



Tremor in Writer's Cramp Patients: A Retrospective Study

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Background: Tremor is one of the important motor phenotypes of dystonia, however, there is a lacuna in the literature regarding the occurrence of tremor in task-specific dystonia such as writer's cramp (WC).

Aims: To delineate the demographic and clinical characteristics of tremor in WC patients.

Methods: This is a detailed chart review of 105 patients of WC who were classified as WC with dystonic action tremor (WCT+) ($n = 39$; 37.1%) and WC without tremor (WCT-) ($n = 66$; 53.9%).

Results: The mean age of patients was 41.10 ± 14.02 years in the WCT+ group and 36.40 ± 14.66 years in the WCT-group. Males were more commonly affected (94.9% in WCT+ and 84.8% in WCT-). The mean duration of disease was significantly higher in WCT+ as compared to WCT- (4.16 ± 4.48 years vs. 2.57 ± 2.81 years, $p = 0.024$). One-third of our patients (29.52%; $n = 31$) were students but, and the majority of them were classified as WCT- (80.64%, $n = 25$, $p = 0.01$). However, tremor was present in all the three doctors in our cohort with WC ($p = 0.03$). The motor overflow to the elbow and shoulder was significantly associated with the presence of tremor (46.1% of WCT+ vs. 15.2% of WCT-patients, $p = 0.001$). Statistically, there were no significant differences between WCT+ and WCT-patients regarding the presence of complex WC (dystonia during other activities as well) ($p = 0.976$), mirror dystonia ($p = 0.211$), and finger flexion/extension abnormalities ($p = 0.111$).

Conclusion: The presence of tremor in WC was associated with a longer duration of disease and the presence of motor overflow.

Keywords: dystonia, writer's cramp, tremor, overflow, mirroring

INTRODUCTION

Tremor is one of the important motor phenotypes of dystonia and can be present in approximately 12–86.7% of patients. [1–3] Among focal dystonia, tremor is more frequently seen in cervical and upper limb dystonia (ULD). [1,2] A remarkable feature of ULD is its task specificity, meaning that dystonia manifests only during a specific motor task such as writing, typing, or playing a musical instrument. [4] Although over time task-specificity may be lost and ULD may become non-task-specific. More commonly ULD starts as non-task-specific manifesting even at rest or during different types of voluntary movements. Tremor has been more frequently reported in non-task-specific dystonia patients compared to task-specific dystonia. [5] There is a lacuna in the literature with respect to the presence of tremor in task-specific dystonia such as writer's cramp (WC), which is one

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TABLE 1 | Demographic and Clinical Characteristics of writer's cramp (WC) patients with tremor (WCT+) and without tremor (WCT-) Foot notes: The mean between the two groups were compared using the *t*-test (*), and frequencies between the various groups were compared using the χ^2 test (**). The *p* value in bold indicates that the difference is significant (<0.05).

Demographic/Clinical characteristics	WCT+ (n = 39)	WCT- (n = 66)	<i>p</i> Value
Age	41.10 ± 14.02	36.40 ± 14.66	0.656*
Sex (Male: Female)	37:2	56:10	0.119**
Duration	4.16 ± 4.48	2.57 ± 2.81	0.024*
Occupation			
Students	6	25	0.01**
Clerks	8	6	0.09**
Businessmen	6	5	0.13**
Policemen	3	8	0.60**
Government officials	2	3	0.78**
Teachers	1	4	0.49**
Shopkeepers	2	3	0.78**
Doctors	3	0	0.03**
Bankers	2	0	0.11**
Paramedics	0	3	0.54**
Labourers	0	2	0.54**
Others (each one)	(Lawyer, Musician, Engineer, Journalist, Security Guard, Paramedic)	(Lawyer, Musician, Librarian, Stenographer, Telephone officer, Quality control inspector, Barber)	
Family history	3 (7.6%)	5 (12.8%)	0.983**
Simple/complex type	32:7 (82.1%:17.9%)	54:12 (81.8%:18.2%)	0.976**
Motor overflow	18 (46.1%)	10 (15.1%)	0.001**
Mirror dystonia	31 (79.4%)	45 (68.2%)	0.211**
Type of movement			
Finger Flexion-12 (30.7%)		Finger Flexion-31 (46.9%)	0.102**
Finger Extension-3 (7.6%)		Finger Extension-5 (7.5%)	0.982**
Finger Flexion + Extension- 24 (61.5%)		Finger Flexion + Extension-30 (45.4%)	0.111**
Wrist flexion-5 (12.82%)		Wrist flexion-15 (22.72%)	0.211**
Wrist extension- 2 (5.12%)		Wrist extension- 8 (11.59%)	0.238**

of the most common idiopathic task-specific upper limb dystonia seen in the clinic. [6] Most studies have included a relatively small number of patients and they have not looked into the different demographic and clinical factors associated with tremor in WC patients. [7-19] Also, as we gradually move to a digital world, writing has become less common these days, leading to an underreporting of WC, and difficulty in establishing the precise onset of dystonia and tremor.

