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Psychosocial intervention for children with narcolepsy: Parents' expectations and perceived support

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Abstract

The study focuses on the parents of children who were affected by narcolepsy after a pandemic influenza and vaccination campaign in Finland. The main aim of the study was to clarify parents' expectations and perceived support from the intervention and to assess their need for additional support. The data were gathered using questionnaires. Fifty-eight parents answered the baseline questionnaire and 40 parents the final questionnaire. Parents' expectations of and perceived support from the intervention mainly related to peer support. The intervention offered an arena for sharing information and experiences and provided encouragement for coping in everyday life. Many expectations were not met, especially those concerning information about needed services, financial benefits and availability of local support. The results highlight that for persons with rare disorders and their families, an inpatient psychosocial intervention can offer an important arena to receive both informal and professionally led peer support. Comprehensive psychosocial and other support services are also needed in the community. Listening to parents' perspectives on the intervention and perceived support can help to establish multiform family-centred support for families with children affected by a rare chronic disabling condition.

Keywords

Childhood narcolepsy, peer support, psychosocial family intervention

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Introduction

Narcolepsy is a rare, lifelong neurological sleep disorder, with a prevalence of around .04% in the general population (Ohayon, 2008). In late 2009 and early 2010, a sudden increase in childhood onset narcolepsy was observed in Finland, occurring soon after the pandemic influenza A (H1N1) epidemic and the ASO3-adjuvanted Pandemrix® vaccination campaign. In 2012, vaccinated children aged 4–19 years were reported to have a 12.7 times higher risk of narcolepsy than unvaccinated individuals in the same age group (Nohynek et al., 2012). A co-occurrence between the vaccine and narcolepsy has been also established in other countries, for example, Sweden (Persson et al., 2014) and UK (Miller et al., 2013).

The typical symptoms of narcolepsy are excessive daytime sleepiness, unintended sleep episodes and often cataplexy, a sudden loss of muscle tone often evoked by emotions. Additional symptoms may include hallucinations, sleep paralysis, fragmented nocturnal sleep and cognitive and emotional problems (Poli et al., 2013). Narcolepsy can lead to deterioration in the health-related quality of life such as physical and mental well-being, relations with friends, leisure time activities and socioeconomic disadvantage (Inocente et al., 2014; Jennum et al., 2012; Ohayon, 2013). Both pharmacological and non-pharmacological treatments for narcolepsy are based on the symptoms and daily challenges linked to the disorder (Lecendreux, 2014; Ohayon, 2013).

The need for psychosocial interventions for children with any chronic disabling condition and their families has been identified (Morawska et al., 2015). So far, there are only some non-pharmacological intervention studies focusing on narcolepsy (Marín Agudelo et al., 2014). However, the need for psychosocial interventions for persons with narcolepsy and their families has been recognized (Dorris et al., 2008; Lecendreux, 2014).

Peer support is one form of psychosocial support that has been defined within healthcare as assistance by a person with experiential knowledge of the stressors and behaviours similar to the target population (e.g. Dennis, 2003). Support from peers can improve knowledge and confidence in managing and coping with the disorder by sharing experiences with others. Peer support can be beneficial for both persons with disabilities and their families (Embuldeniya et al., 2013; Shilling et al., 2013). However, families of children with a rare disorder are often faced with insufficient opportunities for peer support and psychosocial support (Anderson et al., 2013).

The aim of this study was to analyse parents' expectations of and perceived support from a psychosocial family intervention as well as to identify needs for additional support in a situation where a rare disorder was induced by authorized activity, namely, vaccination against a pandemic influenza. The study questions were (1) which symptoms occurring after the onset of the child's narcolepsy were reported by the parents? (2) What were the parents' personal concerns and how much support had they received after the onset of the child's narcolepsy? (3) What were the parents' expectations of the intervention and what was their perceived support from it? (4) What other needs for support were reported? This study is part of a larger study on the implementation of the intervention (Autti-Rämö et al., 2015).

Methods

The participants were parents of children and adolescents diagnosed with narcolepsy, who attended seven inpatient psychosocial rehabilitation interventions in Finland between spring 2012 and autumn 2013. The programme was arranged as two 5-day periods with an approximately 6-month interval for a maximum number of nine children or adolescents per group plus their parents

and siblings. Separate interventions were arranged for clients with different age ranges (6–12, 13–16 and 17–20 years). Altogether 45 children and adolescents with narcolepsy, 63 parents and 46 siblings took part in the first 5-day rehabilitation period. The attendance rate for the second period was 80% ($n = 36$) for the children, 79% ($n = 50$) for their parents and 72% ($n = 33$) for siblings.

