



Since January 2020 Elsevier has created a COVID-19 resource centre with free information in English and Mandarin on the novel coronavirus COVID-19. The COVID-19 resource centre is hosted on Elsevier Connect, the company's public news and information website.

Elsevier hereby grants permission to make all its COVID-19-related research that is available on the COVID-19 resource centre - including this research content - immediately available in PubMed Central and other publicly funded repositories, such as the WHO COVID database with rights for unrestricted research re-use and analyses in any form or by any means with acknowledgement of the original source. These permissions are granted for free by Elsevier for as long as the COVID-19 resource centre remains active.

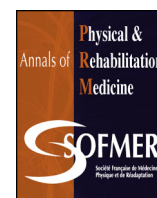


Available online at

ScienceDirect  
www.sciencedirect.com

Elsevier Masson France

EM|consulte  
www.em-consulte.com



## Letter to the editor

**People with Charcot-Marie-Tooth disease and COVID-19:  
Impaired physical conditions due to the lockdown.  
An International cross-sectional survey**



## ARTICLE INFO

## Keywords:

Charcot-Marie-Tooth disease  
Neuromuscular disorders  
COVID-19  
Neuropathies

Dear Editor. The COVID-19 pandemic violently reached Italy by February 2020 and, thereafter, Europe and the United States [1–3], causing a significant burden of disease and death, associated with enormous limitations on the normal lives of people. These limitations included home confinement [4] and the lockdown of schools and absence of most activities. The national health systems resources were absorbed by SARS-Cov-2–infected patients, with consequently decreased access to care for many patients, especially those with chronic diseases.

Our study evaluated the impact of the COVID-19 lockdown on patients with Charcot-Marie-Tooth (CMT) neuropathy, the most common genetic neuromuscular disorder. In particular, we envisaged home confinement as potentially harmful for patients with neuromuscular diseases because they need constant physical therapy, which was suddenly interrupted after the lockdown. Indeed, to date, there are no therapies for the disease, and rehabilitation is the unique effective intervention to reduce the progression of symptoms [5,6].

We designed a simple online questionnaire for CMT patients to investigate how their life habits were transformed during the lockdown and the motor consequences of the restrictions in neuropathic patients. Another aim was to determine whether these limitations affected chronic limb pain and balance, symptoms common to CMT [7,8].

With the support of the Italian CMT Association (ACMT-Rete) and the patronage of the European CMT Federation and other patient advocacy groups, the online survey was distributed to many individuals worldwide (European countries, the United States, and Canada, mainly). The questionnaire, administered in Italian and English from April 6, 2020 to May 11, 2020, contained 5 sections: 1) generic data (age, sex, CMT type) and the “home situation” (the presence of the lockdown in the country, the possibility of staying at home or not); 2) COVID-19 information, accessible only if the patients had the disease; 3) habits and general health situation before the outbreak (e.g., pain in the

extremities, ability to perform exercises at home or outside); 4) how these habits changed after the outbreak; and 5) psychological situation and personal needs.

We used paired-sample t test to compare results for walking, perceived pain and fear of walking before and during the outbreak.  $P < 0.05$  was considered statistically significant. Effect sizes were calculated with Cohen's d coefficient. The STROBE checklist for cross-sectional studies was used for reporting.

In total, 281 patients responded to the survey: 204 women (F/M ratio: 2.72). The most frequent age range was 46 to 55 years (29%). We included all respondents except 2 who stated they had pathologies other than CMT. We included all hereditary neuropathies such as CMT, hereditary neuropathy with liability to pressure palsies and hereditary neuropathy not specified, considering that not all respondents had a molecular diagnosis or could remember/report the precise subtype of their pathology.