Therefore, we undertook this retrospective study of our patients who presented with writing difficulty (with and without tremor) to delineate the demographic and clinical characteristics of tremor.

METHODS

This is a detailed chart review of 105 patients of WC who attended our movement disorder clinic over the past 3 years (2017–2020). In addition to demographic details, notes were also made regarding the occupation. Patients were examined while writing with the dominant and non-dominant hands. During the act of writing, involuntary contraction of elbow and shoulder joints (suggestive of motor overflow) was noted. We systematically looked for the presence of mirror movements in the relaxed dominant hand while writing with the non-dominant hand. It was considered to be present when the mirror movements partially or completely reproduced a dystonic

posture in the dominant hand (similar to that observed when writing with this hand).

WC was classified into simple (presence of dystonia only during writing) and complex (dystonia during other activities as well). Patients were further subdivided into WC with dystonic action tremor (WCT+) and without tremor (WCT-). Patients (*n* = 11) diagnosed to have primary writing tremor (defined as task-specific tremor which occurs while writing or attempting to write with lack of overt abnormal posture) during this period were not included for the review. [3] Patients were treated with oral pharmacological drugs (anticholinergics, benzodiazepines, and beta-blockers) and injection botulinum toxin. Data were entered into Microsoft Office Excel and analysis was performed with IBM SPSS software version 27. Values were expressed as means ± standard deviations and as percentages and ranges. The mean between the two groups was compared using the *t*-test, frequencies between the various groups were compared using the χ^2 test, and *p* values ≤ 0.05 were considered statistically significant.

RESULTS

Demographic

There were 39 patients (37.1%) with WCT+ and 66 patients (53.9%) with WCT- (Table 1). Three patients were left-handed, while the rest were right-handed. Students (*n* = 31, 30%) formed

the largest group, followed by clerks ($n = 13$, 12%), policemen ($n = 11$, 10%), businessmen ($n = 11$, 10%), and government officials ($n = 5$, 5%) (Table 1). One-third of our patients (29.52%; $n = 31$) were students but the majority of them had WCT- (80.64%, $n = 25$, $p = 0.01$). However, tremor was present in all the three doctors in our cohort with WC ($p = 0.03$). Out of 105 patients, 93 (88.6%) patients were males. Among the two groups, 94.9% ($n = 37$) of the patients with WCT+ and 84.8% ($n = 56$) of the patients with WCT- were males. There was no predilection for the presence of tremor based on gender. We found that the onset of the disease among patients with WCT- was earlier in females compared to males ($p = 0.034$), however, for patients with WCT+, no significant association was found ($p = 0.086$) with gender. The mean \pm standard deviation (SD) age of our patients was 41.10 ± 14.02 years in the WCT+ group and 36.40 ± 14.66 years in the WCT- group, but, there was no significant difference between the groups ($p = 0.656$). The mean duration of disease was significantly higher in WCT+ as compared to WCT- (4.16 ± 4.48 years vs. 2.57 ± 2.81 years, $p = 0.024$). Three patients in WCT+ and five patients in WCT- had a family history of writer's cramp.

Clinical Characteristics

Simple WC was more common (82.1% patients in WCT+ and 81.8% patients in WCT-) than complex WC (17.9% patients in WCT+ and 18.2% patients in WCT-) in our cohort. There was no association between the presence of complex WC and the duration of disease ($p = 0.950$). Also, no association was found between the complex WC and the presence of tremor ($p = 0.976$).

Finger flexion abnormalities (30.8% in WCT+ and 47.0% in WCT-) were more common than finger extension abnormalities (7.7% in WCT+ and 7.6% in WCT-), but a combination of finger flexion and extension abnormalities was most commonly seen (61.5% in WCT+ and 45.5% in WCT-). No association was found between the finger flexion/extension abnormalities and the presence of tremor ($p = 0.111$).

