



ELSEVIER

Contents lists available at ScienceDirect

## Parkinsonism and Related Disorders

journal homepage: [www.elsevier.com/locate/parkreldis](http://www.elsevier.com/locate/parkreldis)

Review article

## Pain in focal dystonias – A focused review to address an important component of the disease

Micol Avenali<sup>a,b,\*</sup>, R. De Icco<sup>a,b</sup>, M. Tinazzi<sup>c</sup>, G. Defazio<sup>d</sup>, L. Tronconi<sup>e</sup>, G. Sandrini<sup>a,b</sup>, C. Tassorelli<sup>a,b</sup><sup>a</sup> Department of Brain and Behavioural Sciences, University of Pavia, Pavia, Italy<sup>b</sup> Department of Neurology and Neurorehabilitation, Mondino Foundation IRCCS, Pavia, Italy<sup>c</sup> Department of Neurosciences, Biomedicine and Movement, University of Verona, Verona, Italy<sup>d</sup> Department of Medical Sciences and Public Health, University of Cagliari, Cagliari, Italy<sup>e</sup> Department of Public Health, Experimental and Forensic Medicine, University of Pavia - Mondino Foundation IRCCS, Pavia, Italy

## ARTICLE INFO

## Keywords:

Cervical dystonia  
Focal hand dystonia  
Pain assessment  
Pain treatment  
Neurorehabilitation  
Neuromodulation

## ABSTRACT

Focal dystonia is characterized by involuntary muscle contractions that cause abnormal postures and/or twisting movements in a segment of the body. Motor symptoms have a major impact on disability in this condition, but the presence of pain represents an additional source of impairment and poor quality of life. Notwithstanding that pain occurs in up to 70% of patients with cervical dystonia and in a relevant proportion of subjects with focal dystonia of the limbs, it has received very little attention from researchers and controlled trials are scant.

The aim of this review is to summarize the current knowledge on the clinical assessment and management of pain in focal dystonias. The search results will inform on the types of pain reported in focal dystonias, on the tools that are used to quantify pain and on the efficacy of pharmacological and non-pharmacological approaches. The collated data will hopefully improve the clinical management of focal dystonia and also stimulate future research on dystonia-associated pain. Optimization of the outcome indeed requires the identification and the management of all the factors that determine disability and hence relies on a multidisciplinary approach.

## 1. Introduction

Focal dystonias include a group of movement disorders that are characterized by involuntary movements and postures in a segment of the body. Dystonic movements are typically patterned, twisting, and may be tremulous. Dystonia is often initiated or worsened by voluntary action and associated with overflow muscle activation [1]. Although dystonia is commonly referred to as a “motor” disorder, “non-motor” symptoms greatly contribute to worsen quality of life of dystonic patients.

Pain is the most common and disabling non-motor symptom in patients with cervical dystonia (CD), being reported by 75% of patients [2,3]. Limited data is available as regards the occurrence of pain in other focal dystonias [4]. In addition, the nature of dystonic pain is far from being clearly defined. It is generally agreed that dystonic pain is not simply muscular in origin [5], but it possibly reflects an altered central processing of nociceptive stimuli at the spinal level due to the constant and prolonged afferent input caused by muscle contraction. Alternatively, dystonic pain may be generated in basal ganglia nuclei as

a consequence of the dysfunction of the neurotransmission systems [5–7].

A better recognition and understanding of painful symptoms in dystonia is needed in order to put in place adequate and effective therapeutic strategies. The focus of this review was therefore to analyse available data with the ultimate aim to inform stakeholders and create awareness on existing gaps in evidence-based approach. This effort was part of the Project “The Conference of Pain in Neurorehabilitation”, promoted and funded by the Italian Society of Neurorehabilitation [8–10]. The experts of the Conference identified 4 questions as the most relevant for the clinicians in the assessment and management of pain in patients with focal dystonia.

Query 1- Which are the main types of pain in focal dystonias?

Query 2- Are there any predictors or risk factors for developing pain in focal dystonias?

Query 3- Are there standard methods or criteria to evaluate and quantify pain in focal dystonias?

Query 4- What is the evidence of efficacy for pharmacological and non-pharmacological therapies in the management of pain in focal

\* Corresponding author. Mondino Foundation National Neurological Institute IRCCS, Via Mondino 2, 27100 Pavia, Italy.

E-mail address: [avenali.micol87@gmail.com](mailto:avenali.micol87@gmail.com) (M. Avenali).

<https://doi.org/10.1016/j.parkreldis.2018.04.030>

Received 22 November 2017; Received in revised form 5 April 2018; Accepted 26 April 2018

1353-8020/ © 2018 Elsevier Ltd. All rights reserved.

dystonias?

With our review work, we sought to elaborate an evidence-based answer to these questions wherever possible.

## 2. Search methodology for the identification of studies

The literature search was performed without restrictions on year of publication. We searched English language literature in the following scientific databases: Pubmed, EMBASE, and the Cochrane Library.