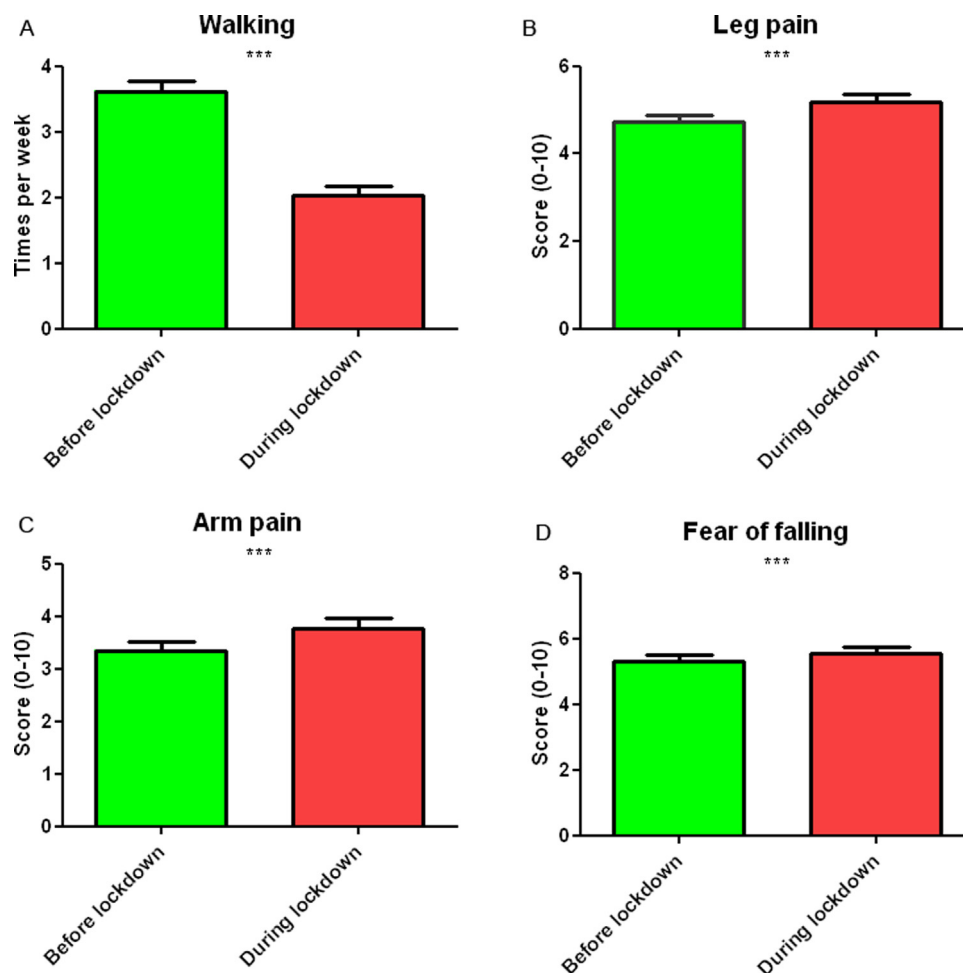
CMT types were represented as follows: CMT1A: 49% ( $n = 137$ ); CMT2 forms: 16% ( $n = 45$ ); CMT1X: 10% ( $n = 28$ ); CMT1B: 5.38% ( $n = 15$ ); and CMT4 C: 2% ( $n = 7$ ). Overall 5% ( $n = 14$ ) of respondents reported rare forms and 11% ( $n = 33$ ) did not specify the type of their neuropathy.

As expected, we found a significant reduction in number of walks per week before and during the lockdown (mean [SD] 3.6 [2.5] vs 2.0 [2.3];  $P < 0.0001$ ; Cohen's d 1.9; Fig. 1A). Also, the perception reporting pain in arms and legs was significantly increased after the lockdown (legs: 4.7 [2.7] vs 5.2 [3.0];  $P < 0.0001$ ; Cohen's d 0.03; arms: 3.3 [2.8] vs 3.8 [3.0];  $P < 0.0001$ ; Cohen's d 0.03; Fig. 1B and C). The fear of falling was also significantly increased (5.3 [3.3] vs 5.4 [3.4];  $P = 0.0004$ ; Cohen's d 0.03; Fig. 1D).

The COVID-19 pandemic has changed our lives in many ways, involving all life domains such as work, social activities and health perception. Healthy people may have experienced a change in their physical performance and perception of physical health, but consequences might have been more severe for people with chronic conditions who need regular exercise to control the disease progression. Also, the massive involvement of health professionals in the pandemic may have left chronic patients behind, with unmet needs.

In the chronic disease CMT neuropathy, constant physiotherapy and physical exercise are important to slow the disease progression and maintain nerve/muscle functionality [6,7]. In our survey, we investigated how individuals' life habits were affected by the pandemic and their health perception. We kept the questionnaire open during the lockdown, with very strict rules in most of the countries. However, the different approaches to the pandemic among countries may in some way have affected our results and their interpretation. Moreover, an online questionnaire could have potential limitations in terms of respondent answers.

For the answers concerning the period before the COVID-19 outbreak and the subsequent lockdown, most patients went out for



**Fig. 1.** Change in (A) walking, perception of (B) leg and (C) arm pain and (D) fear of falling before and during the COVID-19 lockdown by people with Charcot-Marie-Tooth disease.

a walk, with a mean of about 3 days per week. However, during the lockdown, the possibility to walk outside was significantly reduced in terms of days per week [9]. This reduction was probably detrimental for CMT patients because walking is important to stay healthy [6]; moreover, the fear of going outside, as a consequence of self-isolation and social distancing [10], could have further reduced physical activities. The psychological condition of fear was previously highlighted: at the end of the lockdown, many people would prefer to stay at home, feeling more protected, thus not regaining their active routine [11].

These results highlight that health professionals should increase efforts to rehabilitate individuals with CMT after the lockdown and encourage them to resume a life routine comparable to the previous routine before the lockdown, to avoid further impairment and to recover all functionalities lost. A telerehabilitation system could be useful to promote initial motor activity, provide assistance, and, with fear of going outside, inform individuals about safe behaviors to follow for a pleasant and healthy walk [12].

In conclusion, people with CMT worldwide reported significantly impaired physical skills and pain worsening after the COVID-19 lockdown. These symptoms were caused by a relevant decrease in walking and the lack of guidance in physical activity by healthcare professionals. Thus, communication services should be increased and people with CMT helped to resume motor activities and recover the physical skills they lost during the COVID-19 lockdown.

