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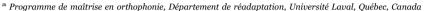
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Language disorders in multiple sclerosis: A systematic review

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ABSTRACT

Background: Although cognitive impairments are common in patients with multiple sclerosis (MS), language impairments are not well defined.

Objective: The goal of this review was to examine the presence and nature of language disorders associated with MS.

Method: A search of PubMed and PsycINFO databases was conducted using combinations of the following terms: language disorders, language pathology, language impairment, multiple sclerosis, communication, language and speech. Studies were chosen based on the original language of the text, year of publication, peer-review status and specificity of the results regarding language and communication disorders.

Results: This review covers 30 articles from 13 countries. The studies involved patients with different types of MS. Various language impairments were reported in MS. However, since the methods used in the studies varied widely, it is difficult to draw any conclusions thus far.

Conclusion: Given the various cognitive deficits in MS, it is expected that higher language abilities would be affected. This aspect should be investigated in future studies.

1. Introduction

According to the Multiple Sclerosis International Federation, about 2,300,000 people worldwide suffered from some form of multiple sclerosis in 2015 (Multiple sclerosis federation, 2016). Multiple sclerosis (MS) is thought to be an autoimmune maladaptive disease that affects the axons of the white matter in the nervous system by destroying the myelin that insulates them (Henry and Beatty, 2006). The disease primarily affects females from 45 to 65 years of age (Statistics Canada, 2016) and manifests itself by acute symptoms known as "attacks" (Cosh and Carslaw, 2014). These "attacks" can be intermittent, with partial or complete recovery of physical and mental functions (relapsing-remitting MS), or functions can be affected on a regular basis and may become progressively undermined (primary progressive MS) (Lublin, 2014). The most common symptoms are sensorimotor or physical and include fatigue, sensory deficits (especially visual), numbness, pain, weakness, spasticity, and balance problems. Communication difficulties are also frequently observed and present most often as motor disorders, called dysarthria (Hartelius et al., 2000). Cognitive deficits have also been reported in many studies, and mostly affect working memory, executive functions, attention, information processing speed, visual perceptual skills and long-term memory (Rao, 1995). Until recently, few studies examined

deficits in other higher cognitive functions such as language in MS patients (Rao, 1995). Language is the cognitive ability to communicate and manipulate symbols. It should be distinguished from speech, which is a motor ability. A number of contemporary studies have investigated the symptoms associated with language disorders in multiple sclerosis. Because of a better understanding and more detailed documentation, speech-motor deficits are now receiving even more attention, as in a virtuous cycle. Speech-language pathologists usually prioritize these symptoms and overlook the potential language difficulties that individuals with MS may experience. However, language impairments can have a major impact on quality of life as well as on the efficiency of rehabilitation. Therefore, identifying and characterizing language disorders associated with MS is very valuable to frontline clinicians as well as rehabilitation professionals. The aim of this article was to review the most significant studies investigating the nature of language impairments in individuals diagnosed with MS.

2. Method

2.1. Review protocol

As a guideline for conducting this review, we used the PRISMA statement for reporting systematic reviews and meta-analyses of

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studies that evaluate health care interventions (Liberati et al., 2009). The PRISMA statement consists of a 27-item checklist and a fourphase flow diagram designed to help authors improve the reporting of systematic reviews and meta-analyses.

2.2. Eligibility criteria

To be included in this review, studies had to: 1) be conducted with participants with MS; 2) focus on language and communication disorders in adults with MS, 3) be published in peer-reviewed journals, and 4) be written in English or French. We excluded studies if they: 1) targeted general cognitive skills; 2) targeted dysarthria and orofacial manifestations, and 3) were published after 1990, considering the uncertainty about the clinical diagnosis of MS before that date (Poser and Brinar, 2001).

2.3. Information sources and search strategy

We identified studies by searching PubMed and PsycINFO with the following keywords used in combination: multiple sclerosis, language, speech (the keyword "speech" was used since the distinction between speech and language is not consistently made in studies), communication, language disorders, language pathology and language impairment. We also reviewed the reference lists of all identified articles for additional studies. The literature search was conducted between May and November 2015.

2.4. Study selection

Two people (LMS, SR) screened titles and abstracts of studies for potential eligibility. Full texts of potentially eligible studies were then retrieved to ensure they met the inclusion criteria and could be included in this review. The agreement on excluded and included studies was 100%.

