



# Caring for Children and Adolescents with Multiple Sclerosis

Exploring the unmet needs and existing supports for paediatric multiple sclerosis caregivers

Rebecca Maguire<sup>1</sup>, Elisabeth Kasilingam<sup>2</sup>, Nora Kriauzaite<sup>2</sup>

# Table of Contents

	Page		Page
<b>Executive summary</b>	<b>1</b>	<b>Section 2: Survey results</b>	<b>22</b>
<b>Background</b>	<b>2</b>	Prevalence of pMS, caregivers and pMS caregivers in countries surveyed	22
Paediatric Multiple Sclerosis	2	Provision of support for MS caregivers and pMS caregivers	24
Caring for people with MS	2	Specific supports available	25
Caring for a child or adolescent with MS	3	Financial supports available	27
Objectives of report	4	Detailed descriptions of supports given	28
<b>Methodology</b>	<b>4</b>	Information on caregiver needs	28
Literature review	4	Other suggestions for supports and resources	28
Phase 1: Rapid systematic review	5	Survey summary	28
Phase 2: Search for additional literature	5	<b>Policy and action gaps</b>	<b>32</b>
Evidence synthesis	5	Addressing the need for psychological support	32
Quality appraisal	6	Addressing the need for information	33
Overview of studies included	7	Addressing the need for practical support	34
Survey of MS societies and experts	10	Addressing the need for educational support	34
Survey development and design	10	Addressing the need for social support	35
Organisations and experts contacted	11	<b>Conclusion</b>	<b>35</b>
Analysis	11	<b>Relevant links and further information</b>	<b>36</b>
<b>Results</b>	<b>13</b>	<b>References</b>	<b>37</b>
Section 1: Unmet needs of pMS caregivers	13	<b>Acknowledgements</b>	<b>38</b>
Need for psychological support	14	<b>Appendix - Survey on services and supports for caregivers of paediatric MS</b>	<b>39</b>
Need for information	16		
Need for practical support	18		
Need for educational support	19		
Need for social support	20		
Needs analysis evaluation and appraisal	20		

## Terms and abbreviations

<b>MS</b> Multiple Sclerosis	<b>EMSP</b> European Multiple Sclerosis Platform
<b>pMS</b> Paediatric Multiple Sclerosis	<b>HCPs</b> Healthcare professionals
<b>caMS</b> Children or Adolescents with Multiple Sclerosis	<b>DMTs</b> Disease Modifying Treatments
<b>Carers/Caregivers</b> Terms used interchangeably to represent those caring for others with MS or pMS/caMS	<b>IEP</b> Individual education plan

## Executive Summary

In recent years, the number of children and adolescents being diagnosed with multiple sclerosis (i.e. paediatric MS) has increasingly been recognised. A diagnosis of multiple sclerosis (MS) at such a young age can place extra demands on caregivers, who may be required to take on many diverse roles and caring responsibilities. Developing a better understanding and treatment of paediatric MS, as well as supporting and empowering MS carers, have been identified as key priority actions within the European Multiple Sclerosis Platform's (EMSP) Code of Good Practice. There has, however, been little focused analysis on the unmet needs of these caregivers, or the supports that may be available to them in a European context. We aimed to investigate these issues by conducting (1) a rapid systematic review of the literature into the unmet needs of caregivers of those with paediatric MS, and (2) a survey aimed at gathering information on the available supports and resources for caregivers completed by national MS societies and experts in the area of paediatric MS.

The results of the literature review, which amalgamates findings from several studies in the area, suggests that caregivers of children and adolescents with MS (typically their parents) can experience various interlinked unmet needs.

While these caregiver needs are particularly apparent around the time of diagnosis, they may continue well beyond diagnosis and highlight the caregivers' difficulties coping with a range of ongoing caring responsibilities. Extant literature points to different ways of helping families to better adapt to a diagnosis of paediatric MS, including through the provision of a range of informational, educational and social supports. The results of the survey of European MS societies and experts, however, suggests that there is considerable cross-country variation in the supports and resources available for paediatric MS caregivers. While some societies offer many tailored resources and information for caregivers of children and adolescents with MS, others lack access to any formal supports, with many regional differences observed.

Our findings point to policy and action gaps in the area of supporting paediatric MS caregivers. In particular, identifying means in which caregivers' needs may be met is something that should be prioritised. These issues are discussed in detail with a view to informing the development of recommendations to support caregivers of children and adolescents with MS.

### These include needs for:

- Psychological support 
- Better information 
- Practical support 
- Educational support 
- Social support 

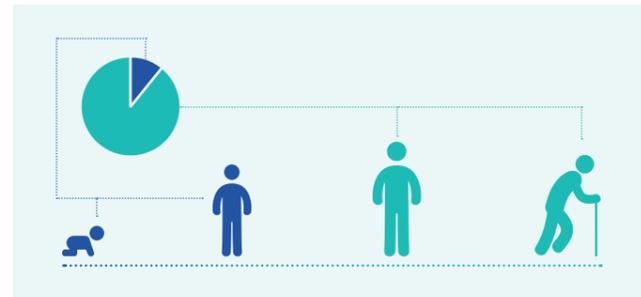
“ This report is really insightful for all the potential obstacles that are faced in the early stages of diagnosis. It will hopefully lead to improved support for caregivers and children and adolescents with MS over the coming years, not only in the UK but all over Europe. Emotionally it touches the heart knowing that a lot of other families are encountering the same issues.”

Jarrad Kitson, parent and carer of Eleonor who was diagnosed with MS at 15 years old

# Background

## Paediatric Multiple Sclerosis

Multiple Sclerosis was once considered a disease occurring exclusively in adulthood, however, there is growing recognition that MS can be diagnosed at any stage of the lifespan, including in early childhood. Due to advances in imaging technologies and other diagnostic procedures (Banwell et al., 2016), **the number of children and adolescents living with MS is increasing worldwide, with approximately 3 -11% of cases of MS now being diagnosed before the age of 18 (Chitnis, Glanz, Jaffin, & Healy, 2009).**



In light of this, in recent years more research attention has been dedicated to the study of Paediatric Multiple Sclerosis (pMS) with numerous research groups set up to investigate the clinical characteristics of the disease (Duignan et al., 2019; Waldman et al., 2016). While such work has shown that pMS shares some similarities with adult-onset MS, this research has also highlighted differences in the progression between the two diseases. For example, while children or adolescents with MS (caMS) have a higher rate of relapse early on in the disease, they often recover more quickly from relapses due to enhanced reparative ability (Banwell et al., 2016; Ghezzi et al., 2017). However, as disease onset and progression in pMS occurs at a time of rapid central nervous system development, this can lead to notable impairments in cognitive function (Amato et al., 2014), which may, in turn, be associated with a number of cognitive challenges, especially in an educational context. Indeed, recent work suggests that early-onset MS is associated with a later risk of cognitive impairment (Ruano et al., 2018). Fatigue is another commonly occurring symptom in pMS, affecting up to 76% of children (Carroll, Chalder, Hemingway, Heyman, & Moss-Morris, 2016). Symptoms such as this can have an impact on daily living, including the development and maintenance of peer relationships. Accordingly, recent reviews of the literature have shown that caMS suffer losses in their quality of life, most notably in the emotional domain (Steeves et al., 2018).

Those who care for caMS, most often their parents, are likely to play an important role in maintaining the wellbeing of the young person they care for, however, the task of caring may itself be associated with several difficulties. It has been well established that caring for a child with any chronic illness or disability can place considerable strain on caregivers, putting them at risk of a lower quality of life (Jensen et al., 2017; Murphy, Christian, Caplin, & Young, 2007; Raina et al., 2005). However, to date, there has been little systematic study of the impact that caring for caMS may have on the caregivers themselves. While there may be initiatives and supports in place for pMS caregivers, no universal recommendations or policies exist for how these carers should best be supported.

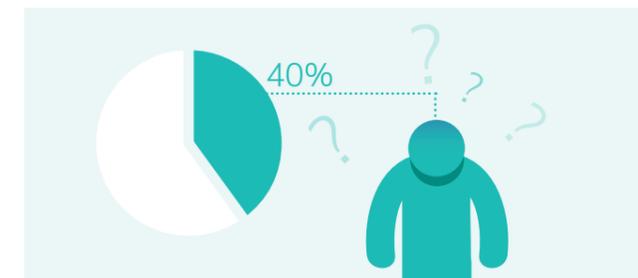
The aim of this report is therefore to present a focused review of literature in this area, as well as to undertake an analysis of existing supports and initiatives that are in place for pMS caregivers in Europe.

## Caring for people with MS

Although little work has focused on caring for those with pMS specifically, it is useful to first examine the literature on MS caregiving more generally, as this is an area which has received a greater deal of research attention. Many studies have explored the experiences of spousal carers of adults with MS (Appleton, Robertson, Mitchell, & Lesley, 2018), as well as those of young carers of parents with MS (Horner, 2013; Pakenham & Cox, 2012). Work in this area has shown that MS carers can be faced with several challenges, both positive and negative (Figved, Myhr, Larsen, & Aarsland, 2007; McKeown, Porter-Armstrong, & Baxter, 2003). While many caregivers adapt to their caring responsibilities over time (Appleton et al., 2018), it is well-established that caring for someone with MS can come with many “burdens”. **These can include financial, psychosocial, physical, and emotional burdens (Corry & While, 2009; Topcu, Buchanan, Aubeeluck, & Garip, 2016).**



A recent report by Merck (2018), in collaboration with IACO (International Alliance of Carer Organisations) and Eurocarers, echoed these findings. This report described an analysis of the experience of adult MS carers in seven countries (US, Canada, France, Germany, Italy, Spain and UK). Results revealed that caring for someone with MS can negatively impact on a many aspects of carers' life, including their physical health, emotional wellbeing, their work/career, and their finances. Almost half reported that caring had negatively affected their life goals or future plans, with many describing the uncertainty inherent with MS as a difficulty. Perhaps unsurprisingly, the most commonly reported challenge in providing care was the emotional/mental strain involved, with **over 40% of carers reporting severe or high stress levels often or all the time.**



The findings of the Merck survey also revealed that the majority of MS carers acknowledged a need for support, but just one in three carers were unaware of the support programmes for which they were eligible. It was noted that this need for support fluctuated depending on the experience of MS (e.g. needs were greater during a relapse). Many expressed a greater need for information, especially around the time of diagnosis. Responses revealed that many carers sourced information about MS online or via social media, rather than from health care professionals (HCPs). This was particularly true of younger carers. Despite expressed needs for support however, just 15% of carers reported connecting with other carers or patient organisations to help cope with the challenges of their role. A strong need for additional financial support was also evident. In a review, Lorefice et al. (2013) observed similar unmet needs for MS caregivers, including a need for more information and better psychological support.

While the Merck survey was limited to just seven countries (and just five European countries), it did reveal differences in the provision of support for carers across countries. For example, within the UK, 60% of carers revealed that progression and care needs were well explained to them, compared to an average of just

44%. Considering their findings, Merck included a number of recommendations in their report, including recommendations aimed at HCPs, MS societies, education and training institutions, employers and governments. These mainly focused on the provision of information and support for MS caregivers, including the provision of financial support and access to respite care. Other studies also highlight how the availability of social support can be beneficial for MS caregivers, which is consistent with these findings (McKeown et al., 2003; Rollero, 2016).

## Caring for a child or adolescent with MS

While those caring for caMS may have some overlap in the experiences of adult MS caregivers, it is also likely that they exhibit additional needs, and so may require a different set of supports. From the young person's perspective, dealing with MS is associated with a unique set of problems. For example, caMS are at risk of lower psychosocial wellbeing than their peers (MacAllister et al., 2013; Thannhauser, 2014; Weisbrot et al., 2010), and can suffer impairments in their educational activities, as well as difficulties with social relationships (Goretti et al., 2010). They may also be at risk of behavioural problems; however, work in this area has shown how important the parent-child relationship is in reducing the risk of such problems (Till et al., 2012).

CaMS may hence require numerous forms of support from parents, especially in terms of helping to meet their psychosocial and educational needs. They also may require greater practical support, including the administration of medication and the management of healthcare. As well as acting as a support for their children, parents may also experience their own complex set of needs as they cope with the psychosocial effects of the disease. It is worth therefore considering pMS caregivers as a unique subset of MS caregivers.

“When a child is diagnosed with MS, life is turned upside down, for the child, for parents and family.”

Elin Katrine Vestly, parent and carer of Ragna-Elise who was diagnosed with MS at 11 years old.

