

Characterization of speech therapy publications in the field of neurological syndromes (2009-2019)

Caracterização das publicações fonoaudiológicas sobre síndromes neurológicas (2009-2019)

Caracterización de publicaciones de logopedia em el campo de los síndromes neurológicos (2009-2019)

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Abstract

Introduction: Characterization of speech therapy publications on neurological syndromes. **Objective:** Identify syndromes surveyed, clinical aspects addressed, methodology used and number of publications in 4 Brazilian journals between 2009-2019. **Methods:** Integrative literature review. **Results:** Speech therapy actively participates in this field, with publications distributed in a balanced way over the period studied, with 36 syndromes surveyed, 8 of them with more systematic studies; the majority in the form of a case study, mainly addressing issues related to the evaluation procedures. **Conclusion:** In Speech Therapy, there is, during the period visited, a constant production of articles related to the field of neurological syndromes. Therefore, this field has a recognized presence in this area. Studies are needed for a more comprehensive characterization of the “state of the art”.

Keywords: Neurosciences; Syndrome; Speech, Language and Hearing Sciences

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Authors' contributions:

BMC: Methodology; Data collection; Study outline; Critical review.

RRRP: Study conception; Study outline; Critical review; Guidance;

MLP: Methodology; Data collection; Critical review;

MMP: Methodology; Data collection.

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Resumo

Introdução: Caracterização das publicações fonoaudiológicas sobre síndromes neurológicas. **Objetivo:** Identificar síndromes pesquisadas, aspectos clínicos abordados, metodologia utilizada e número de publicações em 4 periódicos brasileiros entre 2009-2019. **Método:** Revisão Integrativa da literatura. **Resultados:** A Fonoaudiologia participa ativamente deste campo, com publicações distribuídas de modo equilibrado ao longo do período estudado, com 36 síndromes pesquisadas, 8 delas com estudos mais sistemáticos; a maioria sob a forma de estudo de caso, abordando, sobretudo, questões ligadas aos procedimentos de avaliação. **Conclusão:** Na Fonoaudiologia, há, no período visitado, uma produção constante de artigos ligados ao campo das síndromes neurológicas. Portanto, este campo tem presença reconhecida nesta área. Estudos são necessários para uma caracterização mais abrangente do “estado da arte”.

Palavras-chave: Neurociências; Síndrome; Fonoaudiologia

Resumen

Introducción: Caracterización de publicaciones de logopedia sobre síndromes neurológicas. **Objetivo:** Identificar síndromes encuestados, aspectos clínicos abordados, metodología utilizada y número de publicaciones en 4 revistas brasileñas entre 2009-2019. **Metodos:** Revisión bibliográfica integradora. **Resultados:** La logopedia participa activamente en este campo, con publicaciones distribuidas de forma equilibrada durante el período estudiado, con 36 síndromes encuestados, 8 de ellos con estudios más sistemáticos; la mayoría en forma de estudio de caso, que abordan principalmente cuestiones relacionadas con los procedimientos de evaluación. **Conclusión:** En Logopedia, durante el período visitado, existe una producción constante de artículos relacionados con el campo de los síndromes neurológicos. Por tanto, este campo tiene una presencia reconocida en este ámbito. Se necesitan estudios para una caracterización más completa del “estado del arte”.

Palabras clave: Neurociencias; Síndrome; Fonoaudiología

Introduction

The brain has long been the object of study by scholars, such as Hippocrates, who in his treatise “On Sacred Disease” in the 3rd century BC, began to assign the value of the main organ to the brain, being essential for life and determining in the genesis of diseases¹. Over time, studies have deepened and tried to unravel this organ, initially identifying its structures and, then, its functions.

As the brain structure was already fully identified by anatomical studies in the 18th century, physiological studies were promoted and provided opportunities for localization theories^{2,3}. In the following century, Gall was already suggesting that the brain consisted of 35 regions that would include intellectual faculties and emotional behaviors, already relating brain topography and mental functions⁴. These studies advanced and, at the beginning of the 20th century, Brodman identified 52 distinct functional brain regions in a cytoarchitectonic map⁵.