3. Results

3.1. Study selection

Nine hundred and thirty-eight (938) studies were identified in PubMed and 207 in PsycINFO for a total of 1145 studies. After excluding 16 duplicates, the titles and abstracts of 1129 studies were screened for eligibility, and 110 were earmarked for closer reading. Based on the eligibility criteria, 88 articles were excluded, leaving 22. Eight additional studies were selected from the reference lists of the selected articles, giving a total of 30 studies included in this review. Fig. 1 shows the article selection process flowchart..

3.2. Main characteristics of studies

Data items were first extracted using a chart containing the following variables: number of participants, country of origin, diagnosis, mean age, sex, education, duration of disease and tests used to establish diagnosis. These data are summarized in Table 1.

For each of the studies, data related to language tests and results, and main conclusions were then gathered. The results of this in-depth analysis were compiled in a summary table (see Table 2).

The studies were conducted in 13 different countries (Australia, Brazil, Canada, Finland, France, Germany, Italy, The Netherlands, Spain, Sweden, Turkey, UK and USA). Four case studies were included in the analyses (Moreno et al., 2013; Demirkiran et al., 2006; Zarei et al., 2003; Wallace and Holmes, 1993). The other studies had sample sizes ranging from 10 (Ruchkin et al., 1994) to 461 patients (Andrade et al., 1999; Beatty and Monson, 1990; Bensa et al., 2006; Blackwood et al., 1991; Connick et al., 2013; Friend et al., 1999; Grossman et al., 1995; Klonoff et al., 1991; Kraus et al., 2005; Kujala et al., 1996; Kusçu

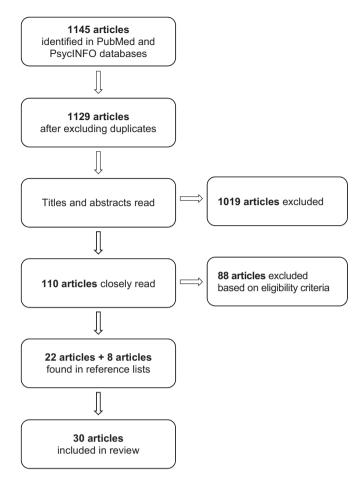


Fig. 1. Article selection process flowchart.

et al., 2012; Laatu et al., 1999; Lacour et al., 2004; Lechner-Scott et al., 2010: Lethlean and Murdoch, 1994: Lethlean and Murdoch, 1997: Mackenzie and Green, 2009; Nocentini et al., 2001; Nocentini et al., 2006; Olivares et al., 2005; Ruggieri et al., 2003; Ryan et al., 1996; Ryan et al., 2012; Tallberg and Bergendal, 2009). In all, 1745 MS patients were included in this review. Twenty-three studies had a control group. In addition to language abilities, cognitive functions were assessed in 20 of the studies for the purpose of establishing formal links between language and cognition or to characterize the clinical population (Moreno et al., 2013; Zarei et al., 2003; Ruchkin et al., 1994; Andrade et al., 1999; Bensa et al., 2006; Blackwood et al., 1991; Connick et al., 2013; Grossman et al., 1995; Klonoff et al., 1991; Kraus et al., 2005; Kujala et al., 1996; Kusçu et al., 2012; Lechner-Scott et al., 2010; Nocentini et al., 2001; Nocentini et al., 2006; Olivares et al., 2005; Ruggieri et al., 2003; Ryan et al., 1996, 2012; Tallberg and Bergendal, 2009). The functions were assessed with various instruments, including tests batteries (e.g., Frontal assessment battery; WAIS) and specific tests (e.g., Rey auditory verbal learning test; Wisconsin Card sorting test).

Twenty-five studies found language deficits in MS patients, two studies reported mixed results (language deficits in a minority of patients) and three studies found no language deficits, although MS patients performed more slowly than controls. Generally, the types of deficits differed widely across the studies. Patients often showed deficits affecting verbal fluency, naming, comprehension or semantic processing. In addition, language impairments were often associated with a general cognitive decline (executive deficits in particular). Language deficits also appeared to be dependent on the tests used in the studies. Some patients presented clear aphasia profiles (Lacour et al., 2004) while others showed no symptoms of aphasia. Jennekens-Schinkel et al. (1990).

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