; Quaedack et al., 2019; Rocca et al., 2016; Stores et al., 2006; Szakacs et al., 2019), and six of the studies assessed quality of life in children and adolescents with narcolepsy (Inocente et al., 2014b; Qu et al., 2022; Rocca et al., 2016; Stores et al., 2006; Szakacs et al., 2019; Zhang et al., 2020).

All of the studies were quantitative in nature, and all but one of the studies used a cross-sectional design. Six of the cross-sectional studies utilised a control group, and the remaining study used a longitudinal design. The lack of longitudinal studies means that it is difficult to prove the long term relationship between narcolepsy in children and adolescents and measures of psychosocial wellbeing and quality of life (Mueller-Peltzer et al., 2020). However, narcolepsy in children and adolescents has only recently been recognised, and consequently this population has historically been subjected to misdiagnosis and under diagnosis (Blackwell et al., 2022). The novelty of this diagnosis in children and adolescents will have made it difficult for researchers to conduct the longitudinal studies needed to draw stronger conclusions about the relationship between psychosocial symptoms, quality of life

and a diagnosis of narcolepsy for this population; future studies, however, should make this a research priority.

### **Population and Recruitment**

All of the studies utilised convenience sampling from specialist narcolepsy services and hospitals. Whilst this method of sampling was appropriate given the nature of the clinical population, it may have missed children and adolescents with milder forms of narcolepsy who were not under the care of specialist services. Consequently, the samples could be skewed towards more severely affected patients (Inocente et al., 2014a). The majority of the studies were also conducted in Western populations such as America, Finland, France, Canada, the Netherlands, Italy and Sweden, with only one study being conducted in an Eastern population (Qu et al., 2022). Therefore, the generalisability of the findings to children and adolescents with narcolepsy are limited to predominantly Western populations.

Furthermore, only two of the studies reported the ethnicity of participants (Guilleminault et al., 1998; Parmar et al., 2019a) and all of the studies failed to comment on or explore differences in ethnicity in relation to narcolepsy. Whilst the relationship between narcolepsy and ethnicity has largely been unexplored, the available research suggests that there are differences in age of symptom onset as well as differences in the most common symptoms between ethnicities (Kiran, 2015). Whilst it is not yet clear whether these differences are due to genetics, environmental factors, of cultural factors, it is important studies explore this in order to help further our understanding of how narcolepsy may present in different individuals, which may contribute to earlier diagnoses and treatments (Kiran, 2015).

The sample size varied between studies; the smallest sample sizes of 30 participants were found in the studies by Parmar et al. (2019a) and Parmar et al. (2019b), and the largest sample size of 267 participants was found in the study by Qu et al. (2022). All of the studies used established methods to confirm a diagnosis of narcolepsy in their population apart from two studies who ensured a diagnosis had previously been given to their participants by a sleep medicine team (Parmar et al., 2019a; Parmar et al., 2019b).

Six of the studies used a control group that allowed for comparisons to be made between children and adolescents with narcolepsy, children and adolescents with other chronic conditions, and healthy children and adolescents. This is particularly important because psychosocial problems and the consequential detriment to quality of life is increasingly common in all children and adolescents. Including a control group in the studies allowed for conclusions to be drawn about whether the findings related specifically to their developmental stage or whether their diagnosis had an impact as well. Four of the six studies that used a control group ensured that the participants were age and sex matched between groups (Quaedack et al., 2019; Rocca et al., 2016; Stores et al., 2006; Szakacs et al., 2019). The two remaining studies used statistical analysis to ensure that no effect was found for age (Inocente et al., 2014b; Parmar et al., 2019b). Three of the studies used healthy controls to compare psychosocial functioning and quality of life in children and adolescents (Inocente et al., 2014b; Quaedack et al., 2019; Stores et al., 2006; Szakacs et al., 2019) and three studies used parallel patient control groups. Parmar et al. (2019b) used secondary data from a chronic pain population as a control, however, there was a large difference in the size of the two groups, and no comparison was made to a baseline group, limiting the strength of conclusions drawn and inferences made in this study. Two studies, however, used parallel patient control groups who experienced daytime sleepiness as well as a healthy control group (Rocca et al.,

2016, Stores et al., 2006). This allowed stronger inferences to be drawn about the psychosocial wellbeing and quality of life in children and adolescents with narcolepsy, and whether the findings were specific to their diagnosis, their symptomology of daytime sleepiness, or their stage of development.

### **Outcome measures**

A variety of outcome measures were used to assess for psychological wellbeing, social functioning and quality of life. Whilst the most common outcome used for measuring psychological wellbeing and specifically symptomology of depression was the Children's Depression Inventory (CDI) (Hovi et al., 2022; Inocente et al., 2014a; Inocente et al., 2014b; Parmar et al., 2019a; Stores et al., 2006; Zhang et al., 2020), the Depression Self-Rating Scale for Children (DSRS-C) was also used (Qu et al., 2022). The SCARED was used in one study to measure symptomology of anxiety (Qu et al., 2022), and the DAWBA (Szakacs et al., 2015) and CBCL (Hovi et al., 2022; Quaedack et al., 2019; Rocca et al., 2016) were used to give a more global measures of psychological wellbeing. Social difficulties were measured using a wide range of outcomes such as the Youth Self Report (YSR) (Hovi et al., 2022;), the SRS (Quaedack et al., 2019) the Conner's' Parent Rating Scale (CPRS) (Inocente et al., 2014a; Inocente et al., 2014b; Zhang et al., 2020), the SDQ (Stores et al., 2006), the ABAS (Szakacs et al., 2019) and non-validated Likert scales and questions composed by the research team (Hovi et al., 2022; Inocente et al., 2014a; Stores et al., 2006). Quality of life was measured by a range of outcomes such as the VSP-A (Inocente et al., 2014b, Zhang et al., 2020), the PedsQL (Parmar et al., 2019b; Rocca et al., 2016) the CHQ (Stores et al., 2006), the KIDSCREEN10 (Szakacs et al., 2019) and the NARQoL-21 (Szakacs et al., 2019).

The range of different assessments and standardised tests in these studies makes the comparison and accumulation of evidence more difficult. Future researchers could focus on coming to an agreement about which outcome measures are appropriate for exploring psychosocial difficulties and quality of life in children and adolescents with narcolepsy to allow for direct comparisons between studies. Guilleminault et al. (1998) was the only study to use an unvalidated questionnaire and referred to assessing psychosocial wellbeing using “a short questionnaire used by one of the authors during his training in child psychiatry that covers overall mental health status” and “school reports”. This reduces the strength of conclusions drawn from this study, however, it is important to note that this was one of the early descriptive studies that looked at multiple aspects of paediatric narcolepsy, such as diagnosis and treatment strategies, and was important in providing a platform for subsequent studies on these constructs.

Finally, eleven of the studies used self-report outcome measures (Guilleminault et al., 1998; Hovi et al., 2022; Inocente et al., 2014a; Inocente et al., 2014b; Parmar et al., 2019a; Qu et al., 2022; Rocca et al., 2016; Stores et al., 2006; Szakacs et al., 2015; Szakacs et al., 2019; Zhang et al., 2020), eleven of the studies used parent-report outcome measures, (Guilleminault et al., 1998; Hovi et al., 2022; Inocente et al., 2014a; Inocente et al., 2014b; Parmar et al., 2019b; Qu et al., 2022; Quaedack et al., 2019; Rocca et al., 2016; Stores et al., 2006; Szakacs et al., 2019; Zhang et al., 2020) and two of the studies used teacher-report outcome measures (Guilleminault et al., 1998; Stores et al., 2006). It is important for studies to use all three of these measurement methods, such as Stores et al. (2006), in order to get a wider perspective and more accurate measurement of the psychosocial difficulties and quality of life in children and adolescents with narcolepsy, and reduce potential sources of bias within report methods (Fisher et al., 2014).

## **Data Analysis and Limitations**

The majority of studies documented their data analysis procedures well and used a range of descriptive and inferential statistical methods to achieve their aims. One study (Guilleminault et al., 1998) did not make reference to their data analysis methods, however, the results were clearly presented. Two studies made reference to how they managed missing data in their analysis (Inocente et al., 2014b; Zhang et al., 2020), and one study acknowledged their limited sample size and the impact of this on their analysis (Parmar et al., 2019a). All of the studies reported key findings in relation to their original aims, and all studies apart from one (Guilleminault et al., 1998) made reference to their study's limitations.

## **Critique Summary**

The majority of the studies in this review were deemed to be of good quality. Guilleminault et al. (1998) was judged to have the lowest quality out of the papers, however, it was the oldest study that intended to give a more global, descriptive picture of paediatric narcolepsy as opposed to ascribing to more specific aims. The field of paediatric narcolepsy is relatively new, which explains the limited number of papers in this review. Future research should focus on using longitudinal research methods with both parallel patient groups and healthy control groups to strengthen the conclusions drawn about psychosocial wellbeing and quality of life for children and adolescents with narcolepsy. All of the studies included in the review were quantitative in nature, and therefore it would also be beneficial to conduct more qualitative research to get a richer picture of these constructs.

## **Synthesis of findings**

The following synthesis summarises the findings of the papers included in the review. The literature summary is broken down into the following three sections to address the aims of the review; psychological wellbeing, social functioning, and quality of life in children and adolescents with narcolepsy.

### **Psychological wellbeing**

Studies that explored psychological difficulties all came to the consensus that this population had marked levels of anxiety and particularly marked levels of depression. A collection of studies found that 20-30% of the children and adolescents with narcolepsy experienced depressive symptomology that scored above the clinical threshold suggesting the presence of depression (Guilleminault et al., 1998; Hovi et al., 2022; Inocente et al., 2014a, 2014b; Parmar et al., 2019a; Szakacs et al., 2015). One study found that 10% of adolescents with narcolepsy experienced suicidal ideation (Parmar et al., 2019a), and depression scores were found to be significantly higher in girls than boys (Inocente et al., 2014a). Participants with higher levels of depression also tended to experience a longer duration between the onset of their narcolepsy and receiving a diagnosis (Inocente et al., 2014a); this could be due to similarities in symptoms causing misdiagnosis or, alternatively, waiting for a diagnosis impacting psychological wellbeing. Further research is necessary to explore this construct and gain a better understand of this relationship. High rates of anxiety were also found in children and adolescents with narcolepsy; 48% of participants reported complaints of anxious symptomology in one study, (Hovi et al., 2022) and 10% met the clinical threshold for general anxiety disorder in another (Szakacs et al., 2015).

A study utilising a control group found that these elevated levels of psychological difficulties were significantly higher than healthy controls (Rocca et al., 2016). Several differences, however, were noted in the causation of psychological difficulties in this population. Stores et al. (2006) found that both of their narcolepsy group and parallel patient group with excessive daytime sleepiness had depression scores significantly higher than healthy controls, but found no significant difference between the two clinical groups. This suggests that the depressive symptomology was a result of excessive sleepiness rather than factors more specific to narcolepsy alone. This is supported by two studies that found depressive scores were positively correlated with excessive daytime sleepiness (Parmer et al., 2019a; Inocente et al., 2014a). Hovi et al. (2022), however, found that children with narcolepsy had overall higher psychological problems than Finnish children with other sleep disturbances, suggesting that this finding is more specific to narcolepsy and not the symptomology of general sleepiness. Finally, one study found higher levels of depression in children and adolescents with narcolepsy and comorbid ADHD compared with those without ADHD (Qu et al., 2022).

## **Social Functioning**

Eight studies exploring social functioning in children and adolescents with narcolepsy found difficulties in family relationships, education and social relationships.

### ***Family Relationships***

Having a child with a diagnosis of narcolepsy was associated with increased parental stress in two studies; parents of children with narcolepsy were found to have higher “total stress” scores than parents of children without narcolepsy (Szakacs et al., 2019). This was due to raised scores on the ‘parent-child dysfunctional interaction’ subscale and ‘difficult



child' subscale, suggesting that parents of children with narcolepsy have more difficult parent-child interactions, and view their child as more difficult to manage than parents of children without narcolepsy. The 'parental distress' subscale, however, was similar for both parents of children with narcolepsy and parents of children without narcolepsy, suggesting that these groups have similar levels of self-esteem in relation to their role as a parent (Szakacs et al., 2019). Similarly, Hovi et al. (2022) found that having a child with a diagnosis of narcolepsy was associated with increased parental and family stress, and that this was related to the level of emotional and behavioural challenges the child had. Overall family functioning was also found to be impaired in parents of adolescents with narcolepsy (Parmer et al., 2019b), and this impairment was found to be similar to parents of adolescents with chronic pain. Parents of adolescents with narcolepsy were also most impacted by 'worry' about their child's condition. Worry is common among caregivers of children with chronic conditions due to the unpredictable nature of their child's illness making it difficult to anticipate their child's health and future (Parmer et al., 2019b).

### ***Educational Difficulties***

Three studies revealed that children and adolescents with narcolepsy experience difficulties at school; one study found that this cohort received negative reports related to poor attention span, memory problems, concentration and falling asleep in class, (Guilleminault et al., 1998) and two studies found that children and adolescents with narcolepsy encountered a higher prevalence of educational challenges compared to controls (Inocente et al., 2014b; Stores et al., 2006). Among those with narcolepsy, 41% reported difficulties at school, whereas only 7.5% of controls faced similar issues. Additionally, within the narcolepsy group, 28% had to repeat a grade due to academic failure, while only 7.5% of the control group experienced the same outcome (Inocente et al., 2014b).

Differences were noted, however, in school absenteeism; one study found significantly higher levels of absenteeism in children and adolescents with narcolepsy than controls (Inocente et al., 2014b), whereas another found no significant group differences (Stores et al., 2006). Finally, adolescents with narcolepsy were found to have lower performances in school than children with narcolepsy, which is comparable to the cohort of children and adolescents without narcolepsy (Inocente et al., 2014b).