The keywords used for the search were the following: “cervical dystonia” OR “hand dystonia” OR “limb dystonia” OR “focal dystonia” AND “pain” AND “pathogenesis” OR “scale” OR “measurement” OR “pharmacologic treatment” OR “non-pharmacological treatment” OR “physical therapy” OR “rehabilitation”.

Due to the scarcity of controlled trials and/or systematic studies, we included also case reports, but we excluded proceedings and abstracts of meetings. The limited availability of focused literature on pain and its management in focal dystonias prevented us from adopting a systematic and explicit approach on the quality of evidence, i.e. the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) Working Group [11]. Nevertheless, based on the findings, we attempted a proposal of recommendations (Tables 1 and 2) to highlight the most problematic areas, also for stimulating future research.

During the first round of selection, two independent evaluators (MA and RDI) analysed the abstract of the papers that were identified in the databases and eliminated literature bearing no relevance to this topic. In the round, the evaluators analysed the full text of the selected studies. The data was extracted from each article using a standardized data extraction form.

The data extracted included: type of pain, pain rating scales, pharmacological treatment of pain, role of botulinum toxin, rehabilitation technique, risk and predicting factors for pain in focal dystonia.

**Table 1**  
Recommendations in patient with cervical dystonia.

Query	Recommendation	Grade
1. Which are the main types of pain in focal dystonias?	At present, there are no specific criteria for pain classification. The studies showed extreme variability and the results obtained were varied and sometimes contradictory.	D
2. Are there any predictors or risk factors for developing pain in focal dystonias?	There are different hypotheses of the origin of pain in dystonic patients. We identified prolonged muscular contraction, alteration of nociceptive processing of painful stimuli and alterations of cortical somatosensory system such as potential risk factor for the development of dystonic pain. More studies are needed.	C-D
3. Are there standard methods or criteria to evaluate and quantify pain in focal dystonias?	3.1 The CCDRS could be considered a valid and reliable scale for the assessment of pain and its impact on daily life in CD. 3.2 Published literature on the assessment of pain related to cervical dystonias by means of neurophysiological tools is scant.	B D
4. What is the evidence of efficacy for pharmacological and non-pharmacological therapies in the management of pain in focal dystonias?	4.1 RCTs on oral drugs treatment focusing on pain in patients with CD are lacking. 4.2 Botulinum toxin, either type A and B, is the only effective treatment in relieving pain of cervical dystonia. 4.3 Physical therapy as an adjuvant to BoNT inoculation is useful in patients with cervical dystonia. 4.4 RCTs studies are lacking on the effectiveness of non-invasive neuromodulation techniques. 4.5 Deep Brain Stimulation (DBS) of the globus pallidus seems effective in pain reduction in CD patients; it should be considered as an alternative treatment in those who are refractory to multiple and repeated traditional interventions. Controlled trials are needed for the identification of specific pain treatments as potential alternatives to BoNT treatment. More studies are needed on large series of patients also to better clarify the impact and disability associated with pain in CD.	D A-B A-B D C

Legend to the grading of recommendations.

A – High, further research is very unlikely to change our confidence in the estimate of effect.

B – Moderate, further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.

C – Low, further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.

D – Very low - Any estimate of effect is very uncertain.

## 3. Results

### 3.1. Query 1- which are the main types of pain in focal dystonias?

Unlike pain associated to Parkinson's disease, we could not identify precise nosographic criteria for pain in focal dystonias. Most of the papers we analysed described pain on the basis of patients' reports.

#### 3.1.1. Cervical dystonia

CD is the most common form of focal dystonia. It is characterised by involuntary contraction of the neck muscles, resulting in a variety of abnormal postures of the head. The majority of CD patients report pain, with a prevalence ranging from 54.6% to 88.9% [4,12–14]. The intensity of pain varies and typically it has the characteristics of musculoskeletal and/or myofascial type. Pain is mostly perceived in the neck and shoulders (68% of patients), but often spreads to the upper back region with some radiations directed up to the head on the bent side and down to the ipsilateral upper limb [3,5,15]. About 10%–20% of CD patients suffer also from chronic daily headache. Headache pain is mainly reported in the occipital (79%) or cervical (72.7%) areas, but it is may also spread to the temporal (43.2%), frontal (36.4%) and retro-orbital (11.4%) regions, with the involvement of the vertex in 25% of subjects [14,16,17]. Although, CD has been recognised as a possible cause of secondary headache, only a minority of them (1.3%) meet the criteria of the International Headache Society for “Headache attributed to craniocervical dystonia” [14,16]. Indeed, very few papers described headache attributed to CD, and a correlation between headache and spontaneous pain in dystonic muscles was rarely found [18]. Different authors describe pain as exhausting, radiating, prickly or “pulling the neck” [2,3]. In some cases of CD, pain is particularly intense in proximity of the tendon insertion of the sternocleidomastoid and trapezius muscles [5], to suggest the origin in an abnormal and protracted contraction of these specific muscle groups.

#### 3.1.2. Other focal dystonias

Limited data is available as regards the occurrence of pain in other

Download English Version:

<https://daneshyari.com/en/article/10227113>

Download Persian Version:

<https://daneshyari.com/article/10227113>

[Daneshyari.com](https://daneshyari.com)