At the same time, histological studies were carried out, which allowed the identification of the structure of neurons, -cell body, dendrites, axons⁶-, as well as pharmacological studies, which highlighted the chemical nature of communication between neural cells, defining the neural tissue as a network⁷.

In the 20th century, studies were developed that aimed to unveil the functional complexes of the brain⁸, which eventually highlighted the need for a multidisciplinary approach to obtain real advances in this field.

In this context, the field of neurosciences has emerged, which is conducive to more vertical and complex studies on consciousness and the brain or the cerebral substrate of the mind^{9,10}.

It is a multidisciplinary field¹¹ that expands into different parts, with particular themes of study - cognition, cells, neural circuits, etc. - that can be approached at different levels - neurobiology, neurocytology, behavioral neuroscience, cognitive

neuroscience, neurolinguistics, among others -, showing its breadth and richness.

This also includes speech-language pathology, since many neurological impairments determine changes in language, hearing, cognition, voice and swallowing¹². Thus, the speech-language pathologist is involved in this discussion, as responsible for studying, evaluating and treating such conditions, as well as studying such skills and functions, in terms of acquisition and development.

One possibility is the elaboration of studies on the speech therapy discussion in the different neurological syndromes in the assessment, diagnosis and treatment, leading to a relevant theoretical-practical overview for the field.

Therefore, this study aimed to identify, through an integrative literature review, publications related to neurological “syndromes” in the last ten years, in Brazilian national journals in the speech-language pathology field. Thus, allowing to prepare a synthesis of knowledge at this specific moment and define the “state-of-the-art” knowledge, which may support clinical decisions and the possible identification of points to be further investigated. The efforts in this direction are explained by being a broad, well-researched and relevant topic.

Method

This is a cross-sectional and retrospective study, which was carried out through an integrative literature review. Due to the nature of the study, the opinion of the Research Ethics Committee was not required.

The guiding question of this study was to understand how national speech-language pathology publications are characterized in themes associated with neurological syndromes between 2009 and 2019.

An integrative literature review was carried out to answer this question in order to identify: (1) number of publications, (2) methodological approaches, (3) neurological issues (syndromes) and (4) clinical issues (assessment/therapy) addressed by the selected studies over the defined period.

The search was carried out in the four national speech-language pathology journals* indexed by the LILACS Database, registered at the Brazilian Institute of Information in Science and Technology (IBICT) and which have the International Standard

Serial Number (ISSN). These journals will be identified hereinafter by the following letters:

- (A) Revista Distúrbios da Comunicação (DIC)
- (B) Revista CEFAC
- (C) Revista CoDAS (*Communication Disorders, Audiology and Swallowing*)/Revista Pró-fono de Atualização Científica/Jornal da Sociedade Brasileira de Fonoaudiologia
- (D) Revista ACR (*Audiology Communication Research*)/Revista da Sociedade Brasileira de Fonoaudiologia.

*the names of the journals have changed over the period studied and, therefore, they are all detailed

The target was as follows: (1) unpublished articles, (2) from 2009 to 2019, (3) in Brazilian Portuguese, (4) related to syndromes, with the investigation of a particular syndrome, (5) addressing speech-language pathology therapy and/or assessment of changes in oral or written language, hearing, voice and orofacial motricity, (6) in any methodological approach. Studies that did not meet all these inclusion criteria were removed, as well as duplicate studies. The following descriptors were used: neuroscience and speech-language pathology and syndrome and speech-language pathology, belonging to the SDS.

It should be noted that the first combination used was “neuroscience and speech-language pathology”, which resulted in a reduced number of publications. Then, the other combination, “syndrome and speech-language pathology”, was used, reaching a more significant number of productions, which led to the decision to use both combinations, excluding duplicate productions.