### ***Social Difficulties***

Social difficulties were found to be significantly higher in children and adolescents with narcolepsy than healthy controls in one study, who had more difficulties in social awareness, social cognition, social communication, social motivation and social preoccupation. Furthermore, 45.3% of children and adolescents with narcolepsy experienced mild-moderate social difficulties and 20.9% of children and adolescents with narcolepsy experienced severe difficulties (Quaedack et al., 2019). Finally, 90% of children in one study reported shame related to their diagnosis and expressed a desire to hide their illness from their peers at school (Guilleminault et al., 1998).

### **Quality of life**

Reduced quality of life in children and adolescents with narcolepsy was found in a collection on studies. Self-reported quality of life was found to be significantly diminished compared with healthy controls on both generic and narcolepsy specific measures of quality of life, with lower scores reported in all domains of the narcolepsy specific measure of quality of life (Szakacs et al., 2019). This cohort also expressed particular concerns about their future, demonstrating the need to support this population throughout childhood and

adolescence so that they are able to meet their future goals (Szakacs et al., 2019). Similarly, Inocente et al. (2014b) found that both children and adolescents with narcolepsy had lower self-reported health-related quality of life than controls.

Some differences were found between the child and adolescent cohorts, however, in regard to the domains most impacted; children with narcolepsy were found to have lower general wellbeing, poorer self-image and lower vitality than controls whereas adolescents with narcolepsy were found to have lower physical wellbeing, fewer friends and leisure activities than controls (Inocente et al., 2014b). Stores et al. (2006) found, however, that among children and adolescents with narcolepsy, only the mental health quality of life subscale exhibited a significant difference compared to control groups. Rocca et al. (2016) similarly discovered that children and adolescents with narcolepsy exhibit significantly lower scores in the psychosocial health domain, whilst the impact on the physical health domain was similar to controls. Overall, there is a consensus that quality of life in children and adolescents with narcolepsy is significantly diminished compared with healthy controls, however the specific domains most impacted is unclear; this may be due to the differences in measures used to assess quality of life across the studies.

The association between having a diagnosis of narcolepsy and a reduced quality of life in children and adolescents is complemented by parental reports. Parents of children and adolescents with narcolepsy reported their children as having overall worse quality of life than parents of children without narcolepsy in both the physical health and psychosocial health domains (Rocca et al., 2016). Parents were also found to have a good perception of their children's quality of life and gave similar ratings to their children across most of the domains (Inocente et al., 2014b).

No significant differences were found, however, in quality of life between children and adolescents with narcolepsy and the parallel patient control group with excessive daytime sleepiness (Stores et al., 2006). This suggests that reduced quality of life in this population may be a result of their excessive sleepiness rather than factors more specific to narcolepsy. Depression was also found to be a factor that was associated with diminished quality of life in children and adolescents with narcolepsy, demonstrating the interrelatedness of these constructs (Inocente et al., 2014b; Zhang et al., 2020). According to Inocente et al. (2014b), depression accounted for 49% of the variability in quality of life scores among children and adolescents with narcolepsy. Similarly, Zhang et al. (2020) found that depression explained 44% of the variability in quality of life scores for children and adolescents with narcolepsy. Finally, comorbid ADHD symptomology was also found to be associated with lower quality of life for this population and further impaired the quality of life of children and adolescents with narcolepsy (Qu et al., 2022).

### **Synthesis of Findings Summary**

Multiple studies examining psychological difficulties among children and adolescents with narcolepsy consistently concluded that this population experiences significant levels of depression and anxiety (Guilleminault et al., 1998; Hovi et al., 2022; Inocente et al., 2014a, 2014b; Parmar et al., 2019a; Szakacs et al., 2015). These elevated levels were also found to be significantly higher than healthy controls (Rocca et al., 2016). It is unclear, however, whether this is a result of excessive sleepiness symptomology, or whether another mechanism is at play (Parmar et al., 2019a; Inocente et al., 2014a; Hovi et al., 2022); further research is needed to clarify this.

Family relationships were impaired in both children and adolescents, with parents of children with narcolepsy experiencing increased parental and family stress (Hovi et al., 2022) and higher levels of stress than parents of children without narcolepsy (Szakacs et al., 2019). Similarly, overall family functioning was impaired in parents of adolescents with narcolepsy (Parmer et al., 2019b), and this impairment was found to be similar to parents of adolescents with chronic pain. Children and adolescents with narcolepsy also reported more difficulties at school compared to those without narcolepsy (Guilleminault et al., 1998; Inocente et al., 2014b; Stores et al., 2006) with school performance being worst in the adolescent cohort (Stores et al., 2006). Finally, social difficulties were higher in children and adolescents with narcolepsy than healthy controls (Quaedack et al., 2019), with this cohort expressing shame about their diagnosis and a desire to hide their illness from their peers at school (Guilleminault et al., 1998).

Overall, there is a consensus that quality of life in children and adolescents with narcolepsy is significantly diminished compared with healthy controls, however the specific domains most impacted is unclear. This may be due to the variation in measures used to assess quality of life across the studies and therefore further research using a consistent outcome measure is needed to clarify this (Szakacs et al., 2019; Inocente et al., 2014b; Stores et al., 2006; Rocca et al., 2016). This consensus is supported by parent reports (Rocca et al., 2016) who were also found to have a good perception of their children's quality of life (Inocente et al., 2014b). It seems, however, that reduced quality of life in this population may be a result of their excessive sleepiness symptomology rather than factors more specific to narcolepsy (Stores et al., 2006); further research is needed to ascertain whether this hypothesis holds true. Finally, higher levels of depression was significantly associated with poorer quality of life in this population (Inocente et al., 2014b; Zhang et al., 2020), and

comorbid ADHD symptomology was also related to an impaired quality of life of children and adolescents with narcolepsy (Qu et al., 2022).

## **Discussion**

To the author's knowledge, this is the first review to consider both the psychosocial functioning and quality of life in children and adolescents with narcolepsy. This review has demonstrated that whilst the research on psychosocial wellbeing and quality of life in children and adolescents with narcolepsy is relatively new, the majority of it has been judged to have an acceptable level of quality. It has also demonstrated that this population have marked psychological difficulties, social difficulties and a diminished quality of life. Consequently, there is a clear need for the development of preventative strategies following diagnosis, as well as psychological interventions targeting these psychological constructs, complemented by family and school interventions.

However, it is still unclear what mechanisms underlie and contribute to these findings; whilst some of the studies in this review suggest that narcolepsy symptomology is responsible for the reported distress (Stores et al., 2006; Parmer et al., 2019a; Inocente et al., 2014a) others suggest this might not be the case and that other mechanisms might be at play (Hovi et al., 2022). The wider research on the psychosocial outcomes and quality of life in chronic illness supports the latter findings, and suggests that symptomology alone is not enough to predict these outcomes, and instead draws on explanations such as Leventhal's Common Sense Model of Illness Representations (Leventhal et al., 1980). Future research should focus on exploring the possible predictors of psychosocial difficulties and quality of life for children and adolescents, such as symptom severity and illness representations, in

more depth. This will enable clinicians to better understand this populations' needs and tailor their interventions accordingly.

There were also a number of methodological limitations that were noted in this review; this has highlighted a range of areas future studies will need to consider when embarking on further research. Future research needs to use longitudinal research methods that utilises both parallel patient groups and healthy control groups. This will enhance the generalisability of findings and allow for the observation in any changes of these constructs across the different life stages of childhood, adolescence and adulthood, whilst enhancing the ability to establish causality and strengthen any conclusions drawn (Mueller-Peltzer et al., 2020). It is also essential that future quantitative research adopts a consistent outcome measure when investigating psychosocial difficulties and quality of life in children and adolescents with narcolepsy. By employing a standardised measure across studies, direct comparisons can be made, facilitating a better understanding of the impact of narcolepsy on these domains. Consistency in outcome measures enhances the reliability and validity of research findings, enabling researchers to identify common patterns and factors that contribute to psychosocial difficulties and quality of life in this population.

To gain a more comprehensive understanding of the psychosocial wellbeing and quality of life for this population, it is essential to conduct qualitative research. This approach would provide a richer description and a more detailed picture of their experiences and perspectives. By employing qualitative methods, researchers can explore the subjective aspects of psychosocial wellbeing and quality of life, capturing the nuances and complexities that quantitative measures may not fully capture. Through techniques such as interviews, focus groups, and observations, qualitative research could offer valuable insights into the

unique challenges and factors influencing the psychosocial wellbeing and quality of life of individuals in children and adolescents with narcolepsy (Shelton et al., 2022).

Finally, in order to obtain a comprehensive understanding of psychosocial difficulties and quality of life in children and adolescents with narcolepsy, it is crucial for researchers to collect data from a wide range of sources using self-report, parent-report, and school-report methods. Self-reports allow individuals with narcolepsy to directly express their thoughts and feelings, while parent-reports offer insights from the perspective of caregivers who observe their child's experiences. Additionally, school-reports provide valuable information on the child's functioning and interactions within the educational environment. By employing these diverse methods, a broader perspective can be obtained, and a more accurate measurement of their experiences can be achieved. This will help to minimize potential sources of bias that may arise from relying solely on one type of report method (Fisher et al., 2014).

Whilst a clear strength of this review is that it closely followed PRISMA guidelines to reduce bias and ensure the search strategy captured all the relevant published literature, it is important to recognise the limitations of the review as well. The quality assessment tools used in this review required the researcher to make subjective conclusions about each study's quality, and therefore different conclusions may have been drawn by other researchers (Linde et al., 2003). Future researchers conducting similar reviews may wish to utilise multiple researchers when making quality assessments to increase reliability and reduce bias.

Another limitation to this review is that it collated 'children and adolescents' into one group due to the limited literature available for this population. There is a great difference in individuals' physical, cognitive and emotional development throughout the various stages of



childhood and adolescence (Field et al., 2003), and consequently there may be differences in the psychosocial wellbeing and quality of life of children with narcolepsy compared with adolescents with narcolepsy. As the body of literature increases for this population, future researchers may wish to conduct separate reviews for children and adolescents respectively.

## **Conclusion**

In conclusion, this review has demonstrated that the majority of research on psychosocial wellbeing and quality of life in children and adolescents with narcolepsy is of good quality. It has also demonstrated that this population experiences marked psychosocial difficulties and has a diminished quality of life. This highlights the need for the formulation of preventive strategies subsequent to diagnosis alongside psychological interventions that target these constructs. Future research should focus on what mechanisms may be underlying these findings, whilst considering the methodological recommendations highlighted in this review.

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Alexandra Burdass BSc (Hons) MSc

Major Research Project

Section B: Empirical Study

An Investigation into the Relationship between Illness Perceptions  
and Psychosocial Outcomes, and the Mediating Role of Coping in  
Young Adults with Narcolepsy

Word Count: 7476 (597)

SALOMONS INSTITUTE FOR APPLIED PSYCHOLOGY

CANTERBURY CHRIST CHURCH UNIVERSITY

## **Abstract**

**Objective:** The aim of this study was to explore illness perceptions, coping strategies and psychosocial outcomes in young adults with narcolepsy, and the potential relationships between these variables.

**Method:** 54 young with narcolepsy adults aged 16-25 took part in this cross-sectional study. They completed quantitative and qualitative questionnaires online assessing their illness perceptions, coping strategies, quality of life and symptoms of anxiety and depression.

**Results:** Descriptive statistics showed that this population experience high levels of perceived threat in relation to their condition, poorer physical and mental quality of life than the general young adult population, and experience moderate levels of anxiety and depression. They also engage in predominantly problem-focused coping strategies and find sleeping, napping and taking medication the most helpful ways to cope with their condition. Regression analyses showed that higher levels of illness threat perception were associated lower physical and mental quality of life. Coping did not mediate the relationship between illness perceptions and psychosocial outcomes.

**Conclusions:** This study has made an important contribution to the limited literature on young adults with narcolepsy. The implications of findings for research and clinical practice with this population are considered.

**Keywords:** *narcolepsy, illness perceptions, coping, psychosocial wellbeing, quality of life*

## **Introduction**

### **The Impact of Narcolepsy**

Narcolepsy is a rare, chronic, neurological condition characterised by excessive daytime sleepiness and a disruption in the sleep-wake cycle. Individuals with narcolepsy may experience sudden and uncontrollable sleep attacks, where they fall asleep involuntarily and unexpectedly. Other symptoms of narcolepsy can include cataplexy (sudden loss of muscle tone), sleep paralysis (temporary inability to move or speak upon waking or falling asleep), and vivid hallucinations during sleep (Barateau et al., 2022). These symptoms can significantly impact daily life, and individuals with narcolepsy often need to make lifestyle adjustments to manage their symptoms effectively as there is no specific cure. This may include implementing regular napping schedules, making accommodations to ensure productivity and safety in daily activities and enhancing wakefulness through medications (Bhattarai et al., 2017). Whilst the onset of narcolepsy symptoms typically occurs during adolescence or early adulthood, it can also develop in children and older individuals. Diagnosis, however, is commonly delayed due to lack of awareness and misinterpretation of symptoms (Zhang et al., 2020).

