

A ROMANIAN SURVEY ON THE IMPACT OF SARS-COV-2 PANDEMIC ON DYSTONIA PATIENTS

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Abstract: *Background.* The SARS-CoV-2 pandemic largely restricted the access of chronic patients to basic healthcare. One of the categories heavily affected by the lockdown were patients with dystonia.

Objective. To assess the impact of botulinum toxin (BoNT) injection therapy discontinuation on the physical status and quality of life (QoL) of patients suffering from dystonia.

Methods. A questionnaire was applied between April 1st and April 15th, 2020 to a group of 50 dystonia patients that met the inclusion criteria for this study.

Results. The majority of the respondents (92%) indicated that their greatest issue during SARS-CoV-2 lockdown was the lack of access to regular BoNT injections. Over 50% of respondents stated that they experienced atypical physical pain, followed by the inability to perform usual activities, with further impact on the QoL. Alteration of the physical status due to the inability to follow an in-clinic rehabilitation program was also reported by up to 25% of patients. In 10% of patients, dystonia became more severe, and led to further afflictions, with a great impact on their QoL. Younger dystonia participants registered lower QoL scores than older participants. Furthermore, isolation and income reduction were found to cause a moderate impact on QoL in most patients and may be associated with mild to moderate forms of depression.

Conclusion. The healthcare field must work to ensure that all these patients have access to their regular treatment plan in a safe environment. Meanwhile, recommendations should be made for these individuals to follow an adequate physical therapy program at home.

Key words: dystonia, botulinum toxin, BoNT, medical rehabilitation, SARS-CoV-2, COVID-19.

INTRODUCTION

In late 2019 a novel type of coronavirus has emerged in Wuhan city of China and in early March 2020, the ongoing coronavirus disease 2019 (COVID-19) outbreak was declared a pandemic by the World Health Organization. The virus was named severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) as it was found to be very similar to the first SARS-CoV (over 80% genetic similarity) and also to MERS-CoV (50% genetic similarity). These are all enveloped RNA viruses, belonging to the Coronaviridae family and can be found in both humans and other mammals. As of June 2nd, 6.403.075 cases have been detected worldwide, out of which, 2.932.299 have recovered and 378.112 have died [1].

The recent pandemic led to the shut-down of several hospitals and wards, leaving numerous chronic patients without the possibility of getting regular treatment for their underlying conditions. Some of the affected categories were neurological patients, including those suffering from various types of dystonia. This affliction is defined as a group of disorders causing movement impairment, characterized by repetitive, intermittent, or sustained muscle spasms, leading to abnormal postures or movements. It can be classified as focal, segmental, multifocal, or generalized [2].

Several countries have released guidelines for patients with dystonia. The Association of British Neurologists issued the following recommendations for neurological patients, according to the estimated risk for each condition: social distancing for all the risk

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categories, including the patients' families and carers; self-isolation for people considered at high risk; the patients in the low and moderate risk groups could be reclassified as high risk if they have other underlying comorbidities, such as lung, kidneys, heart conditions, etc.; frailty was considered an important risk factor for people with neurological conditions or older age, leading to a poor treatment response against COVID-19 [3].

Patients with dystonia were not considered particularly susceptible to SARS-CoV-2 infection, however, people that were known to have suffered from bulbar and respiratory failure, or those that had other underlying diseases, had a higher risk of developing a more severe form of COVID-19. Any disorder that involves respiratory or cognitive impairment and individuals living in care homes could potentially present a poor outcome in the event of SARS-CoV-2 infection [3].

One of the treatment options for dystonia patients is the injection with BoNT in the affected muscles [4]. However, in the current global context of the COVID-19 pandemic, a great majority of patients did not have access to this particular therapy. The scope of the following study was to investigate the effect of discontinuing BoNT injections for the treatment of patients with dystonia during the COVID-19 pandemic. The impact was assessed both on the physical status as well as in terms of QoL.