# Objectives and Methodology

## Objectives of report

The current report is intended to contribute to the development of a set of recommendations to support caregivers of paediatric MS:



**Objective 1:**  
To identify the unmet needs of paediatric MS caregivers

**Objective 2:**  
To identify existing activities, initiatives and resources for paediatric MS caregivers



**Objective 3:**  
To determine policy and action gaps relating to the provision of support for paediatric MS caregivers in Europe

## Methodology

Several different methodological approaches were adopted in order to meet the objectives. Firstly, to meet objective 1, a rapid systematic review of the literature was undertaken, which focused on an analysis of studies that directly explored the experiences of paediatric MS caregivers. Subsequently, to meet objective 2, MS organisations and experts in the area of paediatric MS in different European countries were surveyed to get an overview of existing supports for MS caregivers generally, and paediatric MS caregivers specifically. The findings of both these studies contributed to objective 3, described in the section 'Action and Policy gaps', which is intended to guide the development of recommendations for the support of paediatric MS caregivers.

## Literature review

In order to provide a broad but comprehensive review of relevant literature in the area of paediatric MS caregiving, a search strategy was developed which comprised of two different phases.



## Phase 1: Rapid systematic review

Firstly, in late April/early May 2019, a rapid systematic review of the literature relating to caregivers of pMS was undertaken. In order to achieve this, several databases were searched including Medline, Web of Science, PsycINFO and EMBASE. We wished to investigate any studies that had explored the experiences or unmet needs of paediatric MS caregivers, so kept our initial search strategy broad in order to capture as many relevant articles as possible. To this end, a range of appropriate search terms was developed that related to (1) paediatric MS, and (2) caregivers. Since care may be delivered by a range of people, including parents and other family members, we included search terms reflecting the numerous relationships that may exist (see Table 1 for search terms used).

Any abstracts that included these terms were imported into the systematic review software Rayyan (Ouzzani, Hammady, Fedorowicz, & Elmagarmid, 2016) for further analysis. After duplicate results were removed, titles and abstracts of all results were screened to determine whether they were relevant for the purposes of this study (see description of the SPIDER strategy used for this in the following section). This was followed by a full-text analysis of



## Phase 2: Search for additional literature

Following this initial search, three other papers meeting the inclusion criteria were identified by (1) examining the reference list of the papers identified in phase 1, and (2) examining more recent studies that had cited the papers in phase 1, through Google Scholar citations.

## Evidence synthesis

Our search strategy was guided by the SPIDER Tool for evidence synthesis (Cooke, Smith, & Booth, 2012) which is outlined in more detail in Table 2. Essentially this approach determines which studies to include in the review by specifying (1) the sample, (2) the phenomenon of interest, (3) the design, (4) the evaluation, and (5) the research type.

Table 1: Search terms used in initial review

Terms relating to paediatric MS	Terms relating to caregivers
paediatric multiple sclerosis	Caregivers [MESH]
pediatric multiple sclerosis	Parents [MESH]
childhood multiple sclerosis	Siblings [MESH]
early-onset multiple sclerosis	Family [MESH]
adolescent multiple sclerosis	caregiver*
	carer*
	parent*
	family*
	sibling*

relevant papers. Five of these studies were deemed to meet the study inclusion criteria, which stipulated that papers must: (1) describe a study that collects primary data from paediatric MS caregivers, (2) include at least one measure of the caregiver's experience/needs/quality of life, and (3) take the form of a full paper, rather than, for example, a conference abstract.

Table 2: Search terms and inclusion criteria as guided by the SPIDER approach (Cooke et al., 2012)

SPIDER component	Search terms or inclusion criteria
S Sample	Caregivers of pMS (identified using terms outlined in Table 1)
Pi Phenomenon of interest	Experience of pMS caregivers
D Design	Any study focusing on experience of pMS caregivers was considered, irrespective of design type
E Evaluation	The extent to which studies highlighted unmet needs of, and/or supports available to pMS caregivers
R Research type	Qualitative, quantitative or mixed method studies collecting primary data on pMS caregivers

# Methodology

## Quality appraisal

Given that our review gave rise to largely qualitative studies, with some quantitative studies, we utilised the MMAT (Mixed Methods Appraisal Tool) to appraise the methodological quality of the studies included in the review (Pace et al., 2012). This comprises of two screening questions applied to all studies, followed by four questions which vary depending on the methodological approach taken (see Table 3).

The MMAT enables the appraisal of mixed methods designs, qualitative designs, and a variety of different quantitative design types (e.g. randomized controlled trials, non-randomized designs and descriptive designs). As the studies included in our review were either qualitative or descriptive, we display the criteria for these only in the table below.

**Table 3:** Overview of the qualitative criteria included in the MMAT (Pace et al., 2012)

Question type	Quality criteria
Screening questions (all studies)	<ul style="list-style-type: none"> <li>Are there clear research questions/objectives?</li> <li>Do the data collected address the research questions?</li> </ul>
Qualitative questions	<ul style="list-style-type: none"> <li>Are the sources of qualitative data relevant to address the research questions?</li> </ul>
	<ul style="list-style-type: none"> <li>Is the process for analysing qualitative data relevant to address the research questions?</li> </ul>
	<ul style="list-style-type: none"> <li>Is appropriate consideration given to how findings relate to the context, e.g., the setting, in which the data were collected?</li> <li>Is appropriate consideration given to how findings relate to researchers' influence, e.g., through their interactions with participants?</li> </ul>
Quantitative descriptive	<ul style="list-style-type: none"> <li>Is the sampling strategy relevant to address the quantitative research question?</li> </ul>
	<ul style="list-style-type: none"> <li>Is the sample representative of the population under study?</li> </ul>
	<ul style="list-style-type: none"> <li>Are measurements appropriate (clear origin, or validity known, or standard instrument)?</li> <li>Is there an acceptable response rate (60% or above)?</li> </ul>

Following the MMAT guidelines (Pace et al., 2012), a score was derived for each study based on the proportion of the four criteria met (ranging from 0-100%). We used this score to generate an overall quality assessment rating for each study. Specifically, studies where all criteria were met (100%) were classified as high quality, studies where 2-3 criteria were met (50-70%) were classified as moderate quality, and studies where 0-1 criteria were met (0-25%) were classified as low quality.

The MMAT gives an indication of the methodological quality of studies in isolation. When conducting a review, however, it is also recommended to conduct an appraisal of the amalgamated review findings, especially in cases such as the current project which is intended to guide the development of a set of recommendations to support pMS caregivers.

In order to evaluate confidence in the overall synthesis of review findings, we used an approach guided by the GRADE (Grading of Recommendations Assessment, Development, and Evaluation) – Confidence in the Evidence from Reviews of Qualitative research (GRADE-CERQual) (Lewin et al., 2018; Lewin et al., 2015). We adopted this approach as most studies included in the review were qualitative in design.

The GRADE-CERQual consists of four key criteria – methodological limitations, relevance, coherence and adequacy of data (see Table 4 for an explanation of how these apply to the current project). Using this approach, an overall level of confidence in the various review findings can be estimated (high confidence, moderate confidence, low confidence, or very low confidence). This assessment was derived from both the individual study quality appraisals (as a result of the MMAT screening), and a consideration of the extent to which the overall review findings address the objectives of the review.

**Table 4:** Description of GRADE-CERQual used to evaluate review findings

GRADE-CERQual criteria	Description
Methodological limitations	Extent of methodological concerns about the design or conduct of studies included in the review
Coherence of the review findings	Assessment of how clear the fit is between the data from the included studies and the review findings
Adequacy of the data	Richness and quantity of data supporting the review findings
Relevance of the studies to the review question	Extent to which studies are applicable to the context of pMS caregivers

## Overview of studies included

Eight articles were included in the final qualitative synthesis of results. An overview of these studies can be seen in Table 5. This table includes details of each study's aims and objectives, the country in which the study was based, the sociodemographic details of participants and the caMS cared for, as well as the methodological approach employed and quality appraisal.

As can be seen in Table 5, the vast majority of caregivers in the studies described were parents of caMS (n = 230), with only one study (Harris, 2018) reporting another family member as a caregiver (in this case an aunt). Most caregivers were female. No study investigating the experience of formal caregivers was identified in our search. Most of the studies included involved a qualitative methodological approach (e.g. semi-structured interviews) and, while two quantitatively based studies were also identified (O'Mahony et al., 2018; Uccelli et al., 2013), it is notable that no large scale quantitative analysis of paediatric MS caregivers was found. It is also worth noting that most studies were based in English speaking countries (the UK, US, or Canada), with only one study based in Italy.

Using the MMAT approach described in Table 3, all the studies included in the review were deemed to be either high or moderate quality, so we can be relatively confident in the results. Typically, only minor concerns were evident which related to the potential representativeness of the samples, and the lack of consideration for research reflexivity in some of the qualitative studies.

While many of these articles included data from both pMS caregivers and caMS themselves, we have focused our later discussion of the findings of the caregiver experience only, with a view to identifying commonly reported unmet needs.

## Note regarding additional literature

To supplement the later discussion relating to unmet needs and supports available for pMS caregivers, additional sources were drawn upon that did not meet the inclusion study for the review but were deemed to be informative for the current purposes. This included a review article (Krupp, Rintell, Charvet, Milazzo, & Wassmer, 2016) which included recommendations on supporting families of pMS.

## Characteristics of studies included in the review in alphabetical order

Table 5: Characteristics of studies included in the review in alphabetical order

Authors	Title	Relevant Aims	Country	Caregiver details	Characteristics of caMS	Methodology	Quality appraisal
(Carroll, Chalder, Hemingway, Heyman, & Moss-Morris, 2016)	"It feels like wearing a giant sandbag." Adolescent and parent perceptions of fatigue in paediatric multiple sclerosis	To explore fatigue in pMS and gain insight into how parents and caMS respond to, and manage, fatigue	UK	13 parents (11 mothers and 2 fathers) of caMS aged 32-52 years (mean age = 46.8)	8 females and 7 males aged 9-18 years (mean age = 15.2 years). All had self-reported significant fatigue	Qualitative semi-structured interviews separately conducted with caMS and parents via telephone or in-person.	Moderate
(Cross, Shanks, Duffy, & Rintell, 2019)	Families' Experience of Pediatric Onset Multiple Sclerosis	To understand families' experience of pMS	USA	21 parents (18 mothers, 1 father and 2 couples). Mean age = 42.8	15 girls and 6 boys (mean age = 14.75 years). Mean time since diagnosis = 20 months (range 1-18 months)	Inductive thematic analysis used to identify themes. Analysed using thematic analysis.	Moderate
(Harris, 2018)	A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers	To explore caregiver perspectives of how family factors influence adaptation to pMS	USA	20 caregivers of caMS, all female (19 mothers and 1 aunt) aged 28-55 years (mean age = 44)	14 females and 8 males aged 7-22 years at time of interview. Mean age at diagnosis = 13	Qualitative interviews and completion of 50-item sociodemographic form. Thematic analysis used to identify themes.	High
(Hebert, Geisthardt, & Hoffman, 2019)	Insights and Recommendations From Parents Receiving a Diagnosis of Pediatric Multiple Sclerosis for Their Child	To understand the experience of receiving a pMS diagnosis from the time parents first sought care to diagnosis	USA	42 parents (40 mothers and 2 fathers)	32 females and 9 males aged 8-33 at time of interview (mean age = 17.6). All diagnosed prior to 18 (mean time since diagnosis = 4.25 years)	Qualitative telephone interview and online demographic survey. Thematic analysis used to identify common reactions to diagnosis and recommendations for HCPs.	Moderate
(Hinton & Kirk, 2015)	Paediatric multiple sclerosis: a qualitative study of families' diagnosis experiences	To examine caMS and parents experience of pMS diagnosis and identify facilitators and barriers to early diagnosis	UK	31 parents from 23 families (20 mothers and 11 fathers)	15 females and 6 males aged 8-17 years (mean age = 15 years)	Qualitative semi-structured interviews with caMS and parents. Transcripts analysed inductively using constant comparative model.	High
(Hinton & Kirk, 2017)	Living with uncertainty and hope: A qualitative study exploring parents' experiences of living with childhood multiple sclerosis	To explore the experiences of parents of caMS	UK	Same as Hinton (2015): 31 parents from 23 families (20 mothers and 11 fathers)	Same as Hinton (2015): 15 females and 6 males aged 8-17 years (mean age = 15 years)	Semi-structured conversational interviews with caMS and parents averaging 90 minutes long. Transcripts analysed inductively using constant comparative model.	High
(O'Mahony et al., 2018)	Pediatric-onset multiple sclerosis is associated with reduced parental health-related quality of life and family functioning	To evaluate the impact of MS on families compared to those with monophasic acquired demyelination syndrome (monADS)	Canada	58 parents of caMS, compared with 178 parents of children with monADS	39 females and 19 males (mean age of symptom onset = 13.9 years)	Quantitative. Questionnaires included measures of parents HRQOL and family impact. Statistical analysis used to compare parents of caMS with those of monADS, as well as to explore associations between key variables.	Moderate
(Uccelli et al., 2013)	Lack of information about multiple sclerosis in children can impact parents' sense of competency and satisfaction within the couple	To assess coping with family crisis and distress in couples with caMS compared to those with healthy children	Italy	15 couples of caMS and control group of 29 couples with healthy children	14 provided info – 11 males, 3 females; mean age 13.7. Mean time since symptom onset = 35.8 months	Quantitative. Questionnaire included various measures of psychological wellbeing, anxiety and depression, couples communication, sense of parenting competence as well as knowledge of MS scale. Statistical analysis compared groups and explored relationships.	Moderate

# Methodology

## Survey of MS societies and experts

To get a clearer understanding of the specific supports and resources available for European pMS caregivers, a survey was developed and circulated to representatives in national MS societies and experts in the area of paediatric MS (typically paediatric neurologists). The survey was also designed to collect information on the supports available for MS caregivers more generally.