### **Narcolepsy and Psychosocial Outcomes**

Studies have demonstrated how living with narcolepsy can significantly impact psychosocial functioning, resulting in lower quality of life, and a higher prevalence of mood and anxiety disorders compared with the general population (Barker et al., 2020). These studies, however, have predominantly focused on middle aged and older adults, and more recently children, resulting in a lack of research into the psychosocial functioning of young adults with narcolepsy (Kapella, 2015). Initial findings do suggest, however, that young



adults with narcolepsy experience lower mood and quality of life than young adults without narcolepsy (Kapella, 2015). Young adulthood is a critical period of development where individuals face a multitude of challenges such as the development of their identity, leaving home, entering further education or work, exploring their sexuality, finding a partner or starting a family (Bonnie et al., 2015). Having a chronic illness can significantly disrupt these developmental milestones and act as a barrier to living a 'normal' life (Wilson et al., 2021). Research suggests that young adults who do not meet these developmental milestones are at risk of struggling to adapt to future adult roles (Sansom-Daly et al., 2012). It is therefore essential that services understand the needs of individuals in this phase of life so that effective interventions can be tailored appropriately.

### **The Common Sense Model of Illness Representation**

Whilst the literature documents the psychosocial impact of narcolepsy, the underlying mechanisms contributing to this have not been fully defined (Kapella, 2015). The causes of psychosocial problems in this population have largely been attributed to the primary symptoms of narcolepsy such as sleepiness, with studies using symptom severity as a predictor for quality of life, depression and anxiety (Shang et al., 2021). However, research demonstrates that symptom severity is often insufficient to fully explain the adverse effects on functioning in individuals with chronic conditions, and discrepancies have been found between the underlying pathology of chronic conditions and the level of illness related dysfunction in the mental and social domains (Verhoof et al., 2014). This has resulted in an exploration into how psychosocial correlates may contribute to the individual differences in adjustment and consequential outcomes in people with chronic illness, including the application of Leventhal's Common Sense Model of Illness Representations (Leventhal et al., 1980; Leventhal et al., 1984).

### ***Illness Perceptions***

This model, also known as the Common Sense Model, suggests that as people with chronic conditions obtain new information about their condition, they develop beliefs about their illness, known as ‘illness perceptions’ (Leventhal et al., 1980). Illness perception is a personal understanding of an individual’s health condition across different dimensions, such as the perceived severity, expected duration, perception of symptoms, and perceived control over the illness and its symptoms (Lee et al., 2023). These perceptions can be used to assess how threatening an illness is perceived by an individual, known as threat-related illness representation (Cannon et al., 2022). To the author’s knowledge, there have been no studies to date that explore the illness perceptions of individuals with narcolepsy.

### ***Coping***

The Common Sense Model suggests that these illness perceptions influence the types of coping strategies a person uses to manage their condition and the perceived threat and stress resulting from it. Whilst the model does not explicitly outline specific types of coping, coping dimensions such as problem-focused coping, emotion-focused coping and avoidant coping and have been derived from theoretical frameworks (Lazarus et al., 1984; Endler et al., 1990). Problem-focused coping is characterised by being goal-oriented and involves cognitive efforts aimed at regulating stress. This coping style is more likely to lead to positive outcomes, particularly in people with chronic conditions (Kristofferzon et al., 2018), because it empowers individuals with a sense of control in the process of problem-solving. Examples include use of informational support, positive reframing and planning. Emotion-focused coping is characterised by the management of emotions and regulation of emotional distress. Examples include the use of humour, acceptance, self-blame and emotional support. These strategies may be adaptive in certain situations, such as when a stressor cannot be changed

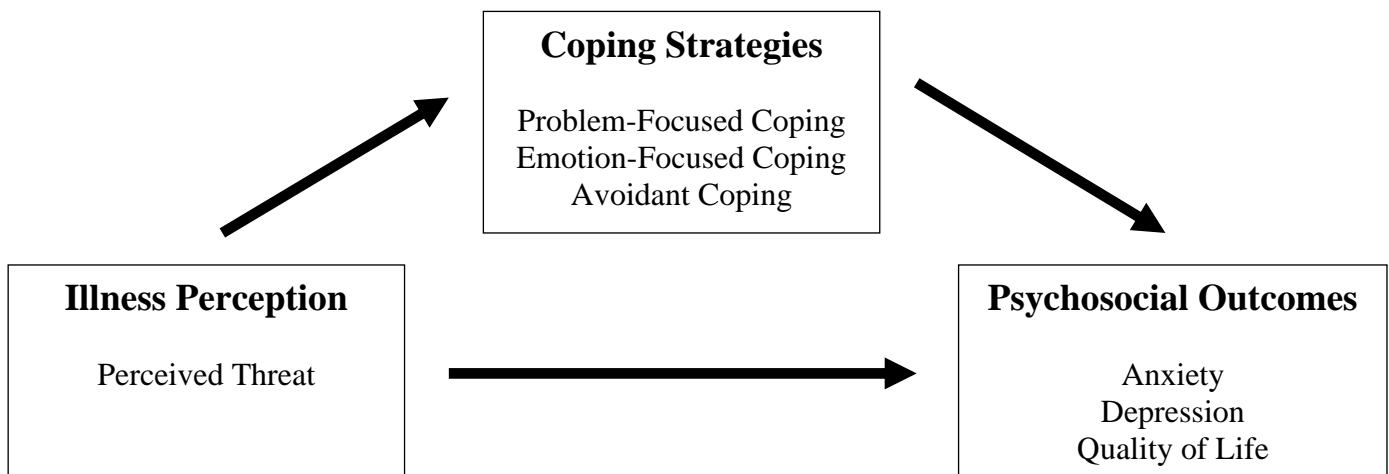
directly, but can be less effective in the long term. In people with chronic conditions, however, emotion focused coping was found to be associated with poorer psychosocial outcomes (Kristofferzon et al., 2018). Finally, avoidant coping involves efforts to redirect attention away from the stressor and is likely to lead to poorer outcomes in people with chronic condition (Eisenberg et al., 2012). Examples include denial, self-distraction and substance use (Green et al., 2010). It is important to note that these coping strategies have been predominantly explored using a single research method in previous research on chronic conditions; using a mixed methods approach can provide a richer and more holistic view of this complex construct, and is demonstrated and recommended by some researchers (Sharour et al., 2020). Furthermore, to the author's knowledge, there have been no studies to date that explore the different coping strategies used by individuals with narcolepsy.

### ***Psychosocial Outcomes***

The Common Sense Model suggests that these different coping strategies, can, in turn, lead to positive or negative outcomes such as anxiety, depression or low quality of life. For example, threat related illness perceptions and avoidant coping styles have been found to be strong predictors of psychological distress (Dempster et al., 2015). The Common Sense Model (Figure 1) is, therefore, a mediation model where coping strategies mediate the influence of a person's illness perceptions on their wellbeing and outcomes (Hagger et al., 2003).

**Figure 1**

*Leventhal's Common Sense Model of Illness Representations (Leventhal et al., 1980)*



A body of research has demonstrated how this model can be applied to a range of chronic conditions, and how illness perceptions and coping are strongly related to health outcomes including anxiety, depression and quality of life (Cheng et al., 2020; Gurkova et al., 2018 ). Furthermore, these findings have been demonstrated in young adults with chronic conditions (Limperg et al., 2020; Stapersma et al., 2019; Verhoof et al., 2014). No research to date, however, has explored this model and its mechanisms in individuals living with narcolepsy.

### **Rationale**

In conclusion there is need for more research on the psychosocial functioning of young adults with narcolepsy, such as the prevalence rates of anxiety, depression and quality of life in this population. There is also a need for research into the patterns of illness perceptions in the narcolepsy population. Coping strategies also need to be explored, both qualitatively and quantitatively to provide a rich understanding of this concept. Finally, an

exploration guided by the Common Sense Model (Leventhal et al., 1980) into the relationship between illness perceptions, coping strategies and psychosocial outcomes such as anxiety, depression and quality of life is needed. If it is found that the Common Sense Model applies to young adults with narcolepsy, it could shed insight and guidance into what interventions might be useful for improving psychosocial wellbeing in this population, such as targeting illness perceptions or coping strategies, resulting in the development of more specific and tailored interventions for this population. Consequently, this study is grounded in the NHS value “Improving Lives”. This study is also the first of the author’s knowledge to investigate these variables in young adults with narcolepsy and apply the Common Sense Model of Illness Representation to this population.

## **Research Questions**

This study aimed to answer the following research questions:

- 1) What is the psychosocial functioning of young adults with narcolepsy, as measured by symptoms of anxiety, depression, and quality of life?
- 2) What are the patterns of illness perception in young adults with narcolepsy, as measured by perceived threat?
- 3) What types of coping strategies do young adults with narcolepsy use?
- 4) Does coping mediate the relationship between illness perception and psychosocial outcomes in young adults with narcolepsy?
  - a. Do coping styles mediate the relationship between illness perception and physical quality of life?
  - b. Do coping styles mediate the relationship between illness perception and mental quality of life?

- c. Do coping styles mediate the relationship between illness perception and anxiety?
- d. Do coping styles mediate the relationship between illness perception and depression?

### **Hypotheses**

- 1) It is hypothesised that young adults with narcolepsy will have poor psychosocial functioning, as measured by symptoms of anxiety, depression and quality of life.
- 2) It is hypothesised that higher levels of illness threat perception will predict poorer psychosocial outcomes
- 3) It is hypothesised that avoidant coping will positively mediate the relationship between illness threat perception and psychosocial outcomes.
- 4) It is hypothesised that emotion-focused coping will positively mediate the relationship between illness threat perception and psychosocial outcomes.
- 5) It is hypothesised that problem-focused coping will negatively mediate the relationship between illness threat perception and psychosocial outcomes.

## **Method**

### **Design**

This study used a mixed method, cross-sectional design. For the quantitative analysis there was one predictor variable (illness threat perception), three mediator variables (problem-focused coping, emotion-focussed coping and avoidant coping) and four outcome variables (physical quality of life, mental quality of life, anxiety and depression). As the study was cross-sectional and used regression analyses to examine the level of association between variables, there was no intention to determine causality. Coping was also assessed qualitatively using free text questions and a content analysis approach.

### **Participants**

Participants were recruited online via advertisements on the research team's Twitter and Reddit accounts and through Narcolepsy UK's online platforms. The following inclusion criteria were applied:

- Participants were required to have a diagnosis of narcolepsy
- Participants were required to be young adults between the ages of 16-25 years old
- Participants had to be able to read and understand English

62 participants provided consent for the study. 8 participants did not continue beyond this point and therefore data was collected from the remaining 54 participants (see Table 1 for demographics). 10 of these participants were excluded from the regression analyses due to ending the study early and leaving incomplete responses on at least one of the measures. 44 participants finished the study and completed all of the measures. Participants were also asked to detail how they received their diagnosis; 51 participants received their diagnosis from clinicians, 3 participants were self-diagnosed and 1 participant did not detail how they

received their diagnosis. Due to the difficulties documented in the narcolepsy literature regarding under-diagnosis and delayed diagnosis (Dunne et al., 2016), combined with the small number of participants recruited to the study, participants were not excluded from the analysis if they did not report receiving a diagnosis from a clinician. However, caution should be taken when drawing conclusions from any findings due to the potential impact on the study's generalisability. Descriptive statistics for individuals with both complete and incomplete data sets were included to observe any differences in the sample characteristics. These can be seen in Table 2.



**Table 1***Participant Demographics*

		<b>Overall Sample (N=54)</b>		<b>Complete Measure (N=44)</b>	
		<b>N</b>	<b>%</b>	<b>N</b>	<b>%</b>
Gender	Male	20	37.0	17	38.6
	Female	28	51.9	23	52.3
	Non-binary/ third gender	5	9.3	4	9.1
Ethnicity	White	43	79.6	36	81.8
	Black	2	3.7	2	4.5
	Asian	3	5.6	2	4.5
	Multiple Ethnic Groups	6	11.1	4	9.1
Nationality	English	27	50	19	43.2
	Welsh	1	1.9	1	2.3
	Scottish	3	5.6	3	6.8
	Irish	1	1.9	0	0
	American	16	29.8	15	34.1
	Canadian	2	3.8	1	2.3
	Indian	1	1.9	1	2.3
	Finish	1	1.9	1	2.3
	Australian	2	3.7	2	4.5
Narcolepsy Type	Type 1	34	63	27	61.4
	Type 2	20	37	16	38.6
Diagnosis Type	Clinician Diagnosis	50	92.6	42	95.5
	Self-Diagnosis	3	5.6	2	4.5
	Missing	1	1.9	0	0
		<b>Mean (SD)</b>	<b>Range</b>	<b>Mean (SD)</b>	<b>Range</b>
Age		21.66 (2.56)	16-25	21.84 (2.51)	16-25
Age at Diagnosis		16.47 (5.80)	1-25	16.29 5.58	1-25

**Measures**

All participants completed a demographic form (Appendix A) assessing information such as age, gender, and type of narcolepsy. The six study variables being investigated were measured by five self-report questionnaires.

### ***Illness Perceptions***

Illness Perceptions was assessed using the Brief Illness Perceptions Questionnaire (Broadbent et al., 2006; Appendix B). This is an eight item scale that assesses the cognitive and emotional representations a person has of their illness including Consequences, Timeline, Personal Control, Treatment Control, Identity, Coherence, Concern and Emotional Response (Broadbent et al., 2006). Items are rated using a 0-10 response scale and an overall score which represents the degree to which the illness is perceived as threatening can be calculated (Broadbent et al., 2006). Higher scores indicate higher experienced threat. The Brief Illness Perceptions Questionnaire has been shown to be a reliable and valid measure of illness perceptions in a range of illness populations, and has been used in research investigating the relationship between illness perceptions and psychosocial outcomes in young adult populations (Verhoof et al., 2014).