MATERIALS AND METHODS

The study was conducted by the Children's Joy Association, between April 1st and April 15th, 2020, on 64 patients suffering from dystonia, who were required to answer a number of questions from a standard questionnaire. All respondents gave their consent for the research use of their data. The study protocol was

in accordance with the ethical prerogatives of the 1975 Declaration of Helsinki and respected Good Clinical Practice (GCP) standards.

Inclusion criteria were as follows: [1] age over 18 years old, [2] focal, segmental, multifocal, or generalized dystonia, and [3] patients previously treated with BoNT injections. Out of the 64 patients that were initially recruited, 14 were excluded for not meeting one or more criteria, or for insufficient information submitted in the questionnaire.

The form comprised 8 questions that were aimed at assessing the impact of restricted access to dystonia treatment during COVID-19 lockdown on the patients' physical status and QoL. The participants were allowed to give multiple answers. Additional information regarding the patient's age and gender was collected separately. The data obtained through the form was compiled using Microsoft Office Package.

RESULTS

After applying the exclusion criteria, the final lot consisted of 50 patients. Their ages varied between 26 and 61 years old. Out of the total, 34 participants (68%) were women and 16 were men (32%), leading to a 2:1 report between the two genders (Fig. 1).

Further on, the following information was retrieved from assessing the survey results (Fig. 2):

- 92% (n=46) of participants reported being highly affected by the inability to benefit from BoNT therapy.
- 24% (n=12) of the respondents complained about not having access to an in-clinic complex rehabilitation program under the direct guidance of a physiotherapist;
- 56% (n=28) stated that they experienced atypical physical pain, followed by the inability to perform usual activities, causing a further decrease in

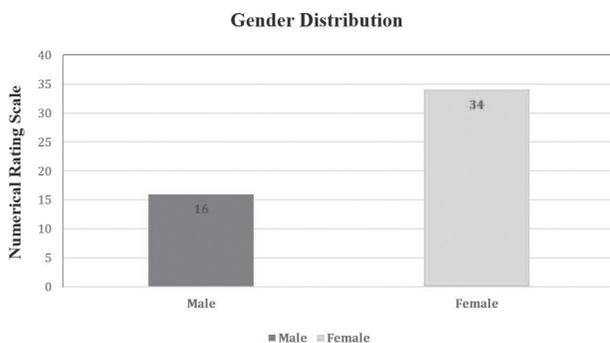


Figure 1. Gender distribution.

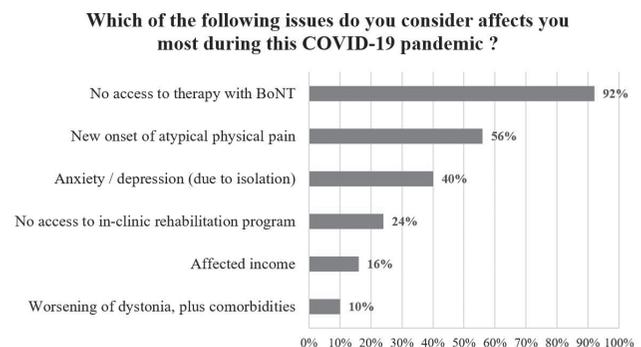


Figure 2. Survey on the most concerning issues of dystonia suffering patients during the COVID-19 pandemic.

the QoL score.

- 10% (n=5) of the patients reported that dystonia became more severe, and even lead to further afflictions, such as carpal tunnel syndrome or urinary incontinence, with a great impact on their QoL;

Other considerations related to QoL assessment:

- Younger dystonia participants had worse QoL than older participants.

- Up to 40% of the participants (n=20) reported being troubled by the lack of social life and isolation, leading to anxiety and mild to moderate depression. Participants who were divorced or separated had significantly worse scores. Nine patients had a mild form of depression and the other eleven suffered from moderate forms. None of the patients had suicidal ideation.

- 16% (n=8) of the participants were highly affected by the impact of the pandemic on their income; Participants with higher socioeconomic status scored significantly higher on the summary index compared to participants without financial stability.

DISCUSSIONS

The study sample consisted of twice as many females than males, which is in accordance with previous findings reporting a female to male ratio of 1,7:1 [5]. The higher prevalence in women may be explained by the fact that they possess certain estrogen receptors in the central nervous system, which were believed to act on the nigrostriatal dopaminergic system [6].