The choice of the descriptor “syndrome” must also be explained, in combination with speech-language pathology, which is the focus of the study. This choice was not an easy decision due to the many different views on the vast field of neurology, which results in different forms applied in the identification of its nosological entities, in addition to the very definition that distinguishes diseases, disorders and syndromes.

Syndrome is a term that refers to a set of signs and symptoms, which is a relevant approach to speech-language pathology, as it has the clinic as its basic element and, thus, it seemed to be an adequate choice.

However, syndromes can be named in different ways, based on the focus of the discussion.

The identification of the syndrome can be based on the name of the scholar (Down syndrome), the name of the causative element (ZIKv or congenital Zika virus syndrome), the life cycle (childhood syndromes), the nature of the condition (genetic syndromes), the anatomical-functional stratum (degenerative syndromes/peripheral syndromes) or even in the pathological resultant (mental disability)¹³. Thus, a broader descriptor, such as “syndrome”, was understood as more relevant in order not to limit the findings in an excessive way, which, despite efforts, is common in studies guided by one or another set of descriptors. In addition, the integrative review aimed at representing the “state-of-the-art” knowledge, without any aspect or element prevailing, as shown in the six inclusion criteria established.

The first search carried out using two combinations of descriptors found 105 articles. Then, the second search included the reading of the title and abstract and, when appropriate, the full reading of the text, which is always performed by more than

one speech-language pathologist, which resulted in 69 articles found. (FIGURE 1)

The selected articles were entered in an Article Classification Form, which consisted of the following aspects: (1) title; (2) year of publication; (3) journal; (4) syndromes; (5) assessment/therapy; and (6) methodology. (FIGURE 2)

Data analysis was carried out using simple descriptive calculations, as shown in figures for easy viewing and subsequent discussion. The figures intended to show the following data: -

- 1- **Syndromes** investigated: for an assessment of trends in the area;
- 2- Aspects covered (**assessment/therapy**): to identify types of speech-language issues in focus;
- 3- **Methodologies** adopted: to identify the methodological solution selected by the area;
- 4- **Number of publications per journal** over the investigated period, to check the increase, reduction or maintenance in the number of publications.

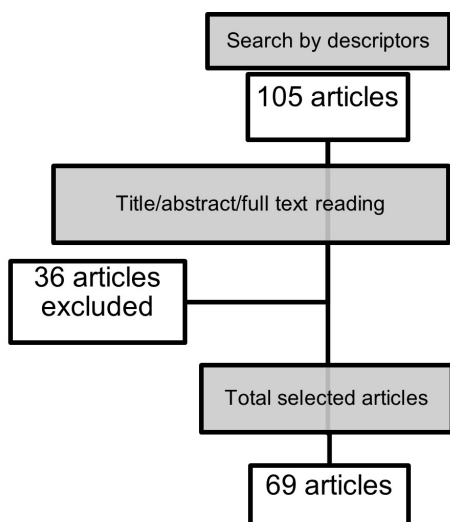


Figure 1

Title:	
Journal	Year:
Syndrome:	Assessment/therapy:
Methodology:	

Figure 2

Results

The 69 selected articles address **36 syndromes**, with **Down syndrome** being the central theme in **21** articles; in addition, **5** articles adopt the **HIV/AIDS**

syndrome as a theme; **4** articles address **Autism Spectrum Disorder and Asperger's syndromes**; **3** articles address the **Fetal Alcohol Spectrum syndrome**, and, finally, **3** articles address the **Prader-Willi, Angelman, Williams-Beuren and Möebius syndromes**. (Figure 3).