### ***Coping***

**Quantitative Measure.** Coping was assessed using the Brief-COPE (Carver, 1997; Appendix C). This is a 28 item scale that measures the use of effective and ineffective coping strategies in response to stressors such as chronic illnesses. Items are rated on a 1-4 response scale from “I haven’t been doing this at all” to “I’ve been doing this a lot”. The scale can determine an individual’s primary coping style using the subscales Problem-Focused Coping, Emotion-Focused Coping and Avoidant Coping. High scores on Problem-Focused Coping indicates strategies that aim to change the stressful situation such as planning and informational support, high scores on Emotion-Focused Coping indicates strategies that aim to regulate emotions with the stressful situation such as humour and acceptance, and high scores on Avoidant Coping indicates strategies that aim to disengage from the stressor such as denial and substance use (Carver, 1997). The Brief-COPE is a well validated measure that has been

used to assess coping strategies in a range of chronic conditions. This well validated measure is also frequently used in studies applying the Common Sense Model of Illness (Hagger et al., 2017).

**Qualitative Measure.** Coping strategies were qualitatively assessed using free text questions based on the existing literature on coping with chronic conditions (Green et al., 2011). These questions included “What are some of the different ways you cope with your narcolepsy? - Are there any coping strategies you use that you find particularly useful or helpful? Are there any coping strategies you use that you find more unhelpful?” (Appendix D).

### *Quality of Life*

Quality of Life was assessed using the Short Form Health Survey (Ware et al., 1992; Appendix E). This 36 item scale includes 36 questions related to an individual’s quality of life and generates two summary scales: a Physical Component Summary (PCS) and a Mental Component Summary (MCS). Scores range from 1 to 100, with higher scores indicating better health status (Bogan et al., 2016) and a mean score of 50 is used as a normative value for both scales. This is a well validated measure that has been utilised in a range of chronic conditions, including narcolepsy (Tadrous et al., 2021).

### *Anxiety and Depression*

Anxiety and depression was assessed using the Hospital Anxiety and Depression Scale (Stern, 2014; Appendix F). This is a 14 item measure comprising of 7 questions for anxiety and 7 questions for depression. Scores can range from 0-21 with high scores reflect greater anxiety or depression (normal = 0-7; mild = 8-10; moderate = 11-14; severe = 15-21).

This is a well-validated measure and been previously used in the narcolepsy population (Kapella et al., 2015).

## **Procedure**

An online study page was created using Qualtrics, containing the various study materials such as the information sheet (Appendix G), consent form (Appendix H), questionnaires and debrief sheet (Appendix I). A narcolepsy team in an NHS hospital initially agreed to advertise the study to their patients in clinic by sending the study advert (Appendix J) in pre-paid envelopes. However, due to change in staff, this arrangement changed and recruitment via this hospital was not possible. The study was subsequently advertised online by Narcolepsy UK, as well as on the research team's Twitter and Reddit accounts. Interested participants were invited to access the study through a computer or mobile device by scanning the QR code or clicking on the study's provided link on the study advert, which directed them to the Qualtrics study page. Participants were required to provide their consent prior to completing the demographic information and the six study questionnaires which were subsequently presented onscreen. The study took approximately 30-35 minutes to complete, and regular reminders to take breaks were incorporated throughout the questionnaires. At the end of the study, participants were provided with information on accessing support if needed (Appendix I). Participants were also given the option of opening a second Qualtrics link to enter a £50 Amazon voucher prize draw, or hear about the results of the study by email. They were informed that their questionnaire data was pseudo-anonymised, and that questionnaire data and personal information would be kept on separate encrypted folders to ensure anonymity.

## **Ethics**

The study was approved by the NHS Research Ethics Committee (Ref no. 308847; Appendix K). All participants provided informed consent. Participant wellbeing was ensured by providing a number and email address to contact if they felt distressed whilst completing the survey. On contacting this number the research team would be able to signpost to services that might be useful for providing further support. The contact details of Narcolepsy UK were also provided for further general advice and support for narcolepsy. Participants were informed, however, that responses were not reviewed immediately by the study team, and that they should actively contact the team if they felt distressed.

## **Service User Involvement**

A young person with narcolepsy acted as a service user consultant for the project, and helped to inform the study's design, the recruitment procedures and the management of the study.

## **Data Analysis Strategy**

### *Descriptive and Correlation Analyses*

The Statistics Package for the Social Sciences (SPSS, Version 24) was used to perform the quantitative analyses. Descriptive statistics explored the sample's demographics and their scores on the different variables. This analysis was used to answer the first three research questions:

- 1) What is the psychosocial functioning of young adults with narcolepsy in this sample, as measured by symptoms of anxiety, depression, and health related quality of life?
- 2) What are the patterns of illness representations of young adults with narcolepsy, as measured by perceived threat?

### 3) What types of coping strategies do young adults with narcolepsy use?

The descriptive statistics also tested the measures' internal reliability using Cronbach's alpha. Pearson correlation analyses explored the relationships between illness threat perception, problem-focused coping, emotion-focused coping, avoidant coping, physical quality of life, mental quality of life, anxiety and depression.

#### *Mediation Analyses*

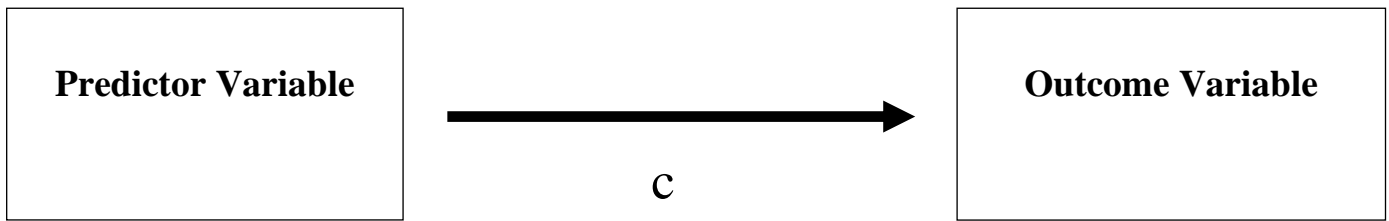
Hayes' (2018) bootstrapping approach to mediation analysis was implemented through the PROCESS macro for SPSS. This statistical method is used to assess and estimate the indirect effect of an independent variable on a dependent variable through one or more mediators. Bootstrapping involves generating multiple resamples of the data and calculating the indirect effect for each resample. Hayes' bootstrapping approach was chosen for this study because it enhances the reliability of mediation analysis by addressing common limitations of traditional methods. For example, it takes into account potential skewness, kurtosis, and bias in the distribution of the indirect effect, providing more reliable estimations of the indirect effect, particularly in cases where the sample size is small (Hayes, 2018). Five thousand bootstrap samples were generated to estimate the indirect effects and bias corrected 95% bootstrapped confidence intervals were calculated for these indirect effects. A total effect (path c) refers to the relationship between a predictor variable and an outcome variable without considering any potential mediator variables. Mediation refers to when there is a total indirect effect (path ab) of the predictor variable on the outcome variable through one or more mediators (see Figure 2). Whilst using Hayes' bootstrapping approach to mediation analysis is particularly useful when dealing with small sample sizes, Fritz et al., (2007) recommends a sample size of 71 to achieve an acceptable power of 80% and detect an

indirect effect comprising of medium sized ‘a’ and ‘b’ pathways on Bias-corrected bootstrapping. Therefore any findings will be interpreted with caution.

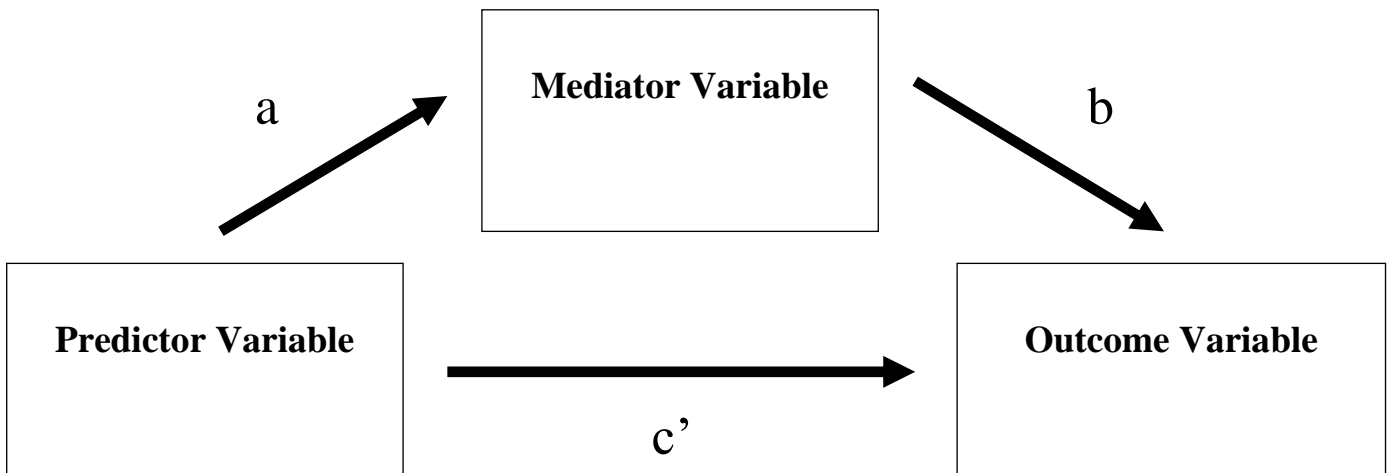
**Figure 2**

*Simple Mediation Model (Hayes, 2008)*

Direct Pathway (Total Effect):



Mediated Pathway (Indirect Effect):



Several parallel mediation models were performed to provide insights into the direct effects of threat perception on each outcome, as well as the potential mediating roles of problem-focused coping, emotion-focused coping, and avoidant coping and answer the research question “Does coping mediate the relationship between illness perceptions and psychosocial outcomes in young adults with narcolepsy?”. To answer research question 4a illness threat perception was entered as the predictor, problem-focused coping, emotion-

focused coping and avoidant coping were the mediators and physical quality of life was entered as the outcome. To answer research question 4b illness threat perception was entered as the predictor, problem-focused coping, emotion-focused coping and avoidant coping were the mediators and mental quality of life was entered as the outcome. To answer research question 4c illness threat perception was entered as the predictor, problem-focused coping, emotion-focused coping and avoidant coping were the mediators and anxiety was entered as the outcome. To answer research question 4d illness threat perception was entered as the predictor, problem-focused coping, emotion-focused coping and avoidant coping were the mediators and depression was entered as the outcome.

### *Qualitative analysis*

Qualitative content analysis was employed to analyse the free text coping questions using a systematic coding process and address the research question “What types of coping strategies do young adults with narcolepsy use?” using the methodology outlined by Bengtsson’s (2016). This approach was chosen in order to broadly ascertain patterns in the short-text data, and focused on developing themes and patterns inductively, without preconceived notions. This allowed for immersion in the data and the emergence of novel insights into the different ways young adults cope with narcolepsy (Al-Yateem et al., 2020). The data collected was divided into two main parts based upon the questions asked: “Coping strategies experienced as helpful” and “Coping strategies experienced as unhelpful”. Potential codes were generated whilst reading the data before developing a coding scheme. Codes were then organised into categories based on meaningful relationships and connections between different codes. The frequency of codes in each category was calculated to understand the most commonly reported coping strategies. A complete list of codes and quotes can be found in Appendix L. The data was then coded by a second member of the research team (Appendix



M) to circumvent any bias; inter-coder percentage agreement was calculated to be 88%, which is an acceptable level of agreement (O'Connor et al., 2020).

## Results

### Quantitative Analysis

#### *Descriptive Statistics*

There were a number of participants who stopped the study early and did not complete all the measures. Descriptive statistics were performed on the overall sample, the sample of participants who completed all measures, and the sample of participants who did not complete all measures. Table 2 show the means, standard deviations (SD) and Cronbach's alphas for the variables in each category.

**Table 2**

#### *Descriptive Statistics*

	<b>Complete measures (n=44) Mean (SD)</b>	<b>Incomplete measures (n=10) Mean (SD)</b>	<b>Overall sample (n=54) Mean (SD)</b>	<b>Cronbach's alpha</b>
Illness Threat Perception	49.75 (10.40)	48.10 (11.11)	49.44 (10.44)	.84
Problem-focused Coping	2.73 (.51)	2.96 (.68)	2.77 (.54)	.73
Emotion-focused Coping	2.42 (.35)	2.46 (.49)	2.42 (.37)	.50
Avoidant Coping	2.02 (.53)	1.71 (.50)	1.97 (.53)	.70
QoL: Physical Component	49.88 (16.91)	55.24 (23.15)	50.71 (17.85)	.70
QoL: Mental Component	42.03 (18.06)	49.01 (22.98)	43.10 (18.82)	.70
Anxiety	10.48 (3.55)	Not available	10.48 (3.55)	.86
Depression	9.36 (3.48)	Not available	9.36 (3.48)	.77

Both the complete and incomplete measures samples had similar illness threat perception scores that were just below the cut off for high experienced threat ( $\geq 50$ ; Kuiper et al., 2022) suggesting that young adults with narcolepsy experience high levels of threat in relation to their condition.

Both the complete and incomplete measures samples had slightly higher problem-focused coping scores than the normative sample (2.67) and slightly higher emotion-focused coping scores than the normative sample (2.34). The complete measures sample had a slightly higher avoidant coping score than the normative sample (1.97), whereas the incomplete measures had a slightly lower avoidant coping score than the normative sample (Buchanan, 2021). This suggests that young adults with narcolepsy engage in higher levels of problem-focused coping strategies and lower emotion-focused coping strategies to the general population.