Following the centralization of the questionnaire answers, the study showed that 92% of the participants were highly affected by the fact that they could not benefit from BoNT therapy. The patients in this study typically required regular injections of BoNT every 3 months to maintain symptomatic control. In all cases, more than 12 weeks had passed since the last injection.

BoNT therapy has proved to be highly effective in the management of many neurological disorders, dystonia being one of them [7, 8]. A study conducted in 2010 by Truong *et al.* on 116 patients diagnosed with cervical dystonia showed a significant improvement in the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) as well as in the Visual Analogue Scale (VAS) for pain in the group of patients that benefited from BoNT injections. The effect of the treatment was maintained for an average of 12 weeks [9].

Taking into consideration that the absence of BoNT therapy was the issue that impacted most of the patients suffering from dystonia, the authors recommend that certain protection measures should be adopted to improve patients' access to therapy, even in the actual circumstances. The time allowed for this procedure should be shortened as much as possible, the patient should arrive on time for the scheduled appointment, wearing a mask and should be required to go through triage upon arrival. Also, the physician should be equipped according to the WHO recommendations [10].

In case the procedure cannot be performed according to safety regulations or if the patient is unable to come to the hospital, certain physiotherapy exercises are recommended by the Canadian Society for Exercise Physiology. Some desired goals when treating a dystonia patient are maintaining a cardiovascular tonus through exercising, range of motion preservation, underutilized muscle strengthening exercises, body posture control, and preventing accidents by using orthoses and moving aids [11]. For the less fit patients, it might be helpful to start by performing breathing exercises for a period of time, followed by gradually introducing more strenuous activities. Another advice for keeping track of progress would be to have a physical exercise journal. Furthermore, the amount of workout per week should average about 150 minutes [11].

A study performed in 2012 by Queiroz *et al.* on 40 dystonia patients analyzed the effect of botulinum toxin and physical therapy versus botulinum toxin alone [12]. The study sample was divided into two groups, the first one only receiving BoNT injections, while the other benefited from both therapies. The physical therapy protocol included kinesiotherapy, functional electrical stimulation (FES) on antagonist muscles, and motor learning exercises. The comparison between the two groups was made by using the TWSTRS and the 36-item Short-Form Health Survey (SF-36)[12]. The results showed an improvement in disease severity in both groups, however, only the group that received physical therapy as well, displayed a decrease in the level of pain and disability [12]. This proves once again that even though the BoNT therapy is highly efficient, the results can be amplified by also adding physical therapy procedures, therefore the patients that do not have access to their regular injections should proceed in following their exercise protocols at home [13].

In our study, the QoL of participants was affected by the limitation of both physical and

social functioning, pain, anxiety, and depression. Socioeconomic factors also had an impact, with poorer QoL in participants with a lower income or whose financial security was threatened by the current pandemic. One recent study by Relja *et al.* (2020), conducted on a wide number of dystonia patients, involving 2858 participants from 30 countries, has investigated the impact of the disease on their QoL [14]. Results showed that 69% of them had their family life affected, 59% their working status, 55% reported an altered social life, while 34% said that every aspect of their life was impacted. The current COVID-19 quarantine could have only worsened these negative influences on the QoL already caused by dystonia.

Lack of access to BoNT treatment has caused greater discomfort in younger dystonia participants than older respondents. Younger people tend to have more physically active lifestyles than older people. As a result, worsening of the dystonic symptoms interferes with their everyday life activities and is likely to have a higher psycho-emotionally negative impact in younger than in older people. These findings are in accordance with other published papers on this subject [15].

In conclusion, the next steps that should be taken are creating a safe environment for this group of patients to be able to benefit from their regular BoNT therapy in order to improve their physical performances and QoL. Where the safety desiderates can not be achieved, patients should be advised by their practitioner to follow a daily exercise routine that should maintain their physical symptoms at a manageable level until the healthcare system will be able to return to normality.

Conflict of interest

The authors declare that they have no conflict of interest.

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