The Social Insurance Institution of Finland (SII) developed the content and structure of the intervention based on the identified needs of the families gathered from treating physicians, parents and officials at the Ministry of Social Affairs and Health. Those needs that could be included in the content and aim of a short psychosocially oriented intervention were selected. The intervention was carried out with a multidisciplinary team in one rehabilitation centre. The aim of the intervention was to strengthen the participants' coping resources and support them in adapting to the disorder and the changes it caused in their daily lives. The intervention was implemented in a group format and aimed to offer both informal peer support and professionally led peer groups. The daily programme lasted 6 hours, comprising a total of 60 hours of supervised activities.

The intervention consisted of a combination of individual and group training or education with a particular emphasis on various group activities. The programme included lectures, psychosocial counselling, group discussions about participants' experiences and skills training. The themes included information about narcolepsy and its treatment, coping with the disorder, information about self-care management and support services and educational counselling. The multidisciplinary team included a nurse, a psychologist, a special education teacher, a social worker, an occupational therapist, a clinical neurologist, at least two nursing aides and other healthcare workers as needed. Access to the intervention was based on the referral by the child's attending physician.

Ethical approval for the study was granted by the ethical committee of the SII. Informed consent to take part in the present study was given by 58 (92%) parents. All of them answered the baseline questionnaire at the beginning of the first rehabilitation period. Forty parents (80%) took part in the second rehabilitation period and filled in the final questionnaire at the end of the period. Altogether 36 parents answered both baseline and final questionnaires.

In addition to demographic variables (age and gender), the following variables were used in the study:

- (a) Frequency of symptoms connected to narcolepsy (10 items, e.g. daytime sleepiness, cataplexy, hallucinations) assessed with a 4-point scale (0 = *none*, 1 = *seldom*, 2 = *frequently*, 3 = *daily*).
- (b) Parents' present concerns (10 items, e.g. child's coping in life, feeling of not being able to help, relationship with partner) assessed with a 3-point scale (1 = *none*, 2 = *some concern*, 3 = *much concern*).
- (c) Support for parents from family members, school and health services (8 items), assessed with a 5-point scale (1 = *no need/none*, 2 = *some*, 3 = *rather much*, 4 = *very much*).
- (d) Expectations of psychosocial and informational support during the rehabilitation intervention (16 items, e.g. escape from everyday worries, peer support for parents/family, information about narcolepsy and treatment) assessed with a 4-point scale (1 = *no need/not at all* ... 4 = *very much*).
- (e) Perceived support during the rehabilitation intervention (same items and assessment scale as in list d).

The questionnaire also included open-ended questions. Answers to them are presented in connection with the corresponding quantitative data. Sample answers to the open-ended questions were included if they added content to the structured questions.

Table 1. Sum scale items of psychosocial and informational support: expectations reported by all subjects at baseline ($n = 57$) and expectations and benefits reported by subjects answering both baseline and follow-up questionnaires ($n = 36$) (% of subjects reporting rather or very much expectations and benefits).

Sum scale		All subjects at baseline ($n = 57^*$)	Subjects answering both baseline and follow-up questionnaires ($n = 36$)	
		Expectations	Expectations	Benefits
Information on services and support	Information on SII benefits	67	64	17
	Information on support services	64	60	–
	Information on where to get support	42	36	3
	Information on narcolepsy and treatment	65	58	11
	Establishing a residential support network	51	53	3
	Assertiveness in care	45	47	17
Peer support and own coping	Escape from everyday worries	42	39	42
	Peer support	70	81	81
	Peer support for the child	86	91	86
	Opportunity to be heard	44	53	39
	Coping as a parent	51	50	17
Coping with the child's emotions	Recognizing the child's strengths	47	47	25
	Coping with the child's emotions	53	50	36
Information on upbringing	Information on the child's upbringing	30	25	17
	Coping with the child's peers	26	22	3

*One parent did not answer this part of the questionnaire.

Data analyses included distributions, cross tabulations, factor analyses and paired t tests using IBM SPSS Statistics 21.