Both samples had lower scores on the mental quality of life component than the physical quality of life component which is in line with previous research on adults with narcolepsy suggesting that narcolepsy has a more significant impact on their mental wellbeing than their physical wellbeing (Tadrous et al., 2021). Both the overall sample and the complete measures sample had lower physical and mental quality of life scores than the normative sample for young adults (53, 49), however, the incomplete measures sample had a higher physical quality of life and similar mental quality of life score to the normative sample for young adults (Ware et al., 2001).

The complete measures sample had ‘moderate’ levels of anxiety and just below ‘moderate’ levels of depression. The participants in the incomplete sample did not complete

the Hospital Anxiety and Depression Scale due to it being the last questionnaire, and therefore a mean is not available for this group.

Overall, the individuals who completed all measures and were included in the regression analyses experienced high levels of threat in relation to their narcolepsy, engaged in higher problem-focused coping strategies, lower emotion-focused coping strategies and higher avoidant coping strategies. They also had lower physical and mental quality of life scores in comparison to young adults in the general population, moderate levels of anxiety and mild/moderate levels of depression. All subsequent analysis was done with the data from this population. Caution should be taken when generalising any conclusions made from the analyses due to the slight differences in variable scores between the complete and incomplete sample.

The Cronbach alpha's for all variables were above the minimum acceptable value (0.70; Bland & Altman, 1997) with the exception of emotion-focused coping (.50). This indicated high internal consistency across the majority of measures. The variable emotion-focused coping was retained in the analysis to provide a more comprehensive view of the data, but should be viewed with caution due to its lower alpha value (Field, 2005).

## **Correlation**

Some of the data was not normally distributed. Therefore two tailed Bootstrapped Pearson's correlation analyses were conducted to explore the relationships between the study's variables. This method is a more robust way of conducting correlation analyses when the underlying assumptions of Pearson's correlation may not hold, such as normality (Hutson et al., 2019). The variables' correlation coefficients can be found in Table 3.

**Table 3***Bootstrapped Pearson's Correlations of Study Variables*

	<b>Illness Threat Perception</b>	<b>Problem- focused Coping</b>	<b>Emotion- focused Coping</b>	<b>Avoidant Coping</b>	<b>Physical QoL</b>	<b>Mental QoL</b>	<b>Anxiety</b>
Problem- focused Coping	.28						
Emotion- focused Coping	.07	.59**					
Avoidant Coping	-.26	-.12	-.22				
Physical QoL	-.48**	.09	-.07	-.12			
Mental QoL	-.75**	-.28	-.28	.24	.58**		
Anxiety	.14	-.03	-.03	.26	-.52**	-.12	
Depression	.04	-.40**	-.21	.25	-.54**	-.13	.46**

*Note:* \*\*  $p < 0.01$ 

Significant relationships were found between the following variables. A moderate negative relationship was observed between illness threat perception and physical quality of life, and a strong negative relationship was observed between illness threat perception and mental quality of life. A strong positive relationship was observed between problem-focused coping and emotion-focused coping, and a moderate negative relationship was observed between problem-focused coping and depression. A strong positive relationship was observed between physical quality of life and mental quality of life and strong negative relationships were observed between physical quality of life and both anxiety and depression. Finally, a moderate positive relationship was observed between anxiety and depression.

## Mediation

### *The relationship Between Illness Threat Perception and Physical Quality of Life, Mediated by Coping Styles*

The first parallel mediation analysis explored the relationship between illness threat perception and physical quality of life, mediated by problem-focused coping, emotion-focused coping, and avoidant coping. There was a significant total effect of illness threat perception on physical quality of life ( $c = -.78, p < .001$ ), indicating that higher levels of threat perception were associated with lower physical quality of life. There was also a significant direct effect of illness threat perception on physical quality of life ( $c' = -1.00, p < .001$ ), suggesting that illness threat perception has a direct impact on physical quality of life, independent of the mediators. The indirect effects through problem-focused coping ( $a^1b^1 = .16, BCI [-.04, .50]$ ), emotion-focused coping ( $a^2b^2 = -.02, BCI [-.22, .10]$ ), and avoidant coping ( $a^3b^3 = .08, BCI [-.04, .26]$ ) were non-significant because all confidence intervals included zero, indicating a lack of mediation. Therefore coping styles did not mediate the relationship between illness threat perception and physical quality of life. These results can be seen in Table 4.

**Table 4**

*Mediation Analysis Results for the Relationship Between Illness Threat Perception and Physical Quality of Life, Mediated by Coping Styles*

<b>Total Effect</b>	<b>Direct Effect</b>	<b>Relationship</b>	<b>Indirect Effect</b>	<b>Boot LLCI</b>	<b>Boot ULCI</b>
<i>Threat perception</i> → <i>Physical QoL</i>	<i>Threat perception</i> → <i>Physical QoL</i>				
- .78 (.001)	-1.00 (.001)				
		Threat perception → Problem-focused coping → Physical QoL	.16	-.04	.50
		Threat perception → Emotion-focused coping → Physical QoL	-.02	-.22	.10
		Threat perception → Avoidant coping → Physical QoL	.08	-.04	.26

*Note:* Boot LLCI represents the Lower Limit of Confidence Interval, and Boot ULCI represents the Upper Limit of Confidence Interval. Confidence intervals that include zero indicate non-significant mediation effects.

***The relationship Between Illness Threat Perception and Mental Quality of Life, Mediated by Coping Styles***

The second mediation analysis explored the relationship between illness threat perception and mental quality of life, mediated by problem-focused coping, emotion-focused coping, and avoidant coping. There was a significant total effect of illness threat perception on mental quality of life ( $c = -1.30, p < .001$ ), indicating that higher levels of illness threat perception were associated with lower mental physical quality of life. There was also a significant direct effect of illness threat perception on mental quality of life ( $c' = -1.26, p < .001$ ), suggesting that illness threat perception has a direct impact on mental quality of life,

independent of the mediators. The indirect effects through problem-focused coping ( $a^1b^1 = .06$ , BCI [-.09, .23]), emotion-focused coping ( $a^2b^2 = -.05$ , BCI [-.26, .13]), and avoidant coping ( $a^3b^3 = -.06$ , BCI [-.23, .04]) were non-significant because all confidence intervals included zero, indicating a lack of mediation. Therefore, coping styles did not mediate the relationship between illness threat perception and mental quality of life. These results can be seen in Table 5.

**Table 5**

*Mediation Analysis Results for the Relationship Between Illness Threat Perception and Mental Quality of Life, Mediated by Coping Styles*

<b>Total Effect</b>	<b>Direct Effect</b>	<b>Relationship</b>	<b>Indirect Effect</b>	<b>Boot LLCI</b>	<b>Boot ULCI</b>
<i>Threat perception → Mental QoL</i>	<i>Threat perception → Mental QoL</i>				
-1.30 (.001)	-1.26 (.001)	Threat perception → Problem-focused coping → Mental QoL	.06	-.09	.23
		Threat perception → Emotion-focused coping → Mental QoL	-.05	-.26	.13
		Threat perception → Avoidant coping → Mental QoL	-.06	-.23	.04

*Note:* Boot LLCI represents the Lower Limit of Confidence Interval, and Boot ULCI represents the Upper Limit of Confidence Interval. Confidence intervals that include zero indicate non-significant mediation effects.

### *The relationship Between Illness Threat Perception and Anxiety, Mediated by Coping Styles*

The third mediation analysis explored the relationship between illness threat perception and anxiety, mediated by problem-focused coping, emotion-focused coping, and avoidant coping. The total effect of illness threat perception on anxiety was non-significant ( $c = -.05, p = .36$ ), indicating that threat perception was not significantly associated with anxiety. Similarly, the direct effect of threat perception on anxiety was also non-significant ( $c' = .08, p = .14$ ), suggesting no direct impact of threat perception on anxiety, independent of the mediators. The indirect effects through problem-focused coping ( $a^1b^1 = -.01, \text{BCI} [-.07, .05]$ ), emotion-focused coping ( $a^2b^2 = -.01, \text{BCI} [-.03, .03]$ ), and avoidant coping ( $a^3b^3 = -.03, 95\% \text{BCI} [-.09, .01]$ ) were non-significant because all confidence intervals included zero, indicating a lack of mediation. Therefore, illness threat perception did not have a significant direct or indirect association with anxiety when mediated by coping styles. These results can be seen in Table 6.



**Table 6**

*Mediation Analysis Results for the Relationship Between Illness Threat Perception and Anxiety, Mediated by Coping Styles*

<b>Total Effect</b>	<b>Direct Effect</b>	<b>Relationship</b>	<b>Indirect Effect</b>	<b>Boot LLCI</b>	<b>Boot ULCI</b>
<i>Threat perception → Anxiety</i>	<i>Threat perception → Anxiety</i>				
-0.05 (.36)	-0.08 (.14)				
		Threat perception → Problem-focused coping → Anxiety	-0.01	-0.07	.05
		Threat perception → Emotion-focused coping → Anxiety	-0.01	-0.03	.03
		Threat perception → Avoidant coping → Anxiety	-0.03	-0.09	.01

*Note:* Boot LLCI represents the Lower Limit of Confidence Interval, and Boot ULCI represents the Upper Limit of Confidence Interval. Confidence intervals that include zero indicate non-significant mediation effects.

***The relationship Between Illness Threat Perception and Depression, Mediated by Coping Styles***

The final mediation analysis explored the relationship between illness threat perception and depression, mediated by problem-focused coping, emotion-focused coping, and avoidant coping. The total effect of illness threat perception on depression was non-significant ( $c = .01$ ,  $p = .78$ ), indicating no significant association between illness threat perception and depression. Similarly, the direct effect of illness threat perception on depression was also non-significant ( $c' = .08$ ,  $p = .13$ ), suggesting no direct impact of illness threat perception on depression, independent of the mediators. The indirect effects through problem-focused coping ( $a^1b^1 = -.03$ , BCI [-0.12, .02]), emotion-focused coping ( $a^2b^2 = -.01$ ,

BCI [-.02, .03]), and avoidant coping ( $a^3b^3 = -.02$ , BCI [-.07, .01]) were non-significant, because all confidence intervals included zero, indicating a lack of mediation. Therefore illness threat perception did not have a significant direct or indirect association with depression when mediated by coping styles. These results can be seen in Table 7.

**Table 7**

*Mediation Analysis Results for the Relationship Between Illness Threat Perception and Depression, Mediated by Coping Styles*

<b>Total Effect</b>	<b>Direct Effect</b>	<b>Relationship</b>	<b>Indirect Effect</b>	<b>Boot LLCI</b>	<b>Boot ULCI</b>
<i>Threat perception → Depression</i>	<i>Threat perception → Depression</i>				
.01 (.78)	0.08 (.13)	Threat perception → Problem-focused coping → Depression	-.03	-.12	.02
		Threat perception → Emotion-focused coping → Depression	-.01	-.02	.03
		Threat perception → Avoidant coping → Depression	-.02	-.07	.01

*Note:* Boot LLCI represents the Lower Limit of Confidence Interval, and Boot ULCI represents the Upper Limit of Confidence Interval. Confidence intervals that include zero indicate non-significant mediation effects.

## Qualitative Content Analysis

### *Coping Strategies Experienced as Helpful*

32 participants wrote about the coping strategies they experienced as helpful. Based on the content analysis, eighteen helpful coping strategies were identified. The frequency and description of each helpful coping strategy can be found in Table 8.

**Table 8**

#### *Frequencies and Descriptions of Helpful Coping Strategies*

<b>Helpful Coping Strategy</b>	<b>Frequency</b>	<b>Description of Results and Quotations</b>
Sleep	16	Sleep was the most commonly mentioned helpful coping strategy. Sixteen participants spoke about the importance of maintaining a regular sleep schedule and getting enough sleep at night: <i>“Habits of sleeping at the same time each night and waking up the same”, “I recognise the importance of sleep, especially night time sleep”</i> .
Naps	13	Thirteen participants wrote about how they find regular naps helpful in managing drowsiness and preparing for busier days: <i>“I nap strategically based on how drowsy I am, how much time I have that day, and what I have to get done that day”</i> .
Medication	10	Ten participants mentioned the different ways they use medication to cope with their narcolepsy such as taking it one hour before getting up and spreading it throughout the day: <i>“I spread my modafinil medication throughout the day - morning, mid-morning, mid-afternoon, late afternoon”</i> .
Eating	9	Nine participants wrote about different aspects of eating they find useful in coping with narcolepsy such as having regular, smaller meals and avoiding sugar: <i>“I avoid consuming foods or beverages that will likely trigger a sleep attack, anything with added sugar, refined carbs, heavy meals”, “coping strategies like regular eating habits and low carb”</i> .
Social	8	Eight participants wrote about how they find both emotional and practical

support		support from friends and family useful: <i>“Have my friends wake me up early”, “support emotionally through friends”</i> .
Exercise	7	Seven participants wrote about the benefits of exercise, particularly walking: <i>“if I can do some exercise, like going for a walk”</i> .
Caffeine	6	Six participants wrote about how caffeine in coffee or diet coke helped them to cope: <i>“I drink coffee throughout the day”</i> .
Pushing through	6	Six participants wrote about pushing through their sleepiness so they can do the things they want: <i>“Trying to go that little harder and power through”, “I keep myself in panic mode sometimes so I can do things without sleep getting in the way”</i> .
Acceptance	6	Six participants wrote about how accepting their condition and learning to live with it helps them to cope daily: <i>“I just accept the fact that I’m going to be more tired than everyone else around me and continue to live my life the best that I can”</i> .
Positive reframing	5	Five participants positively reframed their experience and described this as helpful when coping with their narcolepsy: <i>“My condition is not as bad as people in certain situations, so perspective on this is good”, “Trying to think positively about things if I feel like I’m overreacting or being too negative”</i> .
Pacing	5	Five participants wrote about how pacing their daily activities helps them to cope: <i>“I sit down and take breaks when doing physical activity for more than an hour”</i> .
Professional support	5	Five participants experienced support from healthcare professionals as helpful <i>“speaking to a health care professionals”, “occasionally going to see my psychologist”</i> .
Planning/ Routine	5	Five participants wrote about planning their day ahead to manage and conserve their energy <i>“I try to keep everything ready the night before so I don’t have to put in so much effort in the mornings”</i> .
Self-care/ compassion	3	Five participants wrote about aspect of self-care and being kind to themselves to cope with their narcolepsy: <i>“I really try to not beat myself up anymore”, “little treats to bring up my spirits”</i> .
Peer support	3	Three participants wrote about the benefits of connecting with other people with narcolepsy: <i>“Visiting social media with threads for narcoleptics to feel a sense of community and that I am not alone in my struggles”</i>
Humour	3	Three participants use humour to cope with their narcolepsy: <i>“I like to try</i>

		<i>to keep the conversation around it very light hearted and humorous, even if others don't find it funny".</i>
Travel adjustments	3	Three participants wrote about using different ways to travel to conserve their energy: <i>"Using taxis to commute allows me to move around, preserve energy and sleep in the taxi, and be rest assured I'll arrive at my destination".</i>
Hobbies	3	Three participants wrote about different hobbies they engaged in to cope with their condition <i>"bake, go on walks, read, watch tv, listen to audiobooks, and podcasts".</i>

Maintaining a regular sleep schedule and having naps throughout the day were the most commonly reported helpful coping strategies in this sample which are the primary components of non-pharmacological treatment for narcolepsy (Scammell, 2016). Taking medication and using it in ways that benefited routines, as well as modifying aspects of their eating were also particular common ways this sample found helpful in managing their narcolepsy. These are all problem-focused ways of coping and supports the descriptive finding that this sample engaged in slightly higher problem-focused coping strategies than the normative sample.