## Survey development and design

This survey was developed based on findings from the literature search, as well as from previous questionnaires which involved some assessment of caregiver supports. This included an adaptation of some questions from EMSP's (2015) MS Barometer. A mixture of closed and open-ended questions was included to allow for the collection of a variety of information.

This survey was broken down into different sections (please see the appendix for all the questions included in the survey):

1. Organisational details and background. This collected information on the MS society and/or country in which the society/expert was based.
2. Estimates on the number of pMS diagnoses, number of caregivers, and number of pMS caregivers in the respondent's country, if available.
3. General overview of information programmes available for MS caregivers and pMS caregivers, including programmes aimed at informal and formal caregivers.
4. Details of specific supports available for general MS caregivers and pMS caregivers specifically. Respondents could select various options including information guides/supports, training programmes, telephone helplines, peer support groups (both face-to-face and online), social media supports, psychological services, respite care, a range of financial supports, and educational supports/information for schools.
5. Needs of MS caregivers or pMS caregivers. Here respondents were asked whether their country has undertaken any needs assessment of caregivers and pMS caregivers.
6. Finally, respondents were given the opportunity to indicate any additional articles/resources/supports available for pMS caregivers, as well as any other suggestions relating to supports for this group.

## Organisations and experts contacted

41 EMSP member societies from 36 European countries were invited to take part in the survey. In addition, a further 7 experts in the area of paediatric MS were invited to participate. In total, we received 23 responses from 17 countries (representing a response rate of 47% of countries)

An overview of the countries that participated is shown in Table 6.

Table 6: Details of survey respondents

(n)	Country	Respondents
1	Belarus	National MS society representative
2	Croatia	National MS society representative and expert in paediatric MS
2	Denmark	National MS society representative and expert in paediatric MS
2	Germany	National MS society representative and expert in paediatric MS
2	Greece	Two different MS society representatives
2	Ireland	National MS society representatives
1	Italy	National MS society representative
1	Lithuania	National MS society representative
2	Netherlands	National MS society representative and expert in paediatric MS
1	Norway	National MS society representative
1	Poland	National MS society representative
1	Romania	National neurodegenerative patient association representative
1	Russia	National MS society representative
1	Spain	National MS society representative
1	Switzerland	National MS society representative
1	Turkey	National MS society representative
1	UK	National MS society representative

## Analysis

In cases where there was more than one respondent for each country, we amalgamated these responses to enable an overview of services and supports for that country. Due to the small number of participating countries, we restricted our analysis to a descriptive overview of the data collected. We also used this information to map services and supports across the various respondent countries.

Unfortunately, we did not obtain information from all MS societies contacted, and some surveys were not fully completed. These included partial responses from the Netherlands and Russia. We therefore excluded these countries from the analysis relating to specific supports (described later).





# Results

## Section 1: Unmet needs of pMS caregivers

Based on an analysis of the studies previously described in Table 5, many unmet needs of pMS caregivers were identified using the technique of narrative synthesis. While the literature revealed the diverse experiences of pMS caregivers, most often parents, some key themes emerged in relation to unmet needs, many of which were overlapping. A summary of the main findings in relation to these needs is displayed in Table 7, and the subsequent sections elaborate on these in more detail.

Table 7: Summary of unmet needs of pMS caregivers

pMS caregiver experience	Unmet need
<b>Emotional impact of MS</b> <ul style="list-style-type: none"> <li>• Experience of negative emotions including fear, worry, anxiety, stress, strain, depression, helplessness</li> <li>• Perception that concerns were not addressed by HCPs during consultations</li> <li>• Strain of concealing feelings from caMS</li> <li>• Feelings of uncertainty over the future and disease management</li> </ul>	Need for psychological support prior to, during, and after diagnosis
<b>Access to information</b> <ul style="list-style-type: none"> <li>• Perception that concerns were not addressed by HCPs during consultations</li> <li>• Lack of knowledge about MS</li> <li>• Inconsistencies in information provided</li> <li>• Not knowing where to get information</li> <li>• Unsure how to manage disease and treatment</li> </ul>	Need for information about MS, treatment options and disease management
<b>Practical impacts of MS</b> <ul style="list-style-type: none"> <li>• Impact on schedule, including difficulty in predicting care needs</li> <li>• Impact on finances and employment</li> <li>• Difficulties with treatment administration and adherence</li> <li>• Impact on family relationships</li> </ul>	Need for practical supports, including financial support, care supports and family supports
<b>Experiences with school and educational system</b> <ul style="list-style-type: none"> <li>• Lack of accommodations in some schools</li> <li>• Difficulties with school communication</li> </ul>	Need for educational support, including school accommodations and awareness campaigns towards general public
<b>Social isolation and support</b> <ul style="list-style-type: none"> <li>• Variations in support from family/friends</li> <li>• Beneficial impacts of MS community, especially connecting with other caMS or pMS caregivers</li> </ul>	Need for social support from family/friends and connection with others in the MS community

# Results: Section 1

## Need for psychological support

All the studies analysed discussed at least some emotional impacts of pMS on caregivers, suggesting a need for greater psychological support. The provision of such support seems to be particularly important around the time of diagnosis, however, findings suggest that psychological support may be required at all stages of the disease trajectory. As discussed in more depth below, the most common emotion experienced by pMS caregivers was fear or worry. Helping to alleviate fears may therefore be a particularly important goal for those supporting pMS caregivers.

## Need for support leading up to, and during, diagnosis

As reported by parents in Cross et al.'s (2019) research, the diagnosis of pMS itself was often preceded by months of stress and anxiety, perhaps owing to the uncertainty parents experienced regarding possible causes of their child's symptoms. Hebert et al. (2019) reported that most caMS initially got an incorrect diagnosis and, during this time, parents frequently experienced frustration that their concerns were not taken seriously by HCPs. A similar finding emerged from Hinton and Kirk's (2015) study, where only half of parents felt their concerns were addressed during their initial consultation. This suggests that some fears may be alleviated by HCPs by simply taking time to discuss worries with parents early in the diagnostic process.

Cross et al. (2019) report that the diagnosis itself was often a difficult and upsetting period for parents, a finding echoed by other studies. For example, some parents reported feeling overwhelmed upon diagnosis (Hebert et al., 2019), and were worried that they were somehow responsible (Cross et al., 2019). Harris (2018) also found that stress and strain was a common experience at this time, but that caregivers going through additional stressful life events, such as marital separation, found this to be an even more difficult period. This suggests that an individual's life circumstances should be considered when assessing their support needs.

Other initial emotional reactions to diagnosis included shock, devastation and a difficulty in acceptance (Cross et al., 2019; Harris, 2018; Hebert et al., 2019), however, some parents also reported positive reactions such as relief (Hebert et al., 2019). In such cases, a diagnosis was seen as providing an explanation for the caMS's symptoms, which may have helped parents make better sense of the situation. This suggests that assisting parents to make sense of the diagnosis may be a useful means of helping them cope.

An added emotional difficulty for parents is that they may feel the need to conceal their reactions from their children (Cross et al., 2019). This was echoed in Hebert et al.'s (2019) study, with one participant noting, for example, that **"the hardest part...was having to be strong in front of him and never let him see me cry"**. In this context, parents appreciated being told of their child's diagnosis on their own so that they could contain their emotions when later discussing this with their child. This also gave them an opportunity to request more information from HCPs, which may also address another commonly expressed need (i.e. the need for information, discussed more later).

Interestingly, however, other studies have shown that adolescents themselves valued time alone with HCPs and experienced frustration if they were not part of the consultative process (Krupp et al., 2016). Krupp et al. (2016) acknowledge therefore that the optimal approach for sharing the diagnosis, as well as discussing disease management, may differ depending on the child's age. Balancing the needs of both parents and caMS may be difficult in this respect, and psychological support may be required for the whole family during the diagnostic process.

## Need for continuing psychological support beyond diagnosis

Depression was reported as another early reaction to diagnosis by some parents (Cross et al., 2019), however, Uccelli et al's (2013) study suggests that, overall, pMS parents may be at a greater risk of depression when compared to control parents, even years after diagnosis. This suggests that caregivers may take considerable time to adjust to the diagnosis and their caring responsibilities. Many may therefore require ongoing psychological support.

Interestingly, O'Mahony et al.'s (2018) analysis suggests that parents of caMS reported greater emotional dysfunction and worry irrespective of clinical disease activity. This suggests that all parents need emotional support, regardless of the caMS's disease status.

Fear about the future was a common theme to emerge in many of the study findings (Cross et al., 2019; Harris, 2018; Hebert et al., 2019). For instance, Cross et al (2019) noted that many parents feared a bad outcome, such as their child losing independence (e.g. ending up in a wheelchair), while Hebert et al.'s (2019) study found that some parents simply had a fear of the unknown. Parents also worried about the things that their child may miss out on in the future, as well as the impact that MS may have on their child's mental health (Carroll et al., 2016), academic and career aspirations (Hinton & Kirk, 2017). Some parents also expressed concerns as to what would happen if they weren't around to look after their children in the future. Uccelli et al's (2013) analysis suggests that worries were heightened in mothers, as opposed to fathers, suggesting that they may be particularly vulnerable.

Even on a daily basis, some parents reported reoccurring and intermittent fears. For example, some caregivers in Carroll et al's (2016) study feared that too much activity could lead to a relapse. Cross et al. (2019) also reported the process of MRIs to be a particularly stressful time, as this may lead to a fear of new disease activity. On a more practical note, they also expressed worries relating to the future affordability or efficacy of medications.

A quote from one of Harris' (2018) participants summed up the considerable strain that caregivers experience: **"I don't want to face a lot of stuff that I actually see. I don't really want to talk about it, a lot, because most of the time when I talk about it, it makes me very emotional..."**.

“ I have received psychological support along the way which was very important. I've also had good follow-up with my general practitioner who has had time to listen to my worries and sorrows.”

Elin Katrine Vestly, parent and carer of Ragna-Elise who was diagnosed with MS at 11 years old.

Almost every study described how parents experienced uncertainty to varying degrees, a factor that was largely responsible for the emotional impact of MS on caregivers. One study in particular (Hinton & Kirk, 2017) identified uncertainty as a unifying experience for families living with MS. Harris (2018) also found that the most common concern for parents was the unpredictable nature of the disease and the feeling of having no control. In an effort to combat feelings of uncertainty, parents may engage in behaviours that lead to further psychological strain, such as continuous monitoring of their child (Hinton & Kirk, 2017).

Experiences of uncertainty may also be exacerbated by lack of information, or indeed too much information, so its potential impacts are discussed in more detail below.

“ It is clear that the months before and after diagnosis have a big psychological impact on both the caregiver and a child. The worries that we had include incorrect diagnosis, what does the future hold for the child, the impact of diagnosis, academic aspirations, how do we cope with the future. Our own experience tells that the correct support in that crucial period is not there in the most part. We wish that more psychological support was available to deal with our worries.”

Jarrad Kitson, parent and carer of Eleonor who was diagnosed with MS at 15 years old



## Summary of psychological needs

- Greater psychological support for families is needed, especially around diagnosis – this may simply take the form of enabling caregivers to discuss their concerns with HCPs
- Given that the feeling of uncertainty is a dominant experience, great provision of information (see below) may also help alleviate caregivers' fears and lead them to feel more empowered to manage the effects of MS.

# Results: Section 1

## Need for information

A need for appropriate information was another recurring theme in the studies analysed and, as discussed above, a need that may have contributed to a range of negative emotional reactions experienced by caregivers. For example, some parents acknowledged that their fears stemmed from a lack of knowledge about MS (Hinton & Kirk, 2017). Others had preconceived inaccurate notions of what MS involved, which further exacerbated their fears.