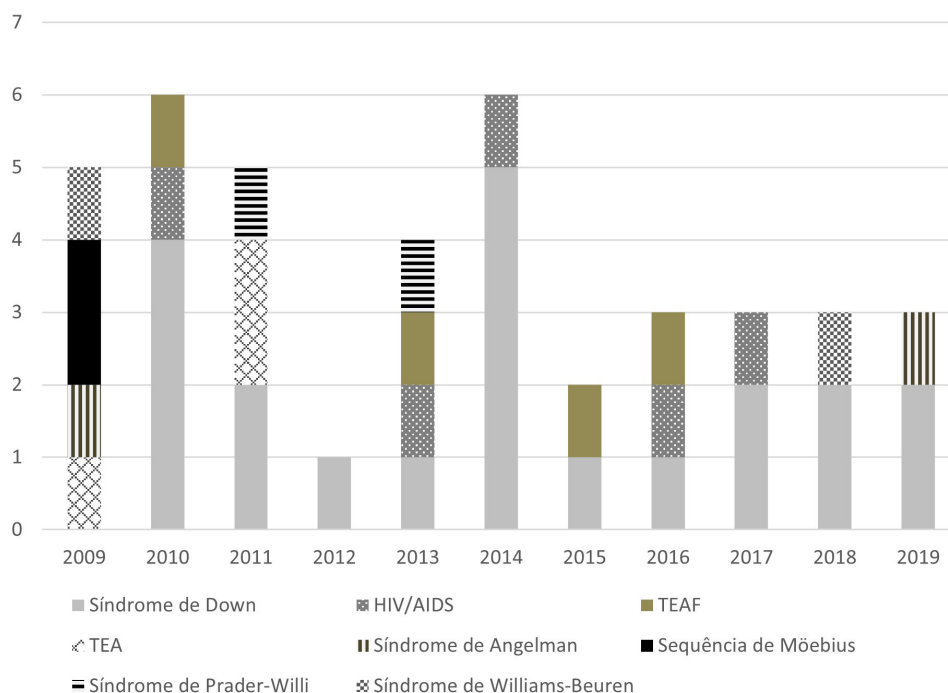


Figure 3. Most researched syndromes over the period studied by number of publications

The remaining **27 syndromes** are reported as the central theme of an article each. As follows: **Pierre-Robin syndrome, Turner syndrome, Kabuki syndrome, Sturge-Weber syndrome, Cat's-eye syndrome, Hunter syndrome, Silver-Russell syndrome, Parry-Romberg syndrome, MELAS syndrome, Fragile X syndrome, Usher syndrome, Meares-Irlen syndrome, Popliteal Pterygium syndrome, Velo-cardio-facial syndrome, Woakes' syndrome, 22q11.2 deletion syndrome, Werdnig-Hoffmann syndrome, Dyslexia syndrome, Kearns-Sayre syndrome, Cornelia**

de Lange syndrome, Smith-Magenis syndrome, branchio-oculo facial syndrome, Crouzon syndrome, musculoskeletal stress syndrome, Arthrogyrosis syndrome, Arnold-Chiari syndrome type III and Burnout syndrome.

Of the **69 articles**, **46** followed the **reporting/case study methodology**, **13** were **case-control studies**, and **10** were an **integrative/systematic literature review**, and in all journals there are publications in the literature review methodology (Figure 4).