### ***Coping Strategies Experienced as Unhelpful***

23 participants wrote about the coping strategies they experienced as unhelpful. Based on the content analysis, thirteen unhelpful coping strategies were identified. The frequency and description of each unhelpful coping strategy can be found in Table 9.

**Table 9***Frequencies and Descriptions of Unhelpful Coping Strategies*

<b>Unhelpful Coping Strategy</b>	<b>Frequency</b>	<b>Description of Results and Quotations</b>
Naps	5	Five participants experienced naps as unhelpful due to feeling worse upon waking: <i>“life I don’t always find taking naps helpful as it can sometimes worsen tiredness episodes”, “sometimes they actually leave me grumpy for some time after waking up”</i> .
Caffeine	4	Four participants wrote about how caffeine is an unhelpful coping strategy for them: <i>“Drinking caffeine. It just makes me anxious and doesn’t help me feel less tired at all.”, “I find I may get a caffeine crash and feel more tired after”</i> .
Substances	3	Three participants wrote about the different substances they find unhelpful in coping with their condition: <i>“I sometimes will have an alcoholic drink when I have a bout of insomnia”</i> .
Social support	3	Three participants experienced talking to others who don’t understand the condition as being unhelpful: <i>“Talking to other people (they just don’t freaking get it)”, “I don’t like talking about it to my loved ones too much - they don’t understand or seem to listen/care”</i> .
Stress/ Pressure	3	Three participants spoke about the unhelpfulness of internal and external pressure: <i>“Pushing myself or ignoring the drowsiness is unhelpful.”</i>
Sleep	2	Two participants did not find sleep helpful in coping with their narcolepsy: <i>“I try to sleep early or get more hours of sleep in, but I find that it makes no real difference. I still feel sleepy no matter what”</i>
Exercise	2	Two participants experienced exercise as an unhelpful coping strategy: <i>“While I am fit and have more physical ability than many people I know, exercise has never helped me to feel more awake or energized. It actively makes me more tired, often to the point that I can’t do anything else after”</i> .
Rumination	2	Two participants explained how over thinking about their condition was unhelpful <i>“I think about the future which opens up Pandora’s box and I begin to overthink”, “lead to thinking about</i>

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the condition more than is useful”.

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Planning/Routine	2	Two participants wrote about how planning their daily routines were not experienced as helpful: <i>“Planning is something many do, but having Narcolepsy, it's less useful for me because I know not what my body will decide for itself”</i> .
Self-criticism	1	One participant wrote about the unhelpfulness of self-criticism: <i>“I beat myself up”</i> .
Eating	1	One participant explained how eating makes them more sleepy: <i>“Eating when I am drowsy is super unhelpful”</i> .
Social withdrawal	1	One participant spoke about the downside of social withdrawal: <i>“Withdrawing socially conserves energy but can get lonely”</i> .
Medication	1	One participant mentioned that their narcolepsy medication did not help them to cope: <i>“Taking modafinil”</i> .

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The coping strategies that this sample found most commonly unhelpful were naps and caffeine. These strategies were reported as beneficial by numerous participants, highlighting the diverse nature of coping styles and their effectiveness within this population.

## Discussion

### Summary of Main Findings

This study aimed to explore illness perception, coping and psychological outcomes in young adults with narcolepsy, and answer the following research questions: first, what is the psychosocial functioning of young adults with narcolepsy, as measured by symptoms of anxiety, depression, and health related quality of life? Second, what are the patterns of illness perception in young adults with narcolepsy, as measured by perceived threat. Third, what types of coping strategies do young adults with narcolepsy use? And finally, does coping

mediate the relationship between illness perception and psychosocial outcomes in young adults with narcolepsy?

This sample of young adults with narcolepsy had ‘moderate’ levels of anxiety and just below ‘moderate’ levels of depression. The overall sample of young adults in this study also had lower physical and mental quality of life scores than the normative sample for young adults, as did the sample who completed all measures. This is in line with findings from paediatric and adult narcolepsy populations who have been found to have poor psychosocial outcomes (Chen et al., 2021), such as high levels of anxiety and depression (Parmar et al., 2019; Barateu et al., 2020) and significantly lower quality of life than controls (Rocca, 2016; Barker et al., 2020), and consistent with initial findings in the young adult narcolepsy population who were found to have lower mood and health-related quality of life than young adults without narcolepsy (Kapella, 2015). The sample of individuals who did not complete all measures, however, had a higher physical quality of life and similar mental quality of life score to the normative sample for young adults. Participants are more likely to engage in research that they perceive as relevant and important, and therefore individuals with lower quality of life may have perceived the research as more relevant and consequently been more motivated to complete all the measures in this study (Brito, 2017).

This sample of young adults with narcolepsy experienced high levels of perceived threat in relation to their condition, which contrasts with research in other chronic illness populations that has found chronic patients, in general, do not perceive their illness as negative and threatening (Adrian-Arrieta et al., 2018). This trend highlights the need to explore illness perceptions in other populations of narcolepsy to understand whether this



experience is specific to young adults or if it is a common experience for the whole narcolepsy population.

This sample of young adults with narcolepsy engaged in higher levels of problem-focused coping strategies and emotion-focused coping strategies to the general population. The overall sample also had the same avoidant coping score as the general population. This supports the qualitative exploration of coping strategies in this population which found that the problem-focused coping strategies of maintaining a regular sleep schedule, having naps throughout the day, taking medication and modifying aspects of nutrition were the most commonly reported helpful coping strategies in this sample. Whilst this is encouraging as problem-focused coping has been found to be predictive of positive outcomes (Hegarty et al., 2021), it is important to note that the coping strategies that this sample found most commonly unhelpful were naps and caffeine. These are two problem-focused coping strategies that were also frequently cited as helpful, reflecting the individuality of the helpfulness of coping styles in this population.

It is of particular interest that only five individuals reported professional support as being helpful in coping with their narcolepsy. This aligns with research indicating that young adults with chronic illnesses often do not view healthcare professionals as their main source of support or guidance in coping with their condition (Wilson, 2017). Whilst it is possible to hypothesise the potential factors behind this finding, such as the impact of burnout on professional-patient relationships (Bhatnagar, 2020) or potential barriers impeding this population from accessing healthcare professionals (Schwarz et al., 2022), more research is need to explore this concept before drawing any conclusions.

This study found a significant direct and total effect of illness threat perception on physical quality of life and mental quality of life, indicating that higher levels of threat perception were associated with lower physical and mental quality of life in this sample. This supports previous research suggesting that chronic illness populations that view their illness as more negative and threatening report worse quality of life (Adrian-Arrieta et al., 2018). The direct and total effects of threat perception on both anxiety and depression, however, were non-significant, suggesting that higher levels of threat perception were not associated with higher levels of anxiety and depression. This contrasts with previous research that has found high levels of threat perception to be strongly associated with symptoms of anxiety and depression (Kuiper et al., 2021). Finally, the indirect effects for all four mediation analyses were non-significant, indicating that coping styles did not mediate the relationship between threat perception and psychosocial outcomes in this sample. Consequently, in contrast to previous findings on young adults with chronic conditions (Limperg et al., 2020; Stapersma et al., 2019; Verhoof et al., 2014) these results do not support the application of the Common Sense Model to this sample of young adults with narcolepsy.

### **Limitations**

The results of this study could have been affected by methodological limitations and practical considerations which will be considered in turn. Whilst using Hayes' bootstrapping approach to mediation analysis is particularly useful when dealing with small sample sizes, Fritz et al., (2007) recommends a sample size of 71 to achieve an acceptable power of 80% on Bias-corrected bootstrapping. Only 44 participants completed all the measures and were therefore included in the mediation analyses, resulting in the study being underpowered. This may have increased the risk of a Type II error when there may have actually been a meaningful relationship between the variables (Fritz et al., 2015). Narcolepsy, however, is a

rare condition, with the estimated prevalence ranging from 0.01-0.05% worldwide. Furthermore, the condition is often underdiagnosed and misdiagnosed, resulting in a small pool of potential participants. Consequently, recruiting large samples sizes and achieving satisfactory statistical power is difficult in this population (Dunne et al., 2016). Many studies examining psychological phenomenon in this population also had small sample sizes (Rocca et al., 2016; Parmer et al., 2019), and studies that have been able to recruit larger sample sizes have been based in and recruited from sleep disorder units and treatment centres (Li et al., 2022; Barateu et al., 2020). Additionally, due to the study's small sample size combined with the difficulties documented in the narcolepsy literature regarding under-diagnosis and delayed diagnosis (Dunne et al., 2016), participants were also not excluded from the analyses if they had not reported receiving their diagnosis from a clinician. Furthermore, whilst the majority of participants did report receiving their diagnosis from a clinician, there was no way of confirming this due to the online nature of the study. Consequently, the degree of confidence with which the study's findings can be generalised to the young adult narcolepsy population is impacted. This highlights the importance of clinicians based within narcolepsy clinics who have access this population engaging in this type of research enabling more reliable findings and conclusions to be drawn. Larger number of participants that have a confirmed diagnosis of narcolepsy by a clinician could be recruited. Furthermore, clinicians that have fostered strong professional-patient relationships with this population could assist with the completion of measures, reducing the likelihood of the participant drop out that was observed in this study.

A second limitation is the variance in how studies exploring the relationship between illness perceptions, coping and psychosocial outcomes are conducted. Mediation analysis methods have evolved over time due to advancements in statistical techniques. However, this

has resulted in a range of different methods being used to explore the relationship between illness perceptions, coping and psychological outcomes in chronic illness populations. For example, studies have used the Baron and Kenny method (Lawson et al., 2010), the Sobel test (Kwon et al., 2018), structural equation modelling (Knowles et al., 2020), and finally bootstrapping (Fan et al., 2012); bootstrapping was also chosen for this study due to its more robust estimation of indirect effects on small sample sizes. Different mediation analysis methods, however, yield different results, impacting the estimation of indirect effects, significance testing and model fit. Consequently, future research applying the Common Sense Model to chronic illness populations such as narcolepsy should consider this when comparing findings and drawing conclusions from their results (Fairchild, 2017).

A third limitation is whilst the Brief-COPE is commonly utilised this area of research when drawing on Leventhal's Common Sense Model (1979) as the theoretical underpinning, the types of coping strategies derived from this measure and entered into mediation analyses are inconsistent. For example, Cheng et al., (2020) separated the scale into five subscales (positive adaptation, denial and disengagement, spiritual coping, support seeking, and self-blame), Minshall et al., (2020) separated it into two coping styles (adaptive and maladaptive coping ) and this study used the three recommended subscales of problem-focused, emotion-focused and avoidant coping (Hegarty et al., 2021). This variation in the way coping is operationalised when using the Brief-COPE may result in potentially misleading findings. Future research exploring the relationship between illness perceptions, coping and psychological outcomes should come to a consensus about the most useful definitions of coping when conducting research with the Brief-COPE in order to effectively compare and apply their findings to the design of interventions improving psychosocial outcomes in chronic illness populations such as young adults with narcolepsy (Deempster et al., 2015).

The qualitative coping analysis, however, demonstrated that the problem-focused coping strategies this sample found most commonly unhelpful, namely naps and caffeine, were also frequently cited as helpful. Not only does this reflect the individuality of the helpfulness of coping styles in this population, but it could also explain why coping was not found to mediate the relationship between illness threat perception and psychosocial outcomes. The Brief-COPE might, therefore, not capture and reflect the dimensions of coping specific to young adults with narcolepsy, and consequently might not be the most appropriate measure to use with this population. Future research could focus on the development and validation of a narcolepsy-specific coping measure that is tailored to the unique challenges and stressors of the condition.

Finally, this study utilised a cross-sectional design, as is common with research examining relationships between illness perceptions, coping and psychological outcomes. Consequently, conclusions cannot be drawn about the direction of the relationship between illness perceptions and quality of life; future research should focus on utilising longitudinal designs so that causal inferences can be drawn about the relationships between these variables (Cai et al., 2022).