A lack of knowledge relating to MS may have wider impacts for family relationships. For example, Uccelli et al. (2013) found that having a low knowledge of MS was associated with lower satisfaction within couples' relationships, as well as with poorer quality communication. They also found that insufficient knowledge of MS was associated with lower feelings of parental competence, which may affect parents' ability to manage the demands of care.

Analysis of the studies' findings suggested that knowledge of MS could be enhanced through the provision of information at various time points, as discussed below.

## Need for information on diagnosis

A need for information seemed especially apparent at the time of diagnosis, however, the provision of information to parents varied widely (Cross et al., 2019; Hebert et al., 2019; Hinton & Kirk, 2015). For example, Cross et al. (2019) reported that while some carers reported receiving helpful information regarding treatment and prognosis from treating physicians, others felt that they were not getting enough information (for example, one parent reported leaving their consultation with **"unanswered questions"**). This finding appears to be consistent with Merck's (2018) survey on general MS carers, where over half stated that HCPs did not explain the disease, its progression or potential care needs when they began their caring role.

In Hebert et al's (2019) study, some reported not knowing where to turn for information, while Hinton and Kirk (2017) noted that some parents were reluctant to seek help for fear of being labelled "neurotic" by medical staff. Some caregivers in Harris' (2018) study also reported struggling due to a perceived lack of resources. Many used the internet to improve their understanding, however, parents acknowledged that this was of limited value. A further danger of searching online for information is the potential for unreliable sources.

## Need for information regarding symptom management and treatment

Caregivers also reported uncertainty regarding how best to manage their caMS's symptoms, pointing to a greater need for information regarding disease management. For example, Hinton and Kirk (2017) noted that it was difficult for parents to interpret the invisible symptoms of MS (i.e. those symptoms that are not clearly apparent to an observer), while some of Cross et al.'s (2019) parents reported trouble in deciding how independent their caMS should be. Carroll et al. (2016) specifically focused on the experience of fatigue in this respect. They found that some parents reported being unsure of how to manage their child's fatigue – concerns which appeared to be heightened by the lack of available information on paediatric MS. Parents emphasised a need for information that was tailored around the guidance and management of fatigue (e.g. "Is there something I should be doing?"). Some expressed specific needs, for example, as to whether they should be modifying activity, sleep or diet. Carroll et al. (2016) also noted that greater accessible and tailored educational information should be provided to teachers, as well as caMS and families, regarding the management of fatigue.

A need for more detailed information regarding treatment was also observed by some of the parents in Cross et al.'s (2019) study. They reported caMS experiencing unexpected adverse effects from treatment (e.g. bruising and scar tissue) that they were not told about by HCPs. This suggests that families were not provided with enough information prior to commencing treatment. Considering Cross et al.'s (2019) study, the authors suggest that those providing information to pMS caregivers need to ensure that methods are developed to answer families' questions and enable them to feel prepared for decision making around Disease Modifying Treatments (DMTs). They also highlight that resources specific to the needs of families caring for pMS are important in this respect. Similarly, Harris reported that some caregivers had difficulty communicating with younger patients, so the development resources aimed specifically at conveying this information to children may be beneficial.



## Summary of informational needs

- Many families report a lack of access to information or difficulty sourcing information
- Information is needed especially at the time of diagnosis, but also during DMT decision making and disease management, and what to expect from treatment
- Information should be provided in a way that families can understand
- The need for information may also include best advice on other practical issues, some of which are discussed in the next section.

“A better link between all the key stakeholders, such as doctors, neurologists, MS nurses to help caregivers to better understand the illness and its potential obstacles is key. More information immediately after diagnosis from the key stakeholders is crucial. Therefore, healthcare professionals should revisit how and what information is delivered to those affected by multiple sclerosis.”

Jarrad Kitson, parent and carer of Eleonor who was diagnosed with MS at 15 years old

# Results: Section 1

## Need for practical support

Harris (2018) reports how caregivers can experience several changes in their roles following diagnosis, which may be gradual changes or sudden/immediate changes. Either way, these changes often present challenges which may be difficult to navigate. **A common challenge for caregivers is balancing the needs of caring for their child's illness, as well as managing employment, family responsibilities and social obligations (Hinton & Kirk, 2017).**



Caring for a child with MS can therefore have many implications for family life. Various forms of practical supports may be of assistance to families as they adapt to their caregiving roles. Some examples of these are discussed below.

## Need to alleviate the burden of care

Uccelli et al. (2013) found that pMS parents reported reduced feelings of parental competence and were less satisfied with their parenting role than control parents, suggesting a need for additional supports to help cope with the demands of the disease. Cross et al. (2019) report that managing tasks such as arranging and travelling to appointments, as well as keeping track of vast amounts of information, can put a considerable strain on families. Similarly, O'Mahony et al. (2018) found that the family functioning of pMS parents was low, and, more generally, that parental health-related quality of life (HRQOL) was also low.

Financial burden appeared to be common for caregivers (Harris, 2018), partly as a result of costs associated with medications and travel to medical appointments. It was also not uncommon for carers to experience losses of earnings due to taking time off work for caring responsibilities, which may even lead to loss of employment in some cases (Cross et al., 2019). Families with lower socioeconomic status, or those with limited insurance may be at a particular disadvantage, especially when it comes to receiving an initial diagnosis (Hebert et al., 2019). For example, in 2018, Merck found that more than a third felt obliged to take time off work because of their caring responsibilities.

## Need for support surrounding symptom management and treatment

PMS caregivers may spend a lot of time helping their children with simple tasks, such as brushing teeth or combing hair (Harris, 2018), however, it is also noted that, owing to the fluctuating support needs of caMS, caring activities may vary considerably (Hinton & Kirk, 2017). This makes it difficult for parents to predict the daily care needs of their caMS (Hinton & Kirk, 2017), which can considerably impact the whole family's schedule. For example, one parent in Carroll et al.'s (2016) study noted that they had to plan everything around their child's levels of fatigue.

Treatment itself also presented a challenge for some families, especially regarding the management and administration of injections. In some cases, needle phobia emerged as an issue for both children and parents (Cross et al., 2019) and the task of administering medication and ensuring treatment compliance was reported as stressful for some (Harris, 2018).

## Need for wider family support

Cross et al. (2019) identified that caregiving can negatively impact relationships, causing problems in marriages and sibling relationships. Harris (2018) also reported cases of difficulties with siblings. For example, some siblings of caMS experienced worries, including whether the disease may be hereditary, while others experienced sibling animosity due to their parent spending more time with the caMS (Harris, 2018). Despite this, Cross et al. (2019) and Harris (2018) both reported that many parents felt that caring for pMS brought the family close together.



## Summary of practical needs

The research demonstrates pMS carers require a variety of practical support that may include:

- Financial support, e.g. to compensate from lack of earnings
- Assistance with management of care responsibilities
- Supports/interventions to combat injection anxiety and treatment adherence
- Family support to reflect and recognise how MS may affect the family dynamic, including relationships with partners and siblings

## Need for educational support

Given the range of cognitive difficulties that may be experienced by caMS, educational activities can be particularly challenging and navigating this system can be difficult for parents (Krupp et al., 2016). Parents in Carroll et al.'s (2016) study, for example, identified fatigue as a particular problem in school.

Cross et al. (2019) found that support varied significantly from school staff. While many caMS had individual education plans (IEP), not all schools were as accommodating. One caregiver in Harris' study reported that their school was not accommodating, even with an IEP in place. This may be due to a lack of awareness or unfamiliarity of MS from school personnel (Krupp et al., 2016).

Managing school communication was difficult too, especially when children were reluctant to share information (Carroll et al., 2016; Cross et al., 2019). Caregivers may feel obliged to take on a role that involves advocating for their child in school to prevent them from falling behind in their work (Harris, 2018), however, this may be a struggle. Many may not feel prepared to ask school for additional support (Krupp et al., 2016). One participant in Carroll et al.'s (2016) study noted that some teachers did not understand the impact that fatigue may have on caMS, although others noted that it was beneficial to disclose this to teachers. Cross et al (2019) suggest that school consultation liaisons could be provided by hospitals or MS societies to educate and inform schools about supporting caMS and their parents.

“ Our daughter was diagnosed with MS 12 months before her exams. This had a detrimental impact on her education because of the lack of support and guidance available to schools. This must be improved.”

Jarrad Kitson, parent and carer of Eleonor who was diagnosed with MS at 15 years old



## Summary of educational support needs

- Greater awareness of the needs of caMS in educational institutions would benefit pMS parents and caMS
- IEPs or similar could be developed depending on the caMS's needs

“ My daughter and I are happy with what the school did. But it cost me a lot of energy and effort to get the school system to work with us. She has had an individualised training plan throughout her years at school, and it has been helpful to stay focused on what has been important to her.

The occupational therapist was at the school and considered what should be arranged, the school followed this up well. However, there should be greater awareness of the needs faced by students with paediatric MS.”

Elin Katrine Vestly, parent and carer of Ragna-Elise who was diagnosed with MS at 11 years old.

### Need for social support

Families can feel isolated following an MS diagnosis (Krupp et al., 2016), and often express the desire to connect with others who have been through the same experience. Establishing social networks for families dealing with pMS can help to address this need.

Interestingly, Hinton and Kirk (2017) report that some parents found social interactions (e.g. with family and friends) to exacerbate negative experiences following diagnosis. One challenge is that, because caMS tend to appear visibly healthy, others may dispute the diagnosis, which also occurred when disclosing a diagnosis to teachers. This was echoed by Harris (2018), who identified that some caregivers found family members to lack understanding or compassion for the caMS.

Conversely, many other caregivers suggested that social support was beneficial, and a number of Cross et al.'s (2019) participants emphasised the value in seeking support from family and friends. In addition to the provision of information support, a number of participants in the studies analysed valued resources that enabled them to connect with others in the MS community. This included the availability of a phone line where questions could be asked (Cross et al., 2019), as well as connection to MS societies and online support groups (Hebert et al., 2019). Hebert et al. (2019) also noted that information on activities such as specialised camps for pMS was helpful. Such camps are particularly popular in the US and Canada (Krupp et al., 2016) and enable caMS to connect with others in the same situation.

In Harris' (2018) study, the centre from which the study participants were drawn was cited as a good source of support and advice, as well as providing opportunities for families to collaborate, network, and improve their educational knowledge of MS. Many used the national MS

- Social support from family and friends can be beneficial, but only if they are aware of the disease impact. Greater awareness of pMS in the wider public may help
- Support from MS communities, including connection with MS societies and others living with pMS via various media can be helpful for caregivers, as well as caMS

society as a further resource, however, an interesting example was given by one of Harris' (2018) participants who noted that many of the MS society events involved people at least 25 years older than her child and, for this reason, she was reluctant to engage. Another participant, however, identified a specific pMS support group as being useful. Parents in Cross et al.'s (2019) study also highlighted the benefits of engaging with social media (e.g. through a pMS caregiver Facebook page).

### Needs analysis evaluation and appraisal

As previously outlined, we employed the GRADE-Confidence in the Evidence from Reviews of Qualitative research (GRADE-CERQual) (Lewin et al., 2015; Lewin et al., 2018) to present an overall judgement of confidence in the review findings. When assessing the evidence using the four key criteria (i.e. methodological limitations, adequacy of the data, coherence of the review findings, and relevance of the studies to the review question), there were typically only minor concerns. We therefore conclude to have moderate confidence in the overall review findings presented. A rationale for this reasoning is provided in the table 8.

Table 8: Assessment of review using the GRADE-CERQual framework

GRADE-CERQual criteria	Concerns	Rationale
Methodological limitations	Minor concerns	Most studies were well-reported, although some studies did not consider the role of researcher reflexivity. Not all pMS caregivers may be represented by the studies
Coherence of the review findings	Very minor concerns	Study findings had typically high levels of coherence (e.g. highlighting common experiences and needs of pMS caregivers)
Adequacy of the data	Minor concerns	Most studies reported a rich amount of data that was relevant to the review
Relevance of the studies to the review question	No concerns	All studies explored some aspect of pMS caregiver experience so are relevant to the research question

GRADE-CERQual also recommends that individual study findings are evaluated using this framework. We present an assessment of confidence relating to the individual unmet needs in the table below. As can be seen here, given the converging evidence, we have a high level of confidence that there are needs for both psychological support and information among pMS caregivers. All studies included in the review highlighted these particular needs, with a high level of agreement among the study findings (e.g. that a common experience was lack of access to appropriate information, and a number of complex emotions, suggesting a need for greater psychological support). We can be moderately confident in relation to the other needs (i.e. needs for practical supports, educational support and social support). Although some convergent findings emerged in relation to these needs, not every study included in the review addressed all of these issues, which is why we exhibit a little more caution in relation to these results. More research is recommended to establish the extent of pMS caregiver needs in this respect.