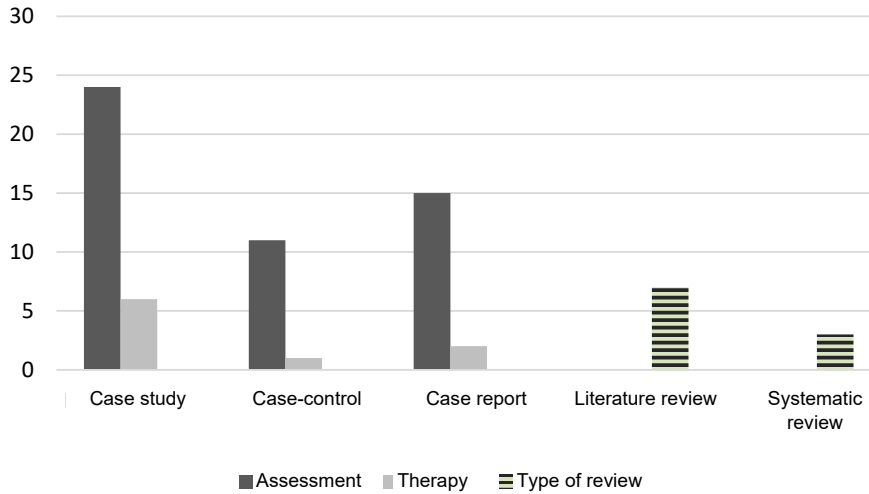


Figure 4. Methodologies adopted vs. issues addressed

Of the 59 articles presented as **case study/case report and case-control**, 50 address issues related to **assessment procedures** and 9 to **therapeutic**

procedures. It should be noted that the ten (10) review articles were not considered (Figure 5).

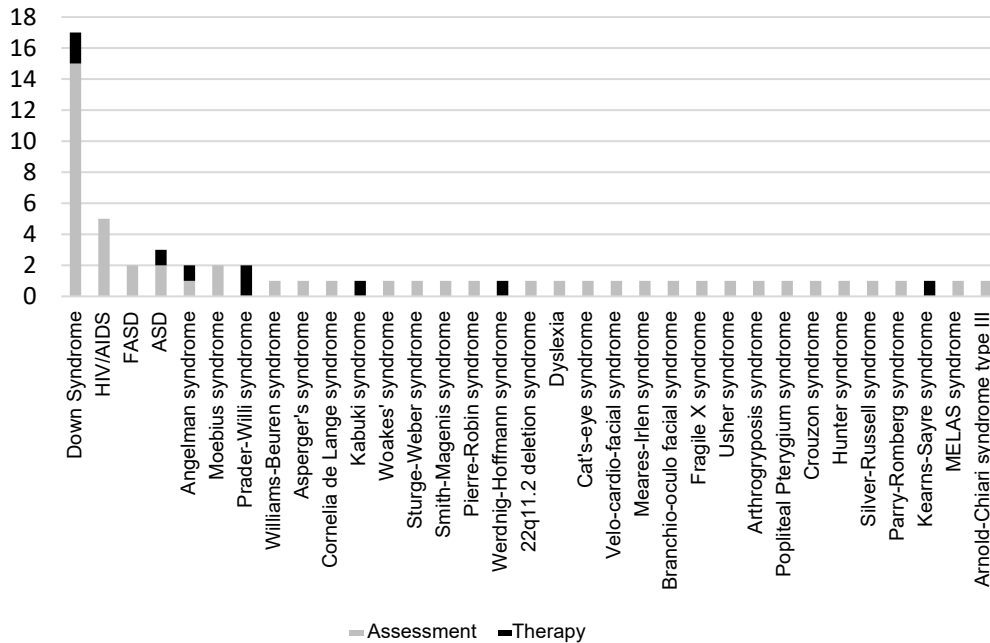


Figura 5. Tipo de questão fonoaudiológica abordada (avaliação/terapia) nas síndromes

2010 was the year with the largest number of publications (11), followed by 2009, 2011, 2014 and 2015 (7); 2013 and 2018 (6); 2012 and 2017

(5); 2019 (4) and 2016 (3). Figure 6 shows that journal D had the highest number of publications (23), followed by journals A (21), C (15) and B (10).