#### Disclosure of interest

The authors declare that they have no competing interest.

#### Funding

Funding was received from AFM Telethon, project number 20821 (Upper Limbs evaluation in hereditary Neuropathies: the ULNA project) and Charitable Donations from ACMT-Rete.

#### Acknowledgments

We thank the ACMT-Rete per la malattia di Charcot-Marie-Tooth Odv and all European CMT Federation associations, CMTA, and HNF for helping us spread the questionnaire worldwide and all the respondents who participated in the survey.

#### Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at <https://doi.org/10.1016/j.rehab.2020.10.001>.

#### References

- [1] Sohrabi C, Alsafi Z, O'Neill N, Khan M, Kerwan A, Al-Jabir A, et al. World Health Organization declares global emergency: a review of the 2019 novel coronavirus (COVID-19). *Int J Surg* 2020. <http://dx.doi.org/10.1016/j.ijsu.2020.02.034>.
- [2] World Health Organization. The coronavirus disease 2019 (COVID-19): situation report-36.; 2020. <http://dx.doi.org/10.3928/19382359-20200219-01>.

- [3] Dey SK, Rahman MM, Siddiqi UR, Howlader A. Analyzing the epidemiological outbreak of COVID-19: a visual exploratory data analysis approach. *J Med Virol* 2020. <http://dx.doi.org/10.1002/jmv.25743>.
- [4] Reluga TC. Game theory of social distancing in response to an epidemic. *PLoS Comput Biol* 2010. <http://dx.doi.org/10.1371/journal.pcbi.1000793>.
- [5] Anens E, Emtner M, Hellström K. Exploratory study of physical activity in persons with Charcot-Marie-Tooth disease. *Arch Phys Med Rehabil* 2015. <http://dx.doi.org/10.1016/j.apmr.2014.09.013>.
- [6] Mori L, Signori A, Prada V, Pareyson D, Piscoquito G, Padua L, et al. Treadmill training in patients affected by Charcot-Marie-Tooth neuropathy: results of a multicenter, prospective, randomized, single-blind, controlled study. *Eur J Neurol* 2020. <http://dx.doi.org/10.1111/ene.14074>.
- [7] Schenone A, Nobbio L, Bragadin MM, Ursino G, Grandis M. Inherited neuropathies. *Curr Treat Options Neurol* 2011;13:160–79. <http://dx.doi.org/10.1007/s11940-011-0115-z>.
- [8] Ribiere C, Bernardin M, Sacconi S, Delmont E, Fournier-Mehouas M, Rauscent H, et al. Pain assessment in Charcot-Marie-Tooth (CMT) disease. *Ann Phys Rehabil Med* 2012;55:160–73. <http://dx.doi.org/10.1016/j.rehab.2012.02.005>.
- [9] Ahorsu DK, Lin CY, Imani V, Saffari M, Griffiths MD, Pakpour AH. The Fear of COVID-19 scale: development and initial validation. *Int J Ment Health Addict* 2020. <http://dx.doi.org/10.1007/s11469-020-00270>.
- [10] Williams SN, Armitage CJ, Tampe T, Dienes K. Public perceptions and experiences of social distancing and social isolation during the COVID-19 pandemic: A UK-based focus group study. *MedRxiv* 2020. <http://dx.doi.org/10.1101/2020.04.10.20061267>.
- [11] Bayen E, Stefanescu F, Robert H, Weil-Chounlamountry A, Villain M, Gouriou C, et al. Will participation restrictions related to the COVID-19 lockdown boost inclusivity? *Ann Phys Rehabil Med* 2020. <http://dx.doi.org/10.1016/j.rehab.2020.04.004>.
- [12] Carl CF. Adoption of telerehabilitation in a developing country before and during the COVID-19 pandemic. *Ann Phys Rehabil Med* 2020. <http://dx.doi.org/10.1016/j.rehab.2020.06.001>.

Valeria Prada<sup>a,\*</sup>, Merhnaz Hamedani<sup>a</sup>, Filippo Genovese<sup>b</sup>,  
Angela Zuppa<sup>a</sup>, Luana Benedetti<sup>a</sup>, Emilia Bellone<sup>a,c</sup>, Marina Grandis<sup>a,c</sup>,

Paola Mandich<sup>a,c</sup>, Angelo Schenone<sup>a,c</sup>

<sup>a</sup>Department of Neurosciences, Rehabilitation, Ophthalmology, Genetics  
and Maternal and Infantile Sciences (DINO GMI), University of Genova,

L.go P. Daneo 3, Genova, Italy

<sup>b</sup>ACMT-Rete per la malattia di Charcot-Marie-Tooth OdV, Bologna, Italy

<sup>c</sup>IRCCS Ospedale Policlinico San Martino, Genova, Italy

\*Corresponding author

E-mail addresses:

[Valeria.prada@gmail.com](mailto:Valeria.prada@gmail.com)[3328613@studenti.unige.it](mailto:3328613@studenti.unige.it) (V. Prada)

Received 29 July 2020

Accepted 5 October 2020