Table 9: Assessment of confidence in individual review findings using CERQual framework

pMS caregiver need from review findings	Studies contributing to finding*	CERQual assessment of confidence	Explanation of CERQual assessment
Need for psychological support	1,2,3,4,5,6,7,8	High	High coherence and relevance; minor concerns about methodology and data
Need for information	1,2,3,4,5,6,8	High	High coherence and relevance; minor concerns about methodology and data
Need for practical support	1,2,3,4,6,7,8	Moderate	High relevance; moderate coherence; minor concerns about methodology and data
Need for educational support	1,2,3	Moderate	Moderate relevance; moderate coherence; minor concerns about methodology and data
Need for social support	2,3,4,6	Moderate	High relevance; moderate coherence; minor concerns about methodology and data

\* Studies numerically represented as follows: (1) Carroll et al (2016); (2) Cross et al. (2019); (3) Harris (2018); (4) Hebert et al. (2019); (5) Hinton & Kirk (2015); (6) Hinton & Kirk (2017); (7) O'Mahony et al (2018); (8) Uccelli et al (2013)

### Limitations and conclusion

It is important to acknowledge that the literature included in the review represents the views of a limited number of caregivers in largely English-speaking countries. There were no identified studies focusing on the experience of formal pMS caregivers pointing to potential research gaps in this area. In addition, most of the studies included in the review took a qualitative approach, which limits their generalisability. The two quantitative studies that were reported also involved small samples. Nevertheless, our review of the literature has highlighted some key recurring needs of pMS caregivers and their families.

Many of these needs are consistent with previously identified needs of general MS caregivers (particularly in relation to needs for information and psychological support), however, it also appears that many additional needs are experienced by this group, including specific needs around practical and educational supports.

There is much potential for these needs to be met by HCPs, MS societies, governmental bodies and the wider MS community, however, there may be variations in an individual country's capacity to meet these needs. In the next section, we detail the results of our survey which captures information on the supports and resources available to both general MS caregivers and pMS caregivers in several European countries. We will later discuss how these supports may be used to help address the needs identified above.

# Section 2: Survey results

## Prevalence of pMS, caregivers and pMS caregivers in countries surveyed

In order to contextualise the supports available for caregivers in the countries surveyed, we will first report on the awareness of rates of paediatric MS and caregiver numbers. As can be seen in Table 10, most respondents (n = 12; 71%) did not have access to accurate information on the number of pMS cases in their country, with 4 (25%) giving no information on the prevalence. Only 5 countries provided definitive numbers, with others providing rough estimates. Countries with more accurate data included Denmark (numbers ranged between 33-55, based on information in the Danish MS Registry), Germany (with 200 reported cases each year, based on data from the German Paediatric Surveillance Unit – ESPED), Lithuania (approx. 58 based on database records), Romania (with 44 undergoing treatment), and the Netherlands (60 cases. Note: although the source was not reported, it is known that the Netherlands has an MS registry).

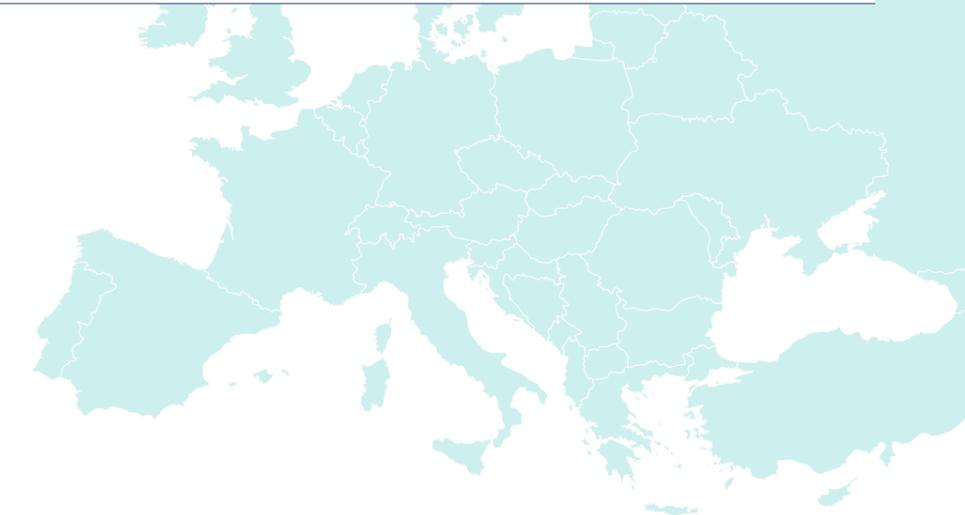
Many other countries provided indications of prevalence based on the proportion of total patients diagnosed with MS in their country (with rates of pMS typically acknowledged as between 5-10%) or of cases which were known to them based on contacts. This highlights the importance and value of registry data relating to MS, including pMS, in order to determine accurate estimates of prevalence.

In terms of caregiving estimates, almost none of the survey respondents were able to provide information on the number of MS caregivers overall, nor the number caring for pMS. A representative from the Polish MS society indicated that about 3,000 people with MS were defined as being incapable of working, which may give some indication of those undertaking care. Within Greece, a representative from the Hellenic Federation of Persons with MS provided an estimate of approximately 28,000-30,000. In terms of caregiving for pMS, one country (Poland) gave an estimate of 2,000 carers, which was based on the assumption of 2 parents per each estimated case of pMS. In contrast, Croatia gave a more conservative estimate of 10 caregivers. No other countries reported estimates.

This suggests that considerable information is lacking on the number of pMS cases as well as the number of caregivers in each country. Having access to such information would be important in terms of ascertaining the extent of support required for these different groups.

Table 10: Estimates of paediatric MS across countries

Country	Prevalence	Source
Belarus, Russia, Spain, Turkey	Unknown	–
Croatia	Estimate of 40	No registry – estimate based on information from colleagues
Denmark	33-50	Population based register
Germany	200 each year (or 5% of 240,000 MS Patients)	ESPED and MS Registry
Greece	Estimate of 750 (5% of MS patients)	Greece National Organization for Medicines
Ireland	20 known	Regional offices of MS Ireland
Italy	Estimate of 3,540-11,800 (or 3%-10% of MS patients)	Published estimate based on approximately 118,000 MS patients
Lithuania	Approx. 58	From database registered
Netherlands	60	–
Norway	Approx. 25-38	–
Poland	Estimate of 1000	Noted no accurate data available
Romania	44 (12-17 years old) with treatment	CNAS (National Health Insurance House)
Switzerland	0.5-1/100,000	–
UK	Estimate of 5-10% of MS patients	–



# Results: Section 2

## Provision of support for MS caregivers and pMS caregivers

Respondents were first asked to indicate whether there were any information programmes available to support MS caregivers generally in their country. This consisted of questions regarding whether information programmes were available (1) for informal MS caregivers (provided by any organisation other than health care professionals), (2) for informal MS caregivers provided by health care professionals, and (3) for formal MS caregivers. The same information was then requested on the provision of programmes for pMS caregivers specifically.

Figure 1 displays an overview of the number of countries reporting caregiver programmes according to the

various categories. One clear finding from this analysis is that there are far more programmes in place for MS caregivers more generally, relative to pMS caregivers specifically.

When analysing these responses according to country, it is clear that great variability in the availability of caregiver programmes (see Table 11). Notably three countries (Belarus, Greece and Russia) had no caregiver programmes in place. Poland also reported no programme for informal caregivers, with only one programme aimed at formal MS caregivers.

In contrast, the only country that reported programmes for all six categories of caregivers was the Netherlands. Other countries reporting specific programmes for pMS informal caregivers were Denmark, Italy, Norway, Romania and the UK.

Table 11: Information programmes available for informal caregivers in MS and pMS

	General caregiver info programmes			pMS caregiver info programmes		
	Informal MS carers	Informal MS carers HCPs	Formal MS carers	Informal pMS carers	Informal pMS carers HCPs	Formal pMS carers
Belarus						
Croatia	x		x			
Denmark	x			x		
Germany	x	x	x		x	
Greece						
Ireland	x	x	x			
Italy	x			x	x	
Lithuania	x					
Netherlands	x	x	x	x	x	x
Norway	x		x	x		
Poland			x			
Romania	x			x	x	
Russia						
Spain	x	x				
Switzerland	x	x	x		x	
Turkey	x	x	x			
UK	x		x	x		

Figure 1: Provision of programmes aimed at general MS caregivers and pMS caregivers specifically (from a total of 15 country responses)

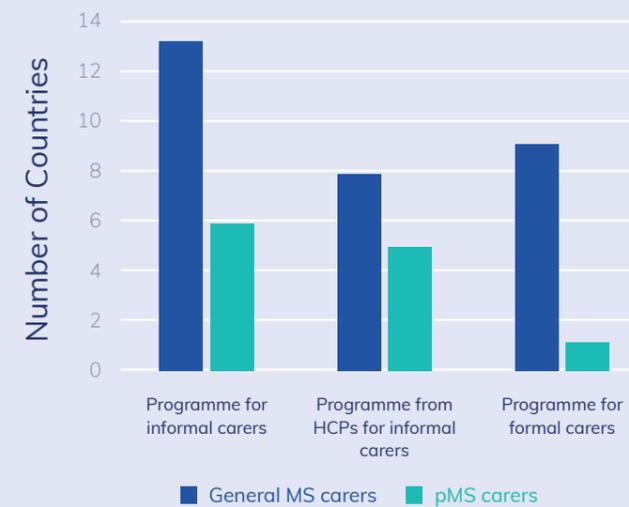
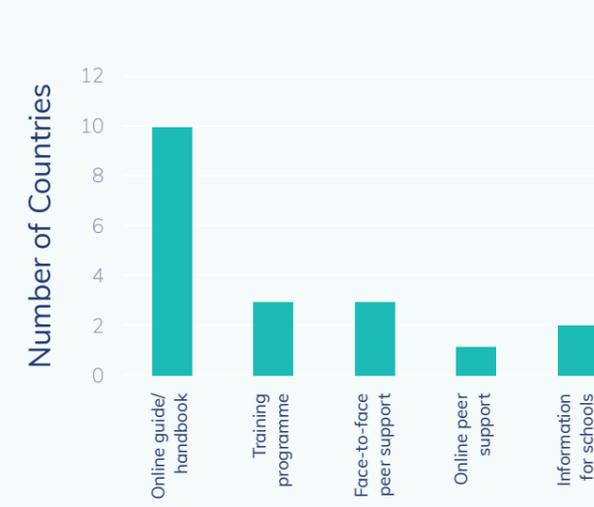


Figure 3: Specific supports for pMS caregivers (from a total of 13 country responses)

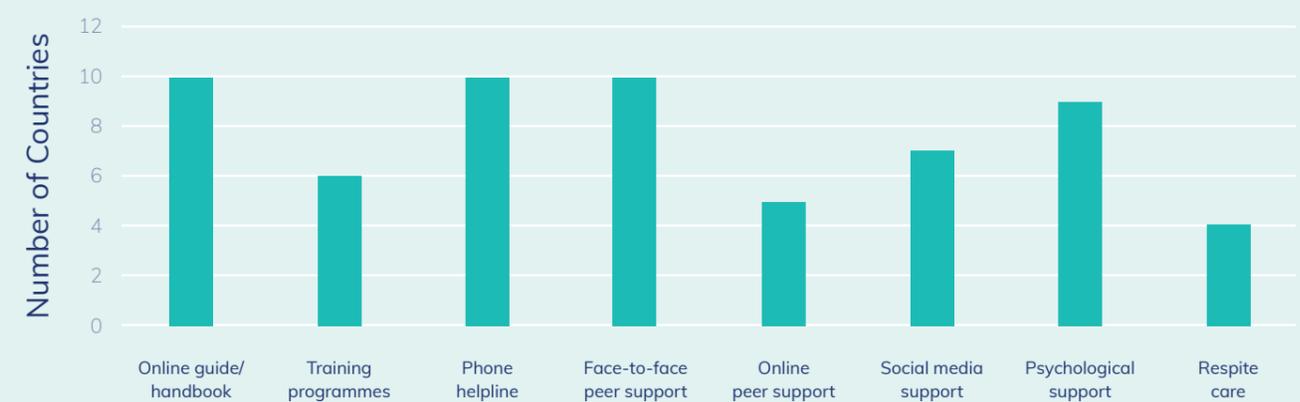


## Specific supports available

Respondents were also asked to provide more detail on the specific supports that were available to caregivers. Figure 1 provides an overview of the supports available for general caregivers, while figure 3 displays an overview of pMS caregiver support specifically. Table 12 provides more detail on the provision of each of these supports in the various countries. Note that in contrast to the information contained in Table 11, the Netherlands or Russia are not listed here due to incomplete data in this section of the questionnaire.