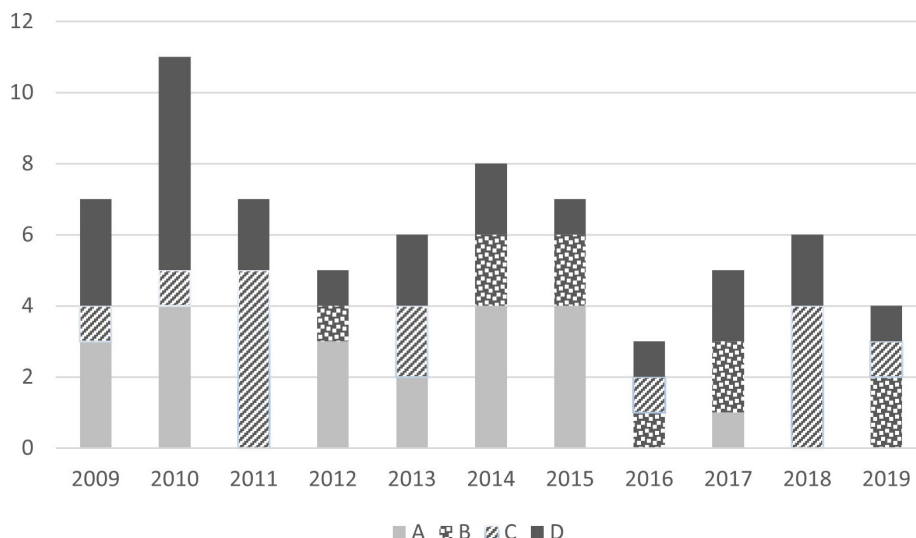


Figure 6. Number of publications in each journal over the period investigated

Discussion

The results obtained by the integrative literature review show that Speech-Language Pathology actively participates in discussions and reflections in the neurological field, addressing several syndromes in their studies.

In 69 selected articles, 36 different syndromes are included as the central topic of discussion, with emphasis on Down Syndrome (DS), which is the focus of 21 studies.

Since its identification and the discovery of its genetic cause in trisomy 21 in 1934, researchers have focused on exploring issues related to DS¹⁴ to develop projects and procedures for prevention, evaluation and treatment. This syndrome has become a source of Early Stimulation, which is a very important initiative, in effect all over the world, in the most diverse environments¹⁵.

In this context, the DS is a frequent topic in the health area, especially in genetic research¹⁶ and in specialized studies in rehabilitation that constantly seeks to improve evaluative, therapeutic and educational procedures for patients diagnosed with the syndrome¹⁷.

In line with the literature^{14,17}, this study found that speech-language pathology research on DS focused on several issues: *language* (13 articles), *alternative communication* (1 article), *listening* (3 articles), *orofacial motricity* (3 articles) and *inclusion* (1). Language and communication are the most explored aspects, since as primary aspects of socialization and learning, they are fundamental conditions for the inclusion of the patient in their community and family environment, promoting autonomy¹⁸.

In this topic, the studies usually address questions about *evaluative procedures* (15 articles) and not much on *therapeutic procedures* (2 articles), in the form of *case study* (13 articles) and *literature review* (4 articles). The prevalence of studies involving issues related to evaluative procedures can be defined by the need for detailed descriptions of the changes as well as the means of accessing them, aiming at a better therapeutic projection. In the period studied, articles about DS are quite common, with emphasis on 2010 (4 articles) and 2014 (5 articles).

Followed by articles related to the HIV/AIDS syndrome. This syndrome is not a recent topic in the

health area, being addressed by speech-language pathology especially in specific areas, perhaps due to its impacts, hearing loss¹⁹ and dysphagia²⁰, with studies on hearing loss being more common.

Five (5) articles were found in the form of case-control study and study/case report, including questions on hearing assessment (3 articles) and swallowing (1 article), as reported in the literature, and only one (1) article was found about reading and writing, distributed throughout 2010, 2013, 2014, 2016 and 2017.

This review also found speech-language studies on the Fetal Alcohol Spectrum Disorder syndrome (FASD), with four (4) articles in 2010, 2013, 2015, and 2016. Two articles are presented as study/case reports, including questions about language assessment and two literature review articles. As it can determine language and hearing development problems, the FASD syndrome is one of the concerns of speech-language pathologists and, therefore, there is an interest in studying assessment procedures for a better clinical description of the condition, which can result in more adequate therapeutic plans.