Four sum scales were formed of the items on expectations and perceived support presented above under points (d) and (e). Factor analysis was performed using principal component analysis as the extraction method. Components with eigenvalues >1 were retained. A varimax rotation was applied to four components, explaining 65% of the total variance. The cut-off of factor loadings was set at .55 when allocating items to the scales. One item did not fulfil this criterion and was excluded from further analyses. The reliability of the sum scales was tested with Cronbach's alpha (α) coefficient and the Spearman–Brown (SB) coefficient. The scales are simple sum scores of the items in question divided by the number of items. The scales are as follows (Table 1): (1) information on services and support (6 items, $\alpha = .84$), (2) peer support and own coping (5 items, $\alpha = .82$), (3) coping with the child's emotions (2 items, SB = .73), (4) information on upbringing (2 items, SB = .57).

Results

Fifty-eight percent of the parents were female, and 42% were aged 40 years or younger, 42% were between 41 and 50 years and 16% were over 50 years. The child's narcolepsy had been diagnosed approximately one year before the intervention started.

The most frequent daily symptoms were daytime sleepiness, mood and behavioural changes. The parents reported increased aggressiveness, irritability and apathy, 'Lacks patience, is pessimistic and aggressive', 'Violent tantrums', 'Cheerless, apathetic'. Eighty percent of the respondents reported frequent or daily occurrence of difficulty in concentrating. Cataplexy was also a common symptom (Table 2).

The majority of the parents were very worried about how the illness would affect their child's ability to cope. Their feelings of inability to help their child and their own coping caused much worry. The parents also expressed concern over not having enough time for their spouse and to what extent the other children in the family would be able to cope with their sibling's illness (Table 3).

Many parents were angry and frustrated with the authorities: 'Trust in the authorities is lost', 'Somebody made a mistake ... and now innocent children are suffering', 'This has been a very traumatic experience ...'

The parents reported receiving much support especially from their spouses (much support reported by 77%) but less from healthcare (27%) or school personnel (23%).

At baseline, the parents' expectations focused on getting peer support for the child and for themselves and on learning more about narcolepsy and its treatment as well as benefits and support services (Table 1).

Differences between the sum scales of expected and perceived support from the intervention are presented in Table 4. For three of the sum scales, the mean scores of perceived support were statistically significantly lower than the corresponding expectations, indicating that the intervention did not offer sufficient support in those areas. Especially expectations concerning information on services and other support systems were rarely met. Received support from peers and for own coping corresponded with the expectations (Table 4.).

Peer support was also mentioned in the open-ended questions: 'I expected to get quite a lot of peer support from this program and that was what I got. By sharing common concerns with others, I got much support and learned new coping skills for the future'. 'The people around us do not understand our current, nightmarish reality. It was really important to meet others that have the same experiences. Understanding each other's situation ... I feel relieved, it feels we are not alone'.

The need for more varied support was reported as well. While some of these needs were unique and individual, many participants expressed a need for psychosocial support for the whole family, information about the health and social services available, educational counselling and financial support. The parents reported that their children had faced educational difficulties due to tiredness, daytime sleepiness and concentration difficulties. Some of the adolescents were weighing their future career choices. As narcolepsy can adversely affect the ability to work in certain professions, parents expressed a need for counselling to help make future plans and reach academic goals.

Financial issues also were of concern. Some parents had to quit their job or work part-time as their child needed more supervision at home than before, while for others, the new situation meant increased medical costs. As the disorder was due to vaccination recommended by the health authorities, the families considered that the government should provide the needed services and financial support.

Discussion

The study participants were parents of children who had developed narcolepsy after the vaccination campaign against the pandemic influenza of 2009. The main focus of the participants'

Table 2. Symptoms of narcolepsy occurring frequently or daily reported at baseline ($n = 58$), %.

	None	Seldom	Frequently	Daily	All
Daytime sleepiness	–	5	26	69	100
Cataplexy	14	26	40	21	100
Sleep paralysis	46	40	15	–	100
Hallucinations	20	45	30	5	100
Memory and learning difficulties	13	27	46	15	100
Difficulties to concentrate	2	18	51	29	100
Involuntary movements	32	35	26	7	100
Visual disturbances	49	36	11	4	100
Behaviour changes	4	16	45	36	100
Mood changes	–	16	40	44	100

Table 3. Parents' concerns reported in the baseline questionnaire ($n = 58$), %.