We observed that involuntary contraction of the musculature of the elbow and shoulders, suggestive of motor overflow, was significantly associated with the presence of tremor (46.1% of WCT+ vs 15.2% of WCT- patients, $p = 0.001$). Mirror dystonia was present in the majority of the WCT+ ($n = 31$, 79.4%) and WCT- ($n = 45$, 68.2%) patients but the difference between the two groups were not significant ($p = 0.211$).

DISCUSSION

Our study of a total of 105 patients with WC observed that 37.1% of the patients with WC had associated dystonic action tremor (WCT+). A review of the literature shows that there is a great deal of variation in the frequency of tremor in WC (5–65%) patients which may be due to several factors such as types of patients included and methods of assessment used (Table 2). [7–22]

We observed that men are affected more commonly than women in WC, which is consistent with the literature. [7,11,23,24] Interestingly, in our study, for all the 12 females with WC, writing was an essential part of their

profession, with 5 (42%) being students, 3 (25%) working as nurses, 2 (17%) as teachers, one (8%) as a policewoman and one (8%) as a doctor. The majority of the students in our cohort did not have tremor, whereas all the three doctors had tremor. This finding is interesting as a significant amount of writing is done by both students and doctors. But, the students had fewer number of writing-years compared to the doctors who were in the medical profession for many years. However, no conclusion can be made considering the very small sample size for the doctor population.

We also observed that the onset of WCT- was earlier in females compared to males ($p = 0.034$), which corroborates the findings reported by other studies. [8,11] However, we did not find a similar association among patients with WCT+ ($p = 0.086$). One study with 957 cases of primary idiopathic dystonia also found a significantly earlier age of onset of WC in females compared to males, contrary to other forms of dystonia (blepharospasm, cervical dystonia) where males had an earlier age of onset. [24]

Our WCT+ patients had a significantly longer duration of disease compared with WCT- patients (4.16 ± 4.48 years vs. 2.57 ± 2.81 , $p = 0.024$). Another study from India also reported that WC patients with dystonic tremor or with associated dystonia in other body parts had a longer duration of symptoms, as well as a delayed onset at presentation. [11] The same study also observed that dystonic tremor was linked with greater disability at presentation compared to all other subtypes of WC. [11] Similar findings have also been observed in cervical dystonia patients where tremor has been observed with a long duration of disease. [25] Our finding lends more credence to the idea that dystonic action tremor may be one of the late manifestations of WC. Also, it is possible that the diagnosis of WCT- was delayed due to the absence of tremor and this hypothesis could be validated in future studies. Further, our findings suggesting more incidence of tremor in doctors compared to students are on similar lines suggesting that tremor is a late feature in WC patients and appears over some time. However, our study findings are limited by less number of patients.

While writing our WCT+ patients had significantly more involvement of ipsilateral proximal joint musculature (elbow and shoulder) suggestive of overflow compared to patients with WCT- ($p = 0.001$). Thus, the presence of motor overflow may be an important marker for tremor in WC patients. Sitburana et al in a study of 30 patients reported that the extent of overflow was significantly associated with the severity of dystonia. [26] They speculated that the high frequencies of motor overflow were due to a loss of normal inhibitory mechanisms leading to widespread abnormalities of motor control. [26] Our findings may point to similar pathophysiology for the occurrence of tremor in WC and further studies are needed to explore this. Mirror dystonia was observed in 72.4% ($n = 76$) of our patients, which is much higher than in other studies. [8,11,14,16,26] However, there was no significant association between tremor and mirror dystonia in our patients.

In our study, over 80% of patients had simple WC and less than 20% of patients had complex WC. However, there was no association between the presence of complex WC and tremor. We

TABLE 2 | Studies reporting tremor in writer's cramp (WC) patients.