### **Clinical Implications**

The results of this study have important clinical implications for young adults with narcolepsy. This sample was found to have poor psychosocial outcomes in terms of levels of anxiety, depression, and quality of life. Whilst this was a small sample that may have been impacted by participation bias (Brito, 2017), young adults with narcolepsy should still be closely monitored by clinicians to ensure their psychosocial wellbeing isn't being negatively impacted, and early screening of poor psychosocial outcomes should be considered (Li et al.,

2021). As this study found that higher levels of threat perception were associated with lower physical and mental quality of life, this population may benefit from interventions that target these negative threat perceptions about their condition (Blanco, 2018). For example, interventions such as mindfulness have been found to be particularly useful for increasing awareness of illness perceptions and encouraging people with chronic illnesses to view their illness differently (Scroevers et al., 2015), and might, therefore, be useful in positively impacting the quality of life in young adults with narcolepsy.

This study also found that young adults with narcolepsy use a wide range of coping strategies, and that the ones they find helpful are predominantly problem-focused. As problem-focused coping has been found to be predictive of positive outcomes (Hegarty et al., 2021), it is important that clinicians share this rich pool of information with newly diagnosed young adults with narcolepsy, particularly as people with chronic conditions value experiential knowledge from peers about coping with and managing their condition (Thompson et al., 2022). It is also important to consider the individuality of each experience young adults with narcolepsy may have, due to the finding that two the coping strategies that this sample found most commonly unhelpful were also frequently cited as helpful. Therefore, clinicians should ensure they are implementing genuine patient centred care when supporting young adults with narcolepsy to manage their condition. This involves integrating evidence from best practice whilst acknowledging the distinctiveness and individuality of each person, and tailoring interventions to meet their specific needs (Stantana et al., 2018).

### **Research Implications**

Whilst higher levels of threat perception were not associated with higher levels of anxiety and depression, and coping styles did not mediate the relationship between threat

perception and psychosocial outcomes in this sample, this contrasts with the body of literature in this area (Adrian-Arrieta et al., 2018; Limperg et al., 2020; Stapersma et al., 2019; Verhoof et al., 2014). It is therefore essential that future research replicates this study whilst addressing the highlighted limitations to ascertain whether both the significant and non-significant findings are representative of young adults with narcolepsy. This will allow further insight and guidance into what interventions might be useful for improving psychosocial wellbeing in this population such as targeting illness perceptions or coping strategies, resulting in the development of more specific and tailored interventions for this population. This includes conducting research within narcolepsy treatment centres in order to reach a larger number of individuals in this population that have a confirmed diagnosis of narcolepsy by a clinician. Clinicians who have good relationships with these individuals should also support participants to take part in the research, contributing to likelihood of complete data sets being achieved. Furthermore, the design of future research should be carefully considered, such as developing and utilising a narcolepsy-specific coping measure and reflecting on the types of mediation analysis used when comparing findings and drawing conclusions from results (Fairchild, 2017). Regarding the qualitative findings, this study provided preliminary data on how young adults cope with narcolepsy. Future qualitative research should expand upon this to gain a more nuanced understanding of this phenomenon such as exploring how coping strategies develop, what makes them helpful or unhelpful, and what factors influence them; a particular area of interest could be why this population do not seem find professional support particularly helpful in coping with their condition.

Finally, longitudinal studies need to be conducted in this population so that causal inferences can be more confidently drawn about the relationships between the variables studied (Cai et al., 2022).

## **Conclusion**

This study has made an important contribution in gaining a better understanding of a population that has received little attention to date. Young adults with narcolepsy seem to have poorer psychosocial outcomes than young adults without this condition, whilst experiencing high levels of perceived threat in relation to their condition. Clinicians should closely monitor this population's psychosocial wellbeing and consider interventions that target this population's illness perceptions. This population, however, also engage in a wide range of coping strategies, with the most helpful ones being predominantly problem-focused. Clinicians should explore the usefulness of individual coping strategies with their patients to ensure they are providing best practice that is tailored to individuals' needs. Finally, this study has provided insights into how researchers can continue to build upon these results and learn about the needs of this population going forward. These should be carefully considered when designing studies so that more definitive conclusions can be drawn and contribute to the development of more specific and tailored interventions for this population, and ultimately enhance the quality of care for young adults with narcolepsy.



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**Section C**  
Appendix of Supporting Material

Section A: Literature Review

Appendix A: CASP Case Control Study Checklist

		<b>Inocente et al., 2014b</b>	<b>Parmar et al., 2019b</b>	<b>Quaedack et al., 2019</b>	<b>Rocca et al., 2016</b>	<b>Stores et al., 2006</b>	<b>Szakacs et al., 2019</b>
1	Did study address clear issue?	Yes	Yes	Yes	Yes	Yes	Yes
2	Did authors use appropriate method?	Yes	Yes	Yes	Yes	Yes	Yes
3	Recruitment method acceptable?	Yes	Yes	Yes	Yes	Yes	Not clear
4	Control selected in acceptable way?	Healthy Yes	Yes (But secondary data)	Yes	Yes	Yes	Yes
5	Exposure accurately measured to minimise bias?	Yes	Yes	Yes	Yes	Yes	Yes
6a	Were groups treated equally?	Yes	No (Secondary data analysis for control group)	Yes	Yes	Yes	Yes
6b	Confounding factors taken into account?	Yes	No	Yes	Yes	Yes	Yes
7	How large were treatment effects?	Significant results	Not significantly different to each other and no comparison to baseline	Significant results	Some significant results	Significant results	Some significant results
8	How precise was estimate of treatment effect?	Confidence interval not reported	Confidence interval not reported	Confidence interval not reported	Confidence interval not reported	Confidence interval not reported	Confidence interval not reported
9	Do you	Yes	Yes –	Yes	Yes	Yes	Yes

	believe results?		comparable to chronic pain				
10	Can results be applied to local population?	Yes	Yes	Yes	Yes	Yes	Yes
11	Do the result of study fit with available evidence?	Yes	First study	Yes	Yes	Yes	Yes

### Appendix B: CASP Cohort Study Checklist

		Guilleminault et al., 1998
1	Did study address clear issue?	Yes
2	Was the cohort recruited in an acceptable way?	Can't Tell
3	Was the exposure accurately measured to minimise bias?	Can't Tell
4	Was the outcome accurately measured to minimise bias?	Can't Tell
5a	Have the authors identified all important confounding factors?	No
5b	Have they taken account of the confounding factors in the design and/or analysis?	No
6a	Was the follow up of subjects complete enough?	Yes
6b	Was the follow up of subjects long enough?	Yes
7	What are the results of this study?	Marked psychosocial difficulties in this population
8	How precise are the results?	Can't Tell
9	Do you believe the results?	Yes
10	Can the results be applied to the local population?	Yes
11	Do the results of this study fit with other available evidence?	Yes
12	What are the implications of this study for practice?	

## Appendix C: AXIS Checklist

		Hovi et al., 2022	Inocente et al., 2014a	Parmer et al., 2019a	Szakacs et al., 2015	Zhang et al., 2020	Qu et al., 2022
1	Aims/objectives clear	Yes	Yes	Yes	Yes	Yes	Yes
2	Appropriate design	Yes	Yes	Yes	Yes	Yes	Yes
3	Sample size justified	No power calculation	No power calculation	No power calculation	No power calculation	No power calculation	No power calculation
4	Target population clearly defined	Yes	Yes	Yes	Yes	Yes	Yes
5	Sample representative of the population	Yes	Yes	Yes	Yes	Yes	Yes
6	Selection process appropriate	Yes	Yes	Yes	Not clearly described	Yes	Not clearly described
7	Measures to categorise non-responders	No	No	Yes - Reasons for non-responders stated	No	No	No
8	Outcome variables appropriate to aims	Yes	Yes	Yes	Yes	Yes	Yes
9	Outcome variables measured appropriately	Yes	Yes	Yes	Yes	Yes	Yes
10	Statistics appropriate	Yes	Yes	Yes	Yes	Yes	Yes
11	Clear method section	Yes?	Yes	Yes	Yes	Yes	Yes
12	Basic data described	Yes	Yes	Yes	Yes	Yes	Yes
13	Response rate	59% for parental questionnaires, 51% for CDI, 52% for YSR  Low response rate justified/ thought about	Not clearly reported – referred to previous study for study methodology	30/35	DAWBA 37/38 PANSS 35/38 ADHD-RS 32/38 ASSQ 32/38 ADHRDS 32/38 Cog Ax 35/38	Not clearly reported	Not clearly reported
14	Information	Non	Not possible	Not	Not	No	No

	about non-responders	responders demographics similar		possible	possible		
15	Results internally consistent	Yes	Yes	Yes	Yes	Yes	Yes
16	All results reported	Yes	Yes	Yes	Yes	Yes	Yes
17	Conclusions justified by results	Yes	Yes	Yes	Yes	Yes	Yes
18	Limitations discussed	Yes	Yes	Yes	Yes	Yes	Yes
19	Declaration of interests noted	Disclosure statement but none declared	Yes	Disclosure statement but none declared	Disclosure statement but none declared	Disclosure statement but none declared	Disclosure statement but none declared
20	Ethical approval or consent attained	Yes	Yes	Yes	Yes	Yes	Yes

## Section B: Empirical Study

### Appendix A: Demographic Questions

1. How old are you?

16, 17, 18, 19, 20, 21, 22, 23, 24, 25

2. How would you describe your gender?

Male

Female

Non-binary / third gender

Prefer not to say

Other (please provide detail)

3. What is your ethnic background?

White - English/ Welsh/ Scottish/ Northern Irish/ British

White - Irish

White - Gypsy or Irish Traveller

White - Other

Mixed/ Multiple ethnic groups - White and Black Caribbean

Mixed/ Multiple ethnic groups - White and Black African

Mixed/ Multiple ethnic groups - White and Asian

Mixed/ Multiple ethnic groups – Other

Asian/ Asian British - Indian

Asian/ Asian British – Pakistani

Asian/ Asian British - Bangladeshi

Asian/ Asian British - Chinese

Asian/ Asian British – Other

Black/ Africa/ Caribbean/ Black British - African

Black/ Africa/ Caribbean/ Black British - Caribbean

Black/ Africa/ Caribbean/ Black British - Other

Arab

Any other ethnic group

4. What country do you live in?

England

Scotland

Wales

Northern Ireland

Other (please provide details)

5. What type of narcolepsy do you have?

Type 1 narcolepsy (narcolepsy with cataplexy)

Type 2 narcolepsy (narcolepsy without cataplexy)

6. What age were you diagnosed with Narcolepsy?

Appendix B: The Brief Illness Perception Questionnaire

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Appendix C: Brief-COPE

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## Appendix D: Qualitative Coping Questions

This last section will ask you about some of the ways you cope specifically with narcolepsy. What are some of the different ways you cope with your narcolepsy?

- Are there any coping strategies you use that you find particularly useful or helpful?
- Are there any coping strategies you use that you find more unhelpful?

Appendix E: Short Form Health Survey

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Appendix F: Hospital Anxiety and Depression Scale

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# Illness perceptions, coping and wellbeing in young adults with narcolepsy.

## Salomons Institute for Applied Psychology

One Meadow Road, Tunbridge Wells, Kent, TN1 2YG

[www.canterbury.ac.uk/appliedpsychology](http://www.canterbury.ac.uk/appliedpsychology)

## Information Sheet

Title of Project: **Psychosocial functioning of young adults with narcolepsy.**

IRAS ID: 308847

Hello. My name is Alex and I am a trainee clinical psychologist at Canterbury Christ Church University. I would like to invite you to take part in a research study. Before you decide whether to take part, it is important that you understand why the research is being done and what it would involve for you.

Please talk to others about the study if you wish to.

**Part 1** tells you the purpose of this study and what will happen to you if you take part.

**Part 2** gives you more detailed information about the conduct of the study.

### **PART 1**

#### **What is the purpose of the study?**

This study hopes to provide information about what young adults with narcolepsy think about their condition, how it might impact different areas of their life, and finally how they cope.

This information will help professionals to understand what support might be useful to offer young adults living with narcolepsy.

#### **Why have I been invited?**

You have been invited to take part in this study because you are a young adult between the ages of 16 and 25, and have a diagnosis of narcolepsy.

#### **Do I have to take part?**

It is up to you to decide whether to join the study. If you agree to take part, you will be asked to complete an online consent form. You are free to withdraw at any time, without giving a reason. This would not affect the standard of care you receive.

#### **What will I be asked to do?**

If you do decide to take part in this study, you will be asked to complete some online questionnaires. The first questionnaire will ask for information describing you (e.g. your age and gender) and five questionnaires that are related to your experience of living with narcolepsy, the ways you cope and your general wellbeing. These should take around 30-35 minutes to complete in total but you can take breaks as needed. Completing the consent form will be taken as evidence of your giving informed consent for your questionnaire answers to be used in the study.

### **What are the possible disadvantages and risks of taking part?**

Whilst we do not anticipate any disadvantages or risks of taking part in this study, if you experience any concerns you will be able to drop out of the study without having to give a reason, and/or contact me via email if you have any questions.

### **What are the possible benefits of taking part?**

We cannot promise the study will help you personally, but the information we get from this study will help improve our understanding of living with narcolepsy as a young adult, which may in turn help professionals understand what support might be useful to offer young adults living with narcolepsy.

### **What if there is a problem?**

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

*This completes part 1. If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.*

## **PART 2**

### **How will we use information about you?**

We will need to use information from you for this research project. This information will include your anonymised questionnaire data and your email address (if you would like to be entered into the prize draw entered of £50 and/or hear about the results of the study in the future). People who do not need to know who you are will not be able to see your contact details. Your data will have a code number instead. We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no one can work out that you took part in the study. Once your participation in the study is complete, your data will be stored for 10 years, as this is generally considered the minimum length of time suggested by the Medical Research Council for basic research.