	No	Some concern	Much concern	All
Child's coping in life	–	24	76	100
Coping of siblings	33	53	14	100
Family member's illness	54	37	9	100
Not being able to help the child	5	47	48	100
Insufficient time for one's spouse	32	42	26	100
Relationship with partner	61	25	14	100
Coping as a parent	10	55	35	100
Job stress	31	50	19	100
Financial situation	33	51	16	100
Unemployment or fear of lay-offs	70	23	7	100

Table 4. Expected and perceived psychosocial and informational support at baseline and final questionnaires: mean scores of the sum scales (M), differences between means (t) and statistical significances (p).

Sum scale	Expected support	Perceived support	n	t	p (two tailed)
	M (SD)	M (SD)			
Information on services and support	2.57 (.68)	1.59 (.46)	35	8.65	.000
Peer support and own coping	2.87 (.68)	2.69 (.67)	32	1.38	.179
Coping with child's emotions	2.50 (.78)	2.04 (.74)	36	2.96	.005
Information on upbringing	2.15 (.53)	1.60 (.56)	36	4.80	.000

expectations and perceived support from the intervention was on peer support for both children and parents, and to be heard (cf. Järviöskoski et al., 2015). Studies have shown that peer support can produce several beneficial impacts and can lead to empowerment and increased control over one's life and promote positive well-being outcomes (e.g. Embuldeniya et al., 2013; Pfeiffer et al., 2011; Shilling et al., 2013). However, more research is needed to confirm the optimal forms and structures of peer support and its long-term effects as well as to clarify possible adverse outcomes identified in some earlier studies (e.g. Skea et al., 2011).

Although some expectations of the intervention were met successfully, a majority of the parents were not satisfied with the information they received concerning narcolepsy and its treatment, the social and health services available, the local network and possibilities for financial support. The unmet needs ranged from more intensive and longer term support to insufficient expert knowledge of the rare childhood onset narcolepsy. This emphasizes the need for improving the information provided to participants about the aims and contents of the intervention. Service providers also need to construct a more profound understanding of the families' needs. Comparing parents' experiences can, however, be somewhat problematic as the respondents' viewpoints can change due to various factors, such as response shifts (cf. Schwartz et al., 2007). Moreover, it should be noted that the unusual contextual factors – the vaccine-induced situation – raised dissatisfaction with the authorities, which may have affected the parents' perspectives.

In addition to the symptoms of narcolepsy, other remarkable changes in the child's psychological well-being were reported as the consequence of the disorder. The parents were worried not only about their child's ability to cope but also about their own coping resources. The parents reported that before the intervention, they had received support mainly from close family members, but that support received from professionals was less common. This may be due to the rareness of the disorder and the consequent initial focus of the authorities on the diagnostic and medical challenges at the expense of psychosocial well-being.

A common challenge identified in this study was how individuals and families with rare disorders can get the help they need in their own community. Persons with rare disorders and their families are confronted with inequities and difficulties in access to treatment (EURORDIS, 2009). Rare disorders have been a priority area in the health programmes and policies of the European Union. Actions to promote empowerment and to put patients with rare disorders on a more equal footing with those with more common disorders have been considered important (European Commission, 2014).

The psychosocial needs of families and children facing narcolepsy have been poorly recognized. This disorder with multiple symptoms has an impact on the psychosocial well-being of the whole family. It is well known that childhood illnesses are related to heightened risk of adverse well-being consequences not only in the affected children but also in their parents and siblings which needs to be considered when developing interventions (Burton et al., 2008; Giallo et al., 2014).

The participation rate of this study was high in both questionnaires when compared to other studies of rare diseases (Anderson et al., 2013; EURORDIS, 2009). Several limitations can, however, be identified. The study was based on a simple before–after design and no long-term follow-up was conducted. As the study did not include a comparison group, the effect of the length of time elapsed after the diagnosis on the need and content of the psychosocial support could not be studied. The study included participants in the first seven rehabilitation interventions for a rare disorder. It is possible that with more experience, the rehabilitation professionals might have been able to better meet individual needs. The questionnaires did not include standardized or generic measures and thus, we are not able to compare the results with other disorders. Future research is needed to compare the perceived experiences and psychosocial outcomes across intervention programmes in families living with narcolepsy.

Conclusion

A child's chronic illness may threaten the well-being of the entire family and the needs of the whole family should be considered when planning support services. Peer support was found to be an important form of social support for families with childhood onset narcolepsy. Inpatient psychosocial intervention can offer an important arena to meet peers and get professionally led support as it can be difficult to find local peer support for rare disorders. Developing comprehensive psychosocial and other support services in the local community for individuals with rare disorders and their families is also needed to help them cope with challenges in their daily lives.

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