The Autism Spectrum Disorder (ASD)/Asperger syndromes were also found in four (4) studies, in 2009 and 2011, in the form of a case study (3) and a case-control study (1), addressing aspects related to evaluative procedures.

Although ASD syndrome is a very current topic, it is still discreetly included in publications, perhaps due to the descriptors selected for this literary review, which probably led to the absence of numerous publications on such an important topic over the last decade^{23,24}. In this study, the identified articles mainly address issues related to communication evaluative procedures.

Two studies were found on the Prader-Willi (PWS), Möebius and Williams-Beuren syndromes. The findings are in line with the literature, both in terms of the clinical issues involved (assessment and therapy) and the methodologies adopted.

The articles on PWS, from 2011 and 2013, are case studies addressing therapy issues that cover not only language issues, but also orofacial motricity. This syndrome is of interest to the area, as it may cause problems in orofacial motricity, especially in early life due to tonicity²⁵, and language problems²⁶, in the presence of a general condition of developmental delay.

The articles that discuss Angelman syndrome, from 2009 to 2019, in the form of a case study, address issues of therapy and evaluation in the communication aspect. This subject is not widely addressed in studies, but it is under the attention of the field due to the important changes it causes in language, including lack of communication²⁷.

In turn, Möebius syndrome is presented in the form of a case study, with questions to assess orofacial motricity, due to important facial changes, including facial nerve palsies, sometimes with cleft palate, hearing loss and language problems²⁸.

Publications on Williams-Beuren syndrome, which results in cognitive, behavioral and motor impairment²⁹, are identified in 2009 and 2018, addressing behavioral assessment procedures in the form of a case-control study and systematic review, respectively.

This review identified only one publication for each of the other 27 syndromes over the selected period, the majority in the form of a case study, mainly on assessment procedures, a restriction that may have been determined by the descriptors selected for the study. Despite this, it is important to note that these syndromes were identified and, to this extent, are part of a problem that the field of speech-language pathology has been facing, whether in clinical terms or in research. This idea is reinforced by the case study being the preferred methodology in research, suggesting an effort to propagate clinical experiences, which make up an important space for reflection for all speech-language pathologists.

Journals A (21) and D (24) were the ones that most published articles on speech therapy associated with syndromes, being followed by journals C (15) and B (10), mainly in 2010 (13) and 2014 (8), with a certain stability in the other years, however, with a decrease in 2016 (only 3 articles). In turn, only 4 publications were identified in 2019, but there is a study search limit.

Summary

This literature review answers the guiding question, showing a constant production of several articles related to the field of neurological syndromes, most of them presented in the form of a case study, which is an interesting methodological possibility addressing clinical issues, and presenting difficulties and solutions. Furthermore,

the articles mainly address issues related to the evaluative procedures, which results in the possibility of increasing the clinical descriptions of the conditions, as well as discussing and suggesting ways to adequately perform the speech-language assessment. Therefore, the field of neurological syndromes has a recognized presence in the field of speech-language pathology.

Conclusion

The literature review developed in this study showed an important participation of speech-language pathology in research in the field of neurological syndromes, using several syndromes as a central theme. Due to the nature of the field, almost all selected articles are case studies, which is a relevant and favorable methodological option for discussing significant issues associated with the use of assessment procedures and therapeutic treatment. In addition, it is a suitable methodology for the dissemination of clinical experiences, providing the opportunity to learn about different clinical resolutions and to be able to improve their own practice.

Due to the descriptors selected in this study, only a limited set of articles was selected, which certainly does not correspond to the complete production in the area. Thus, further studies are required from other focuses and using other descriptors, such as life cycles, the nature of the alteration or even the anatomical-physiological stratum involved, in order to increasingly expand the development of a theoretical-practical scenario to be considered and used by speech-language pathologists in their clinical, scientific and even academic activities.

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