### **What are your choices about how your information is used**

You can stop being part of the study at any time, without giving a reason. If you wish to have any data collected during the study to be removed and destroyed this will be carried out as soon as possible. **You will need to remember the study ID number provided in order for us to identify and withdraw your data; it is a good idea to write this down and keep it somewhere safe at the beginning of the study.** If you wish to withdraw your data, please contact us by 1<sup>st</sup> March 2023.

### **Where can you find out more about how your information is used?**

You can find out more about how we use your information by emailing the research team at [a.burdass1439@canterbury.ac.uk](mailto:a.burdass1439@canterbury.ac.uk). Alternatively you can leave a message on the 24-hour voicemail phone number 01227 927070. Please leave a contact number and say that the message is for Alex Burdass and I will get back to you as soon as possible.

**What if there is a problem?**

If at any point throughout your participation in the study, you feel you have not been treated appropriately and wish to make a complaint, please find details of how to do this below.

**Concerns and Complaints**

If you have a concern about any aspect of this study, or feel distressed whilst completing the questionnaires, you should ask to speak to me and I will do my best to address your concerns and signpost you to further support. You can contact me by emailing [a.burdass1439@canterbury.ac.uk](mailto:a.burdass1439@canterbury.ac.uk). Alternatively you can leave a message on the 24-hour voicemail phone number 01227 927070. Please leave a contact number and say that the message is for Alex Burdass and I will get back to you as soon as possible. Please note that your responses will not be reviewed immediately by the study team, and therefore we encourage you to actively contact us if you feel distressed. If you remain dissatisfied and wish to complain formally, you can do this by contacting Dr Fergal Jones, Clinical Psychology Programme Research Director, Salomons Institute for Applied Psychology [fergal.jones@canterbury.ac.uk](mailto:fergal.jones@canterbury.ac.uk)

**What will happen to the results of the research study?**

The results of this study will be shared with you if you would like them to be. I will email a letter once the study has been completed. This will not comment on individual participants but will explain the findings of the study overall.

This study may be written up and submitted for publication in the future. You will not be identified in any reports or publications.

**Who is sponsoring and funding the research?**

Canterbury Christ Church University is sponsoring this study.

**Who has reviewed the study?**

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the NHS Research Ethics Committee.

**Further information and contact details**

If you would like to speak to me and find out more about the study, seek advice on whether you should participate, or have any other questions about it answered, you can contact me by emailing [a.burdass1439@canterbury.ac.uk](mailto:a.burdass1439@canterbury.ac.uk). Alternatively you can leave a message on the 24-hour voicemail phone number 01227 927070. Please say that the message is for Alex Burdass and leave a contact number so that I can get back to you.

If you are dissatisfied with the study and want to make a complaint, you can do this by contacting Dr Fergal Jones, Clinical Psychology Programme Research Director, Salomons Institute for Applied Psychology [fergal.jones@canterbury.ac.uk](mailto:fergal.jones@canterbury.ac.uk)

## Appendix H: Consent Form

### CONSENT FORM

Title of Project: **Psychosocial functioning of young adults with narcolepsy.**

IRAS ID: 308847

Name of Researcher: Alex Burdass

In order to proceed with the study, all consent clauses must be agreed to and selected. Once you have selected all the consent clauses, please take a screenshot of this page so that you have a copy of your consent form. Alternatively, please contact the research team by emailing by emailing a.burdass1439@canterbury.ac.uk who will be able to provide you with a copy of the consent form .

- I confirm that I have read and understand the information sheet for the above study.
- I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care being affected.
- I agree that anonymous quotes from my questionnaire and other anonymous data may be used in published reports of the study findings.
- I agree for my anonymous data to be used in further research studies.
- I agree to take part in the above study.

ID Number Here is your ID number: `{e://Field/Random%20ID}`

**Please write this down and keep it somewhere safe as it will be required if you wish to withdraw your data.**

## Appendix I: Debrief Sheet

End of Survey! **Thank you for completing the questionnaires!**

Please open the link below if you would like to be entered into the prize draw of £50 and/or hear about the results of the study in the future:

[https://cccsocialsciences.az1.qualtrics.com/jfe/form/SV\\_3PmYXXlCRspms0m](https://cccsocialsciences.az1.qualtrics.com/jfe/form/SV_3PmYXXlCRspms0m)

The link will be opened in a new tab so that your email address is stored separately from your questionnaire data.

---

### Further Information **Concerns and Complaints**

If you have a concern about any aspect of this study, or feel distressed from completing the questionnaires, you should ask to speak to me and I will do my best to address your concerns and signpost you to further support. You can contact me by emailing [a.burdass1439@canterbury.ac.uk](mailto:a.burdass1439@canterbury.ac.uk). Alternatively you can leave a message on the 24-hour voicemail phone number 01227 927070. Please leave a contact number and say that the message is for Alex Burdass and I will get back to you as soon as possible. Please note that your responses will not be reviewed immediately by the study team, and therefore we encourage you to actively contact us if you feel distressed. If you remain dissatisfied and wish to complain formally, you can do this by contacting Dr Fergal Jones, Clinical Psychology Programme Research Director, Salomons Institute for Applied Psychology [fergal.jones@canterbury.ac.uk](mailto:fergal.jones@canterbury.ac.uk)

### **Narcolepsy UK**

For further general advice and support for narcolepsy, you may wish to contact Narcolepsy UK. Narcolepsy UK is a charity that supports people with narcolepsy, their families, carers and others interested in improving their quality of life. If you have a specific narcolepsy related issue on which you need advice, please call their helpline: 0345 450 0394 or visit <https://www.narcolepsy.org.uk/>



## Call for participants!



Salomons Institute for Applied Psychology  
One Meadow Road, Tunbridge Wells, Kent TN1 2YG  
[www.canterbury.ac.uk/appliedpsychology](http://www.canterbury.ac.uk/appliedpsychology)

### Are you a young adult living with Narcolepsy?

We are trying to understand more about coping with Narcolepsy so that we can improve available care and support.

**What the study involves:**

Online questionnaires lasting around 30-35 minutes

**Who can take part:**

Young adults between the ages of 16 and 25 with a diagnosis of narcolepsy

**What the participants will receive:**

You can enter into a prize draw of £50



If you are interested in taking part in this study, please scan the QR code or open the link below:

[https://cccusocialsciences.az1.qualtrics.com/jfe/form/SV\\_cMGtM5EEI7I5tVs](https://cccusocialsciences.az1.qualtrics.com/jfe/form/SV_cMGtM5EEI7I5tVs)

Name of Researcher: Alex Burdass [ab1439@canterbury.ac.uk](mailto:ab1439@canterbury.ac.uk)

Appendix K: Ethics Approval Letter

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Appendix L: Content Analysis Codes and Quotes

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Appendix M: Inter-coder Reliability Codes and Quotes

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## **Illness perceptions, coping and wellbeing in young adults with narcolepsy**

### **Background**

Living with narcolepsy can have an impact on daily life, but this has not been explored in young people with narcolepsy. The aim of this study was to explore the way young adults with narcolepsy perceive their condition, understand what coping strategies they use, and explore their psychosocial wellbeing.

### **Method**

We developed an online research pack that consisted of five questionnaires that measured illness perceptions (the way people perceive their condition), coping strategies, quality of life, and symptoms of anxiety and depression). 54 young adults with narcolepsy took part in the study. Analyses (descriptive statistics, correlation, multiple mediation and content analysis) were carried out to look at the patterns of these variables and any relationships between them.

### **Results**

Descriptive statistics showed that young adults with narcolepsy experience high levels of perceived threat in relation to their condition, poorer quality of life than young adults without narcolepsy, and experience moderate levels of anxiety and depression. They also engage in predominantly problem-focused coping strategies and find sleeping, napping and taking medication the most helpful ways to cope with their condition. Finally, participants that had higher levels of illness threat perception also had lower quality of life.

### **Conclusions**

Clinicians working with young adults with narcolepsy should closely monitor their psychosocial wellbeing and consider interventions that target their illness perceptions to help improve their psychosocial wellbeing. They should also explore the usefulness of different coping strategies with their patients to ensure that care is tailored to individuals' needs.

## Declaration of the end of a study

### (For all studies except Clinical Trials of Investigational Medicinal Products)

**To be completed in typescript by the Chief Investigator or sponsor representative and submitted to the Research Ethics Committee (REC) that gave a favourable opinion of the research within 90 days of the conclusion of the study or within 15 days of early termination**

**For questions with Yes/No options please indicate answer in bold type.**

#### 1. Details of Chief Investigator

Name:	Alexandra Burdass
Address:	Salomons Institute for Applied Psychology Lucy Fildes Building  1 Meadow Rd, Tunbridge Wells  TN1 2YG
Telephone:	01227 927070
E-mail:	Ab1439@canterbury.ac.uk

#### 2. Details of study

Full title of study:	An Investigation into the Relationship between Illness Representations and Psychosocial Outcomes, and the Mediating Role of Coping in Young Adults with Narcolepsy.
IRAS ID:	308847
Name of REC:	HRA and Health and Care Research Wales

	(HCRW) Approval
REC reference number:	22/PR/1387
Date of favourable ethical opinion:	12 December 2022
Sponsor:	Canterbury Christ Church University

### 3. Study duration

Date study commenced:	1 January 2023
Date study ended	02/06/2023
Did this study terminate prematurely?	<b>No</b>

### 4. Recruitment

Number of participants recruited	<b>54</b>
Proposed number of participants to be recruited at the start of the study	112
If different, please state the reason or this	The local NHS Trust who had agreed to support recruitment had a change in staff, and were therefore unable to support the study. Participants were consequently recruited online (as detailed in the original ethics application).

### 5. Circumstances of early termination

What is the justification for this early termination?	
---	--

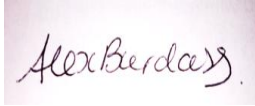
### 6. Potential implications for research participants

<p>Are there any potential implications for research participants as a result of terminating the study prematurely?</p> <p>Please describe the steps taken to address them.</p>	
---	--

## 7. Final report on the research

Have you submitted a Final Report?	No  If no, please submit a Final Report within 12 months of the end of the study (or for paediatric CTIMPs, within 6 months).  More information is available on the <a href="#">HRA website</a>
------------------------------------	---

## 8. Declaration

*Signature or Electronic Authorisation of Chief Investigator/sponsor representative:  *Please print below or insert electronic signature	
Print name:	<b>ALEXANDRA BURDASS</b>
Date of submission:	<b>13/07/2023</b>



## Appendix P: Feedback Summary to Ethics Panel

**Background:** Living with narcolepsy can have an impact on daily life, but this has not been thoroughly investigated in young people with narcolepsy. The aim of this study was to explore illness perceptions, coping strategies and psychosocial outcomes in young adults with narcolepsy, and the potential relationships between these variables.

**Method:** 54 young adults with narcolepsy took part in this study. They completed quantitative and qualitative questionnaires online assessing their illness perceptions, coping strategies, quality of life and symptoms of anxiety and depression.

**Results:** Descriptive statistics showed that this population experience high levels of perceived threat in relation to their condition, poorer quality of life than young adults in the general population, and experience moderate levels of anxiety and depression. They also engage in predominantly problem-focused coping strategies and find sleeping, napping and taking medication the most helpful ways to cope with their condition. Regression analyses showed that higher levels of illness threat perception were associated lower physical and mental quality of life. Coping did not mediate the relationship between illness perceptions and psychosocial outcomes in this study.

**Conclusions:** This study has made an important contribution to the limited literature on young adults with narcolepsy. Clinicians working with young adults with narcolepsy should closely monitor their psychosocial wellbeing and consider interventions that target their illness perceptions to help improve their psychosocial wellbeing. They should also explore the usefulness of different coping strategies with their patients to ensure that care is tailored to individuals' needs.

## Appendix Q: Journal of Sleep Research Author's Guidelines

### AUTHOR GUIDELINES

#### Sections

1. [Submission and Peer Review Process](#)
2. [Article Types](#)
3. [After Acceptance](#)

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- The title page of the manuscript, including:
  - Your co-author details, including affiliation and email address. (*Why is this important? We need to keep all co-authors informed of the outcome of the peer review process.*)

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You should list all funding sources in the Acknowledgments section. You are responsible for the accuracy of their funder designation. If in doubt, please check the [Open Funder Registry](#) for the correct nomenclature.

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- The full names of the authors with institutional affiliations where the work was conducted, with a footnote for the author's present address if different from where the work was conducted;
- Acknowledgments;
- Abstract unstructured;
- Up to six keywords;
- Main body: formatted as introduction, methods, results, discussion, conclusion;
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Article Type	Description	Word Limit	Abstract / Structure	Other Requirements
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<b>Review Articles</b>	critical reviews of the literature, including systematic reviews and meta-analyses	No limit	Yes, unstructured	<ul style="list-style-type: none"> <li>· Data Availability Statement</li> </ul>
<b>Short Reports</b>	preliminary findings of research in progress or a case report of particular interest	2000 words (including abstract, keywords, and main text)	Yes	<ul style="list-style-type: none"> <li>· Data Availability Statement</li> <li>· IRB Statement</li> <li>· Maximum 4 figures or tables</li> <li>· Maximum 15 references</li> </ul>

<b>Editorials</b>	This journal will not consider unsolicited editorials, they will only be invited by the Editor.	800-1000 words	<ul style="list-style-type: none"> <li>Maximum 10 references</li> <li>Title should not exceed 85 characters</li> </ul>
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