Author and year	Number of patients	Sex (Male/Female)	Age in years: Mean \pm standard deviation (range in years)	Mean duration of disease in years	Type of WC (simple/dystonic)	Number of patients having action tremor (percentage)	Comments
⁷ Choudhury 2018	22	22/0	55 \pm 10.5	--	--	10 (45%)	
⁸ Jhunjhunwala 2015	125	109/16	36.8 \pm 14.3	4.9 \pm 5.7 (simple WC); 7.2 \pm 8.5 (complex WC)	103/22	36 (29%)	Family history of WC ($n = 8$) Mirror dystonia ($n = 44$; 35.2%)
⁹ Schneider 2010	27	20/7	43.9 \pm 11.6	11.5	14/13	11 (41%)	
¹⁰ Pekmezovic 2009	19	8/11	40.9 \pm 15.2	6.1 \pm 5.2	--	1 (5%)	
¹¹ Das 2007	141	112/29	33.2 \pm 8.17	4.11 \pm 2.81	108/33	15 (11%)	family history of essential tremor ($n = 11$), or dystonia ($n = 4$) Mirror dystonia ($n = 65$)
¹² Baur 2006	23	10/13	41.5 \pm 4.8	9.1 \pm 9.5	7/16	15 (65%)	
¹³ Schenk 2004	50	21/29	44.3 \pm 11.0	7.5 \pm 7.8	35/15	11 (22%)	
¹⁴ Djebbari 2004	44	19/28	46.1 \pm 14.0	7.6 \pm 8.1	--	6 (14%)	Mirror dystonia ($n = 9$)
¹⁵ Pohl 2002	10	7/3	46.3 \pm 18.6	--	7	2 (20%)	family history of focal dystonia ($n = 7$) History of hand trauma ($n = 3$)
¹⁶ Jedynak 2001	65	42/23	44.2 (19–75)	--	32/33	19 (29%)	Mirror dystonia ($n = 29$)
¹⁷ Wissel 1996	31	15/16	34 \pm 8	12 \pm 3	--	12 (38%)	----
¹⁸ Karp 1994	53	30/23	44 \pm 2 (20–73)	5 \pm 1	21/32	9 (16.98% had postural tremor)	----
¹⁹ Poungvarin 1991	25	17/8	36.8 \pm 10.2	5.88 \pm 7.14	18:7	6 (24%)	----
²⁰ Marsden 1990	91	--	--	--	47/14 30 patients with progressive WC	20 (22%)	----
²¹ Rosenbaum 1988	18	18/10	43.2 (11–65)	4.5	--	10 (56%)	----
²² Sheehy 1982	29	18/11	29.3 (4–53)	16 (range 4–39 years)	13/8 Progressive WC ($n = 8$)	14 (48%)	History of hand trauma ($n = 2$)

did not find any association between complex WC and duration of disease, similar to the study by Jhunjhunwala et al. [8] They speculated that if complex WC were a progression of simple hand dystonia, then it would have been associated with a longer duration of disease. Since this was not the case, they postulated that complex WC might be a separate entity altogether. [8] On the other hand, Das et al reported that complex WC was associated with a longer duration of disease. [11] Thus, further studies are needed to get a more definitive answer about the pathophysiology of complex WC.

Our study findings have some important limitations. First, there may be some information bias inherent to any retrospective study. Second, our center is a tertiary care setting, the results are likely to be influenced by referral biases. Third, we have not provided any data regarding any lag in the diagnosis of our cohort or any misdiagnosis due to musculoskeletal or orthopaedic causes. Fourth, we have not provided any information regarding the effect of botulinum toxin and tremor. One study showed that the presence of tremor in WC was a predictor of poorer response to botulinum toxin. [14] Further studies are

needed to explore how the presence of tremor affects the therapeutic response to various treatment modalities.

Conclusion

Our study has provided detailed demographics and clinical characteristics of tremor in patients with WC. We observed tremor in 37.1% of our patients which was significantly associated with the duration of disease, as well as the presence of motor overflow. Larger follow-up studies are needed to further explore the clinical characteristics seen in WC with writing tremor that will help differentiate it from other disorders such as primary writing tremor and essential tremor.

DATA AVAILABILITY STATEMENT

The data analyzed in this study is subject to the following licenses/restrictions: Data availability is subject to the institutional norms. Requests to access these datasets should be directed to sanjaysgpgi2002@yahoo.co.in.

ETHICS STATEMENT

The studies involving human participants were reviewed and approved by the Institutional ethics committee, Maulana Azad Medical College, New Delhi. Written informed consent for participation was not required for this study in accordance with the national legislation and the institutional requirements.

AUTHOR CONTRIBUTIONS

AD: Research Project Conception, Research Project Organization, Research Project Execution, Statistical Analysis Design, Statistical Analysis Execution, Statistical Analysis Review and Critique, Manuscript Writing of the First Draft, Manuscript Review and Critique. NB: Research

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CONFLICT OF INTEREST

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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