As reflected by the overall analysis, there were fewer supports for pMS caregivers overall, apart from the provision of an online guide or handbook for pMS caregivers, which was available in 10 of the respondent countries. Only 2 counties (Germany and Italy) indicated that there were guidelines in place specifically targeted at schools involved in the care/education of a young person with MS, however, Norway reported having individual training plans for students, which may apply to MS.

Figure 2: Specific supports for MS caregivers (from a total of 13 country responses)



# Results: Section 2

**Table 12:** Reported supports available for caregivers and pMS caregivers in countries surveyed (see key for explanation of support)

	General caregiver supports								pMS caregiver supports				
	Info guide	Carer training	Phone helpline	F2F support	Online support	Social media	Psych. services	Respite care	Info guide	Carer training	F2F support	Online support	School info
Belarus			x			x							
Croatia	x	x	x	x		x							
Denmark	x		x	x	x		x	x	x		x		
Germany	x	x	x	x		x	x		x				x
Greece	x			x		x	x		x				
Ireland	x	x	x	x		x	x						
Italy			x	x			x		x	x			x
Lithuania		x		x			x			x	x		
Norway	x		x	x	x		x	x	x				x
Poland	x		x				x		x				
Romania	x	x	x	x	x	x			x	x	x		
Spain	x						x		x				
Switzerland	x	x	x	x	x	x	x	x	x			x	
Turkey													
UK	x	x	x	x	x	x	x	x	x				

## Key



### Info guide

An online guide or a handbook on caring for someone with MS



### Carer training

Training programmes or interventions on how to care for someone with MS



### Phone helpline

A telephone helpline that MS caregivers can call for support



### F2F support

Face-to-face peer support groups with other MS caregivers



### Online support

Online peer support groups with other MS caregivers



### Social media

Social media supports (e.g. via Facebook MS groups)



### Psychological services

Psychological services or supports for MS caregivers



### Respite care

Respite care for MS caregivers (e.g. provision of alternative paid care to give caregivers a break from their caring responsibilities)

## Financial supports available

A series of questions were also asked regarding financial supports available to carers and patients. This information is displayed in Table 13. It is acknowledged, however, that this does not capture information on the extent of financial support available. Thus, for example, while nearly all countries responding to this section (bar Belarus and Turkey) reported some financial support for carers from the government, it is unclear as to whether this support marks up for loss of earnings from carer responsibilities. Only two countries reported some

employer support for carers, suggesting that across Europe, more could be done by employers to support those with caregiving responsibilities. The provision of this support however may also be shaped by the economic climate. A respondent from Greece for example noted that, while some financial supports were in place, the provision of many of these were negatively affected by the economic crisis, and that the government had recently been cutting down on financial support measures.

**Table 13:** Reported financial supports available in countries surveyed

	Carer government support	Carer employer support	Patient government support	Extra financial help	Support for travel costs	Flexible working arrangements	Sick leave	Child disability allowance
Belarus								
Croatia	x			x		x	x	
Denmark	x		x	x	x	x	x	x
Germany	x		x	x	x	x	x	x
Greece	x	x	x	x	x	x	x	x
Ireland	x		x	x		x	x	x
Italy	x		x			x	x	
Lithuania						x		
Norway	x		x	x	x	x	x	x
Poland	x		x		x		x	x
Romania	x		x	x	x		x	
Spain	x			x		x	x	x
Switzerland	x	x		x	x			x
Turkey								
UK	x		x	x	x	x	x	x

“ In Norway, we have good social care, which is important to a caregiver. I have been paid full salary when I have had to stay at home with my daughter for long periods of time. My employer has also been patient with my long absence. They have arranged for me to have a home office for a period of time and I attended meetings via Skype and telephone.”

Elin Katrine Vestly, parent and carer of Ragna-Elise who was diagnosed with MS at 11 years old.

# Results: Section 2

## Detailed descriptions of supports given

Respondents were also asked to describe the types of supports and resources available to caregivers in their country. Responses included links to websites offering information to caregivers, as well as descriptions of the supports available. Links to relevant websites are included at the end of this report and a description of some of the supports, as indicated by survey respondents, is provided in Table 14 overleaf.

As can be seen in Table 14, there are a variety of supports available for both general caregivers and pMS caregivers, which are mainly provided by national MS societies. However, as with our previous analysis, there are considerable variations across European countries. Notably, while countries such as Norway, Germany, Italy and the UK have supports in place for pMS caregivers, others, such as Poland, Turkey, Belarus, Greece and Croatia, appear to lack any substantial supports.

## Information on caregiver needs

Respondents were also asked whether they had ever conducted a survey investigating the needs of MS caregivers in their country. Of these, just two countries (Denmark and Italy) reported having done so, while another two countries (Spain and the UK) noted that surveys were ongoing. The Spain survey is also capturing information on caregivers of pMS, however, no other studies on the needs of caMS were reported.

## Survey summary

While we did not capture all resources available for MS and pMS caregivers across European countries, our analysis provides a good snapshot of the variety of supports available, as well as the lack of supports in certain countries. A key finding from our analysis is that there is considerable discrepancy in the range of supports available. It appears that there are regional differences in the provision of resources to support caregivers. This points to policy and action gaps, which are discussed in the final section.

## Other suggestions for supports and resources

The survey concluded by asking respondents if they had other suggestions regarding what supports are needed, or may be beneficial, for those caring for pMS. While most respondents did not reply to this question, there were some interesting suggestions. For example, a representative from Poland noted that psychological support would be beneficial for parents and whole families, including siblings and children with MS. This was echoed by the Romanian representative who noted that psychological support is mandatory, which is consistent with the conclusions from our literature review which suggest that all family members may require ongoing psychological support. An expert in paediatric MS from the Netherlands also provided suggestions that may be important to consider in the care of pMS, including coping with chronic disease, transition to adult care, future planning, education and relationships. These are topics which could be addressed as part of a focused information or training programme for caregivers. A representative from MS Ireland highlighted the potential of having an EMSP project to assist member state organisations in conducting a pan European survey of the needs of caregivers. They also noted that this may help in localising results which could increase capacity in member organisations. However, a representative from the Lithuanian MS society noted that, as caregivers lack time, it would be impossible to arrange more than 2-4 training seminars per year. They also suggested that such sessions might need to involve some form of payment in order to increase motivation to participate. The representative from Romania also noted that extra leave days, flexible programmes, personal budgets and facilities would be beneficial, in addition to greater provision of information and training programmes, as well as educational support/interventions.



Table 14: Descriptions of supports available for caregivers and pMS caregivers in Europe

Country	General caregiver resources and supports	pMS caregivers resources and supports
Belarus		
Croatia	<ul style="list-style-type: none"> <li>• SOS telephone line (MS society)</li> <li>• Different clinics provide education</li> <li>• Personal assistant project for non-profit organisations in Croatia (“osobni asistent”).</li> </ul>	<ul style="list-style-type: none"> <li>• Courses with social workers and psychologists</li> </ul>
Denmark	<ul style="list-style-type: none"> <li>• Information and courses with social workers and psychologists</li> <li>• Informal meeting with MS society representative and neurologist</li> </ul>	<ul style="list-style-type: none"> <li>• Courses with social workers and psychologists</li> </ul>
Germany	<ul style="list-style-type: none"> <li>• Programme delivered by the German MS society</li> <li>• Consultation available with each MS society chapter</li> <li>• Training courses for MS Nurse (“MS-Schwester”) and Caregivers of MS (“Pflege bei MS”) provided by MS society</li> <li>• Kompetenznetz Multiple Sclerosis provides further information and resources</li> </ul>	<ul style="list-style-type: none"> <li>• Supports and information available from DMSG website (German MS Society)</li> <li>• The Chapters of the German MS Society offer special groups for parents and young adults, where social workers of the MS societies give information concerning the support of the social and educational system in Germany.</li> </ul>
Greece		
Ireland	<ul style="list-style-type: none"> <li>• MS Ireland runs focus groups, information sessions, peer support and facilitated meetings</li> <li>• Symptom management programmes delivered by AHPs, emotional support programmes delivered by accredited professionals</li> <li>• MS Ireland Annual Conference</li> <li>• Private agencies provide accredited training in many aspects of care delivery (e.g. FETAC Level 5 training in Manual Handling, care of the elderly, infection control, physio therapy assistant etc)</li> </ul>	
Lithuania	<ul style="list-style-type: none"> <li>• Lithuanian MS Union organises 4 seminars for the informal caregivers every year in the different Lithuanian regions</li> <li>• There are “Baltic MS Nurses seminars “every year, organised by the rotations principle</li> </ul>	<ul style="list-style-type: none"> <li>• Seminars, workshops, face to face meetings with family members, psychological support groups</li> </ul>
Italy	<ul style="list-style-type: none"> <li>• Informational events provided by local MS society branches on topics of interest-based needs</li> </ul>	<ul style="list-style-type: none"> <li>• Information on topics including what is MS, medications, rehab, rights, emotions, school, etc.</li> <li>• Online video of/for adolescents with MS</li> </ul>
Netherlands	<ul style="list-style-type: none"> <li>• Yearly patient day with fun/informative programme</li> <li>• Yearly study group meetings for formal caregivers</li> </ul>	<ul style="list-style-type: none"> <li>• Ped MS Centre Website sponsored by MS research foundation</li> <li>• Booklet for MS patients in print</li> </ul>
Norway	<ul style="list-style-type: none"> <li>• Information from local branch of Norwegian MS society (including facts about MS, my life as a relative, peer work, to be a relative – growth and challenges, information about services from the community)</li> <li>• Some neurology departments arrange courses for adults with MS where relatives are invited to participate. MS nurses offer to talk with relatives including children of adults with MS</li> <li>• The EMSP project MS-Nurse pro (for formal caregivers) has been translated into Norwegian</li> </ul>	<ul style="list-style-type: none"> <li>• Oslo University Hospital, Children’s Department for Neurology and the MS Society collaborate on an information programme on topics such as: information about the MS organisation, diagnosis and treatment, cognitive and psychosocial challenges including fatigue, family perspective, rights, challenges in the family, school and education, workshops</li> <li>• The Norwegian MS competence centre has two online self-help programs – one for children and one for young people who have a parent with MS – devised by Norwegian psychologists</li> <li>• The Norwegian MS Society has a helpline operated by two parents</li> </ul>
Poland	<ul style="list-style-type: none"> <li>• No programmes for informal caregivers</li> <li>• Some branches of Polish MS Society employ and train assistants for MS patients with severe disabilities</li> </ul>	
Romania	<ul style="list-style-type: none"> <li>• Annual event for caregivers (organised by APAN in December)</li> <li>• Access to monthly meetings with patients (as well as some meetings having as the subject the family/ couple implications)</li> <li>• Several non-medical books and printed materials</li> <li>• Seminars organised directly by some pharma companies (eg. TEVA)</li> </ul>	<ul style="list-style-type: none"> <li>• In 2017 APAN translated the MSIF brochure in Romanian and distributed to the biannual conference of the Romanian Society of Pediatric Neurology, as well to the only MS Center specialising in Paediatric MS. The brochure is available in both printed and electronic formats.</li> <li>• APAN organises an annual meeting event for MS caregivers and children with MS. The event features a paediatric neurologist, child psychologist, psychologists for parents, and social and teambuilding support etc.</li> </ul>
Spain	<ul style="list-style-type: none"> <li>• Resources available from MS Society (Fundacio Esclerosi Multiple)</li> <li>• Cemcat Vall d Hebron University Hospital in Barcelona organise information sessions</li> </ul>	<ul style="list-style-type: none"> <li>• Different teaching programs in Cemcat include paediatric MS</li> </ul>
Switzerland	<ul style="list-style-type: none"> <li>• Resources available on MS Society website for both informal and formal caregivers</li> </ul>	<ul style="list-style-type: none"> <li>• Inselspital Bern provides training for formal caregivers</li> </ul>
Turkey	<ul style="list-style-type: none"> <li>• MS society provides tips on caregiving, and visits three times per week</li> <li>• Supports for helping caregivers to cope with daily problems</li> </ul>	
UK	<ul style="list-style-type: none"> <li>• A range of supports available on MS society website and through helpline</li> </ul>	<ul style="list-style-type: none"> <li>• Range of supports and information provided from MS society</li> </ul>

# Policy and action gaps

The results of both the rapid systematic review and the survey point to potential policy and action gaps that need to be addressed to better support paediatric MS caregivers. More broadly, this is consistent with initiatives aimed at supporting and empowering informal carers across Europe (Eurocarers, 2019).

Firstly, our analysis of the literature suggests that there are many key interlinked needs experienced by pMS caregivers and we can be moderately confident in these findings. These studies also offer some suggestions for ways in which needs may be met, however, it is evident that families of pMS experience considerable variation in their experiences of support. Because of this, caregivers may continue to exhibit unmet needs long beyond diagnosis. Examination of the survey results suggests that not all these needs may be successfully addressed in a European context. There appears to be considerable variation in the range of supports and resources available to pMS caregivers.

There are, however, examples of good practice taking place to support pMS caregivers in EMSP member organisations. For example, the Norwegian MS society provides comprehensive information and online self-help programmes to supplement support from HCPs. A further feature reported by the Norwegian MS society involves the helpline run by parents for parents of pMS, which is a good example of peer support in action. Many other countries provided further interesting example of programmes that are in place to support caregivers (e.g. as per Table 14). It may be beneficial for MS societies and HCPs to share these examples of good practice in order to help better support pMS caregivers.

In the final sections of this report, we will consider the extent to which the earlier identified needs are being met in the countries included in the survey. We will conclude with suggestions aimed at guiding the development of a set of recommendations to support caregivers of pMS.

## Addressing the need for psychological support

Receiving a diagnosis of MS can be a stressful experience for families, and this was evident from the literature review. Such a finding is also consistent with other studies in the general MS caregiving literature, where the emotional strain was reported as the biggest challenge for caregivers (e.g. Merck, 2018), however, parental caregivers of pMS may experience additional strain as they may feel the need to conceal their fears from their child.

In our survey, we asked societies and experts whether caregivers were able to avail of any psychological services in their country. Most respondents indicated that this was an option available (except for Belarus, Croatia, and Turkey), however, it is unclear whether all pMS caregivers are aware of these services, or indeed of the amount of support available for them. Another issue is the extent to which these services are effective in addressing the problems faced by caregivers. Further work that evaluates these services is therefore merited.

Psychological support may be particularly important around the time of diagnosis, as families may experience anxiety around treatment and progression. As is evident from the review of studies, the extent of emotional distress experienced may be influenced by the caregiver's knowledge of MS, or indeed the extent of information that is available to them. Therefore, addressing the need for information (detailed in the next section) may also assist in alleviating potential psychological distress.

Our review also found that interactions with HCPs were a crucial means of alleviating parental concerns. Having in place informational programmes from HCPs may therefore be beneficial, however, only four countries (Germany, Italy, the Netherlands and Switzerland) reported the presence of such programmes. Ensuring that families have enough time to raise their concerns with HCPs during consultations seems an important first step in providing psychological support.

Other less formal forms of support (such as social supports) may also help alleviate psychological distress, however, again there was considerable variation in the provision of these supports across European countries.

It therefore appears that, overall, caregiver needs for psychological support are not being adequately addressed in all European countries. Simple measures such as the provision of information, more time with HCPs during consultations, and social supports are potential ways of addressing these needs.

## Addressing the need for information

As previously mentioned, the provision of information for caregivers is particularly important to help alleviate emotional distress. This may help parents make sense of the pMS diagnosis, as well as aid in assisting them in understanding how best to manage their child's condition.

Our survey revealed that while there were information programmes in place for informal MS carers in many countries, only five countries reported information programmes specifically targeted at caMS caregivers. Given that parents of caMS may have particular concerns, finding ways to address their informational needs is particularly important.

On a positive note, many countries had some form of online guide or handbook aimed at helping those caring for pMS. Some examples of these guides are included at the end of this report. However, it is acknowledged

that such handbooks often entail the passive delivery of information, which may answer all the questions caregivers may have. Having the opportunity to actively raise concerns with HCPs and other experts may be particularly important, however, only one country (Italy) reported having a pMS caregiver training programme in place.

In addition, identifying ways to convey information to caMS in an age-appropriate fashion is important. While some child-friendly resources exist, they may not be available in all languages.

Eastern European countries appear to be at a disadvantage in the provision of information. For example, Belarus, Croatia and Turkey did not have any information in place relating to caring for pMS. Meeting the informational needs of carers should therefore be a key priority of those involved in providing support for this group.

“ We have always experienced the support of health professionals, doctors and nurses, which is crucial. My daughter had an aggressive development of her MS and new outcomes that were not treated satisfactorily. When I expressed our desire for a specific treatment, I felt doctors heard me, even though there was no approved treatment for children and recently approved treatments for adults. I got to participate in the discussion of opportunities, and we got good information.

There was a lot of work and contacts with specialists in other countries before she was treated with a specific treatment as the first child in Norway. We have always experienced being listened to, doctors have always put in plenty of time for questions in the consultations and we have had good cooperation and opportunity to participate in treatment. They have also been good at approaching her so that she can tell them how she is doing. It has strengthened our confidence in the health care system.

Elin Katrine Vestly, parent and carer of Ragna-Elise who was diagnosed with MS at 11 years old.

# Policy and action gaps

## Addressing the need for practical support

Our review found that practical supports are often necessary to help caregivers manage their caring responsibilities effectively.

As caring can be associated with financial burdens, or even employment loss, having financial supports in place is particularly important. While many countries in our survey reported having some form of government support for carers, the extent of this support is unclear, and it is unlikely to fully compensate for the costs associated with care. Overall, there appear to be wide variations in the financial supports provided. Some countries, especially those in Eastern Europe reported very few supports, with none in the case of Belarus and Turkey.

Helping caregivers manage their care responsibilities may be another important form of practical support. One example of this is through respite care, so that carers may have a break from their duties. Such support may also help alleviate the emotional strain of caring, thereby partially addressing unmet needs for psychological support. Only four countries (Denmark, Norway, Switzerland and the UK) reported that respite care was an option available to carers. This again points to clear inequalities across European countries in the provision of practical supports for carers.

Our review also found that a commonly reported problem for pMS caregivers surrounded medication administration and adherence. While this is another example of a practical support need for families, this is likely to be at least partially addressed through appropriate information and training from HCPs. Ongoing work in this area (Ghezzi et al., in preparation) suggests that parents who are actively involved in a programme that evaluates QoL and acceptance of DMTs may be beneficial in this respect. As discussed previously, however, there are inconsistencies in the availability of such programmes for pMS caregivers.

A final practical need uncovered from our review was the need to support all family members as they learn to adapt to the MS diagnosis. As caring for pMS may impact on family relationships, it is important that support is made available to siblings and others who may be directly affected. Again, this need may be met through psychological support, appropriate information, and social support.

## Addressing the need for educational support

One key specific need relating to the care of pMS requires tailored support in an educational context. School difficulties are reported in nearly all studies describing cognitive impairment in pMS. Fatigue is a common symptom, potentially impairing both attention and executive functions. This may also have an impact on longer-term cognitive functioning, as adolescents transition into adulthood.

Most, if not all, caMS are in full-time education, however, it is clear from the results of our survey that few countries reported specific guidelines aimed at schools in the education or care of caMS. This finding highlights an important policy gap. The literature review revealed that many parents struggled with school communication and that there were considerable variations in the availability of support. This may be a particular difficulty given the invisible nature of many MS symptoms, coupled with the caMS's concerns surrounding peer relationships. One simple solution may involve informing school personnel and teachers about the problems associated with pMS. Addressing needs for educational support is crucial for both caregivers and children/adolescents themselves.

## Addressing the need for social support

Finally, another important need expressed by pMS caregivers was for greater social support. Many of the studies included in the review highlighted the considerable benefits of support from both friends and family members, as well as from those in the MS community. Some parents, however, reported misunderstandings and misconceptions about MS among family and friends, pointing to a greater need for awareness of MS among the general public.

The provision of social support from others in the MS community was particularly helpful for pMS caregivers, including making connections with others via social media platforms. In our survey, we found that some countries offered face-to-face peer support for general caregivers, as well as opportunities to connect with others via social media, however, only one country (Denmark) noted that this form of support was provided specifically for pMS caregivers. Switzerland was the only country reporting online peer support for pMS caregivers. Interestingly, the provision of online support was quite low among the countries surveyed, however, this is something that may be relatively easy to implement. Given that these forms of support are particularly useful in addressing other needs of pMS caregivers, identifying ways to introduce these should be a key priority.

## Conclusion

Many adults diagnosed with MS can live well independently and often do not require significant care from others, especially in cases where their condition is successfully managed. In contrast, those diagnosed with MS in childhood or adolescence require ongoing care from their parents or guardians. The mean onset time for paediatric MS is between 14-15.7 years (Hebert et al., 2019), which can be a turbulent time for any teenager as they cope with a range of psychosocial and educational issues. A diagnosis of MS can bring additional challenges, including symptoms such as fatigue and cognitive difficulties. Caregivers must help caMS navigate these challenges at home and in school, in addition to maintaining their own psychological wellbeing. While caring can often be a positive experience for caregivers, caring for pMS can place considerable strain on parents.

Our review of the literature suggests that various needs may be commonly experienced by caregivers, particularly relating to psychological support and information. It is vital that policy makers take steps to address these needs to ensure the ongoing wellbeing of pMS caregivers, as well as their care recipients. This report has revealed considerable variation in the supports available for pMS caregivers. We have also highlighted how little is known about the needs of pMS caregivers in Europe. More focused research in this area should help further our understanding of policy gaps which may ultimately lead to a core set of recommendations for how best to support caregivers.

# Relevant links and further information

A number of resources provided by MS societies are listed below. In addition, we also obtained other resources available for pMS caregivers using a combination of an online search and from information directly passed on by experts in the area. It should be noted, however, that one limitation of this search was that results were generally confined to those in the English language.

## Canada

Kids get MS too: A guide for parents with a child or teen with MS <https://mssociety.ca/en/pdf/kidsgetmstoo.pdf>

## Eurocarers

Inform Care <https://eurocarers.org/portfolio-item/informcare/>

## Germany

DMSG <https://www.dmsg.de/>

## Germany

Kompetenznetz Multiple Sclerosis <https://www.kompetenznetz-multiplesklerose.de/en/>

## Ireland

MS Ireland <https://www.ms-society.ie/>

## International Paediatric Study Group

<http://www.ipmssg.org/>

## Spain

Fundacio Esclerosi Multiple <https://www.fem.es/>

## Switzerland

MS society [https://www.multiplesklerose.ch/de/unsere-angebote/beratungen/medizin-und-pflegerberatung/#a\\_pflegerische-beratung-fuer-angehoerige](https://www.multiplesklerose.ch/de/unsere-angebote/beratungen/medizin-und-pflegerberatung/#a_pflegerische-beratung-fuer-angehoerige)

## MS International Federation

Childhood MS: A guide for parents <https://www.msif.org/wp-content/uploads/2016/06/Childhood-MS-A-Guide-for-Parents-email-version.pdf>

## Netherlands

KinderCentrum [www.kinderscentrum.nl](http://www.kinderscentrum.nl)

## Norway

Information for parents of caMS <https://www.ms.no/leve-med-ms/barn-og-unge-under-18-aar-med-ms>

## UK

Carer supports <https://www.mssociety.org.uk/care-and-support/support-for-carer>

## UK

Supports from caring for caMS <https://www.mssociety.org.uk/about-ms/what-is-ms/children-and-ms>

## UK

Digesting Science: activities aimed at teaching children about MS <http://digestingscience.co.uk/>

## UK

UK & Ireland Childhood CNS Inflammatory Demyelination Working Group <http://www.childdemyelination.org.uk/>

## USA

National Institute of Neurological Disorder and Stroke: Patient and Caregiver Information for MS <https://www.ninds.nih.gov/Disorders/Patient-Caregiver-Education/Hope-Through-Research/Multiple-Sclerosis-Hope-Through-Research>

# References

Amato, M. P., Goretti, B., Ghezzi, A., Hakiki, B., Nicolai, C., Lori, S., . . . Patti, F. (2014). Neuropsychological features in childhood and juvenile multiple sclerosis: five-year follow-up. *Neurology*, 83(16), 1432-1438.

Appleton, D., Robertson, N., Mitchell, L., & Lesley, R. (2018). Our disease: a qualitative meta-synthesis of the experiences of spousal/partner caregivers of people with multiple sclerosis. *Scandinavian journal of caring sciences*, 32(4), 1262-1278.

Banwell, B., Arnold, D. L., Tillema, J.-M., Rocca, M. A., Filippi, M., Weinstock-Guttman, B., . . . Sormani, M. P. (2016). MRI in the evaluation of paediatric multiple sclerosis. *Neurology*, 87(9 Supplement 2), S88-S96.

Carroll, S., Chalder, T., Hemingway, C., Heyman, I., & Moss-Morris, R. (2016). "It feels like wearing a giant sandbag." Adolescent and parent perceptions of fatigue in paediatric multiple sclerosis. *Eur J Paediatr Neurol*, 20(6), 938-945. doi:10.1016/j.ejpn.2016.06.004

Carroll, S., Chalder, T., Hemingway, C., Heyman, I., & Moss-Morris, R. (2016). Understanding fatigue in paediatric multiple sclerosis: a systematic review of clinical and psychosocial factors. *Developmental Medicine & Child Neurology*, 58(3), 229-239.

Chitnis, T., Glanz, B., Jaffin, S., & Healy, B. (2009). Demographics of pediatric-onset multiple sclerosis in an MS center population from the Northeastern United States. *Multiple Sclerosis Journal*, 15(5), 627-631.

Cooke, A., Smith, D., & Booth, A. (2012). Beyond PICO: the SPIDER tool for qualitative evidence synthesis. *Qualitative health research*, 22(10), 1435-1443.

Corry, M., & While, A. (2009). The needs of carers of people with multiple sclerosis: a literature review. *Scandinavian journal of caring sciences*, 23(3), 569-588.

Cross, T. P., Shanks, A. K., Duffy, L. V., & Rintell, D. J. (2019). Families' Experience of Pediatric Onset Multiple Sclerosis. *Journal of Child & Adolescent Trauma*, 1-11.

Duignan, S., Brownlee, W., Wassmer, E., Hemingway, C., Lim, M., Ciccarelli, O., & Hachohen, Y. (2019). Paediatric multiple sclerosis: a new era in diagnosis and treatment. *Developmental Medicine & Child Neurology*.

Figved, N., Myhr, K.-M., Larsen, J.-P., & Aarsland, D. (2007). Caregiver burden in multiple sclerosis: the impact of neuropsychiatric symptoms. *Journal of Neurology, Neurosurgery & Psychiatry*, 78(10), 1097-1102.

Ghezzi, A., Bianchi, A., Baroncini, D., Bertolotto, A., Malucchi, S., Bresciamorra, V., . . . Patti, F. (2017). A multicenter, observational, prospective study of self-and parent-reported quality of life in adolescent multiple sclerosis patients self-administering interferon-1a using RebiSmart™—the FUTURE study. *Neurological Sciences*, 38(11), 1999-2005.

Goretti, B., Ghezzi, A., Portaccio, E., Lori, S., Zipoli, V., Razzolini, L., . . . Viterbo, R. (2010). Psychosocial issue in children and adolescents with multiple sclerosis. *Neurological sciences*, 31(4), 467-470.

Harris, Y. C. (2018). A Qualitative Descriptive Study Exploring the Adaptation of Families of Children with Multiple Sclerosis from the Perspective of Caregivers. The University of Alabama at Birmingham.

Hebert, D., Geisthardt, C., & Hoffman, H. (2019). Insights and Recommendations From Parents Receiving a Diagnosis of Pediatric Multiple Sclerosis for Their Child. *Journal of child neurology*, 0883073819842420.

Hinton, D., & Kirk, S. (2015). Paediatric multiple sclerosis: a qualitative study of families' diagnosis experiences. *Archives of disease in childhood*, 100(7), 623-629.

Hinton, D., & Kirk, S. (2017). Living with uncertainty and hope: A qualitative study exploring parents' experiences of living with childhood multiple sclerosis. *Chronic illness*, 13(2), 88-99.

Horner, R. (2013). Interventions for children coping with parental multiple sclerosis: a systematic review. *Journal of the American Association of Nurse Practitioners*, 25(6), 309-313.

Jensen, M. P., Liljenquist, K. S., Bocell, F., Gammaitoni, A. R., Aron, C. R., Galer, B. S., & Amtmann, D. (2017). Life impact of caregiving for severe childhood epilepsy: results of expert panels and caregiver focus groups. *Epilepsy & Behavior*, 74, 135-143.

Krupp, L. B., Rintell, D., Charvet, L. E., Milazzo, M., & Wassmer, E. (2016). Pediatric multiple sclerosis: perspectives from adolescents and their families. *Neurology*, 87(9 Supplement 2), S4-S7.

Lewin, S., Booth, A., Glenton, C., Munthe-Kaas, H., Rashidian, A., Wainwright, M., . . . Garside, R. (2018). Applying GRADE-CERQual to qualitative evidence synthesis findings: introduction to the series. In: *BioMed Central*.

Lewin, S., Glenton, C., Munthe-Kaas, H., Carlsen, B., Colvin, C. J., Gülmezoglu, M., . . . Rashidian, A. (2015). Using qualitative evidence in decision making for health and social interventions: an approach to assess confidence in findings from qualitative evidence syntheses (GRADE-CERQual). *PLoS medicine*, 12(10), e1001895.

MacAllister, W. S., Christodoulou, C., Milazzo, M., Preston, T. E., Serafin, D., Krupp, L. B., & Harder, L. (2013). Pediatric Multiple Sclerosis: What we know and where are we headed? *Child Neuropsychology*, 19(1), 1-22.

McKeown, L., Porter-Armstrong, A., & Baxter, G. (2003). The needs and experiences of caregivers of individuals with multiple sclerosis: a systematic review. *Clinical rehabilitation*, 17(3), 234-248.

Murphy, N. A., Christian, B., Caplin, D. A., & Young, P. C. (2007). The health of caregivers for children with disabilities: caregiver perspectives. *Child: care, health and development*, 33(2), 180-187.

Ouzzani, M., Hammady, H., Fedorowicz, Z., & Elmagarmid, A. (2016). Rayyan—a web and mobile app for systematic reviews. *Systematic reviews*, 5(1), 210.

O'Mahony, J., Marrie, R. A., Laporte, A., Bar-Or, A., Yeh, E. A., Brown, A., . . . Banwell, B. (2018). Pediatric-onset multiple sclerosis is associated with reduced parental health-related quality of life and family functioning. *Multiple Sclerosis Journal*, 1352458518796676.

Pace, R., Pluye, P., Bartlett, G., Macaulay, A. C., Salsberg, J., Jagosh, J., & Seller, R. (2012). Testing the reliability and efficiency of the pilot Mixed Methods Appraisal Tool (MMAT) for systematic mixed studies review. *International journal of nursing studies*, 49(1), 47-53.

Pakenham, K. I., & Cox, S. (2012). The nature of caregiving in children of a parent with multiple sclerosis from multiple sources and the associations between caregiving activities and youth adjustment overtime. *Psychology & Health*, 27(3), 324-346.

Raina, P., O'Donnell, M., Rosenbaum, P., Brehaut, J., Walter, S. D., Russell, D., . . . Wood, E. (2005). The health and well-being of caregivers of children with cerebral palsy. *Pediatrics*, 115(6), e626-e636.

## For citing this report, please use the following:

Maguire, R., Kasilingam, E. & Kriauzaitė, N. (2019). *Caring for children and adolescents with multiple sclerosis: Exploring the unmet needs and existing supports for paediatric multiple sclerosis caregivers*. European Multiple Sclerosis Platform.

## For further information:

Rebecca Maguire, Department of Psychology, Maynooth University, Ireland  
Email: [rebecca.maguire@mu.ie](mailto:rebecca.maguire@mu.ie)

Nora Kriauzaitė, European Multiple Sclerosis Platform  
Email: [programme.coordinator@emsp.org](mailto:programme.coordinator@emsp.org)



## Acknowledgements

We would like to thank partners and, in particular, **Angelo Ghezzi and Gurli Vagner** for their helpful feedback on the content of this report. We would also like to thank all the EMSP Member Organisations who facilitated the collection of data.

We are also grateful to our sponsors who supported EMSP project – Caregivers of paediatric MS – within which this report was produced. EMSP, however, is solely responsible for the content of this report.

## Sponsors and supporters



## Appendix

### Survey on services and supports for caregivers of paediatric MS

#### Part A- Organisation details and background

Are you:

- a) Representative of a national MS society
- b) An expert in the area of paediatric MS
- c) Other (specify)

If yes, to (a) above, please give:

The name of your organisation   
 The country in which you are based

If yes to (b) or (c) above, please provide:

Your affiliation   
 Area of expertise in relation to paediatric MS:

If known, what is the approximate number of paediatric MS patients (up to 18 years old) in your country?

If known, what is the approximate number of caregivers of MS patients in your country?

If known, what is the approximate number of caregivers of paediatric MS patients in your country?

## Survey on services and supports for caregivers of paediatric MS

### Part B- General overview of resources and supports available for MS caregivers and paediatric MS caregivers

#### B1.1

Is there any information programme for MS informal caregivers (e.g. family members or friends) in your country, other than the information provided by healthcare professionals?

- Yes
- No
- Not known

#### B1.2

If yes, please indicate who delivers this programme (indicate all that apply):

- MS society
- A state authority
- Health insurers
- Other bodies (please specify)

#### B1.3

If yes, please provide details of the programme(s) available;

#### B1.4

Is there any information programme for MS formal caregivers (i.e. professionals) in your country?

- Yes
- No
- Not know

#### B1.5

If yes, please indicate who delivers this programme (indicate all that apply):

- MS society
- A state authority
- Health insurers
- Other bodies (please specify)

#### B1.6

If yes, please provide details of the programme(s) available;

#### B2.1

Is there any information programme specifically designed for informal caregivers of those with paediatric MS (e.g. parents) in your country, other than that the information provided by healthcare professionals?

- Yes, there is a specific programme for paediatric MS caregivers
- No, but there is a programme for general MS caregivers
- No
- Not known

#### B2.2

If yes, please indicate who delivers this programme (indicate all that apply):

- MS society
- A state authority
- Health insurers
- Other bodies (please specify)

#### B2.3

If yes, please provide details of the programme(s) available;

#### B2.4

Is there an information programme specifically designed for formal caregivers (i.e. professionals) of those with paediatric MS in your country?

- Yes, there is a specific programme for paediatric MS caregivers
- No, but there is a programme for general MS caregivers
- No
- Not known

#### B2.5

If yes, please indicate who delivers this programme:

- MS society
- A state authority
- Health insurers
- Other bodies (please specify)

#### B2.6

If yes, please provide details of the programme(s) available;

## Survey on services and supports for caregivers of paediatric MS

### Part C- Details on specific supports available for MS caregivers and paediatric MS caregivers

To your knowledge, which of the following supports are available for MS caregivers in your country? Indicate all that apply:

- An online guide or handbook on caring for someone with MS
- An online guide or handbook on caring for a child or adolescent with MS
- Training programmes or interventions on how to care for someone with MS
- Training programmes or interventions on how to care for a child or adolescent with MS
- A telephone helpline that MS caregivers can call for support
- Face-to-face peer support groups with other MS caregivers
- Face-to-face peer support groups with other paediatric MS caregivers
- Online peer support groups with other MS caregivers
- Online peer support groups with other paediatric MS caregivers
- Social media supports (e.g. via Facebook MS groups)
- Psychological services or supports for MS caregivers
- Respite care for MS caregivers (e.g. provision of alternative paid care to give caregivers a break from their caring responsibilities)
- Financial supports for MS caregivers from the government
- Financial support for MS caregivers from employers
- Financial support for the person cared for from the government
- Extra financial help, such as disability allowance and/or assistance for hardship
- Financial support for travel costs
- The right to request flexible working arrangements from employers
- Sick leave entitlements
- Child disability allowance
- Information or guidelines for schools involved in the education of a child or adolescent with MS
- Any other forms of support for MS caregivers (specify):
- Any other forms of support for paediatric MS caregivers (specify):

### Part D- Needs of MS caregivers and needs of caregivers of paediatric MS patients

#### D1.1

Has your country undertaken a survey on the needs of MS caregivers?

- Yes
- No
- Not known

#### D1.2

If yes, please provide the following details: number of respondents to the survey, the year of the survey, the link to the survey.

#### D1.3

If yes and available, please provide the survey questionnaire and survey results to [programme.coordinator@emsp.org](mailto:programme.coordinator@emsp.org)

#### D1.4

Are there any other articles/resources/initiatives on the needs of MS caregivers in your country? Please provide details, below.

#### D1.5

Have you any other suggestions regarding what supports are needed, or may be beneficial, for MS caregivers?

#### D2.1

Has your country undertaken a survey on the needs of caregivers of paediatric MS patients?

- Yes
- No
- Not known

#### D2.2

If yes, please provide the following details: number of respondents to the survey, the year of the survey, the link to the survey.

#### D2.3

If yes and available, please provide the survey questionnaire and survey results to [programme.coordinator@emsp.org](mailto:programme.coordinator@emsp.org)

#### D2.4

Are there any other articles/resources/initiatives on the needs of caregivers of paediatric MS patients in your country? Please provide details below.

#### D2.5

Have you any other suggestions regarding what supports are needed, or may be beneficial, for those caring for a child/adolescence with MS?



EUROPEAN  
MULTIPLE SCLEROSIS  
PLATFORM

Dedicated to International Children's Day 2019