

## PAPER

# Secondary motor disturbances in 101 patients with musician's dystonia

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**Objective:** Musician's focal dystonia is usually considered to be task specific but secondary motor disturbances have been reported also. We carried out a detailed evaluation of the incidence of these secondary motor problems in 101 patients.

**Method:** Symptoms were assessed using clinical histories, neurological examinations and observation of instrumental manoeuvres.

**Results:** 53.5% of patients reported secondary motor disturbances in activities other than playing their main instrument, with the onset delayed in some cases by up to 12 years from the awareness of dystonic symptoms. 46.5% suffered from simple, 19.8% from complex and 33.7% from progressive cramps. Plucked string players (guitarists) mainly suffered from simple cramps while keyboardists more frequently displayed the progressive form. In all patients, symptoms were focal, and the type of cramp was unrelated to the severity of the perceived symptoms. Those patients playing a second instrument similar to their main instrument showed symptoms which worsened to a higher degree than those playing either only one instrument or whose second instrument was different.

**Conclusions:** Longer follow-up assessments may reveal secondary motor symptoms that are not visible over shorter examination periods. Therefore, a thorough evaluation of everyday life motor activities should be considered in any clinical and treatment protocol. We speculate that the avoidance of movements that are similar to the main affected task may be of help in limiting symptoms. Consequently, focal dystonia may be considered more movement than task specific.

The term occupational cramps is used to describe "a group of maladies in which certain symptoms are excited by the attempt to perform some often repeated muscular action, commonly one that is involved in the occupation of the sufferer".<sup>1</sup> The term task specific dystonia is used to address the fact that one specific task and not other manual activities are affected. The best known focal dystonia is probably writer's cramp, which inhibits the ability to write.<sup>2</sup> Dystonia has been diagnosed in, for example, surgeons, milkers, money counters, golfers, tennis and dart players and bowlers,<sup>3–5</sup> as well as in musicians.<sup>6–9</sup> Focal dystonia can manifest with a high degree of specificity, for example during longhand and not shorthand writing,<sup>3</sup> or while playing classical but not electric guitar.<sup>10</sup> Nevertheless, approximately 50% of writer's cramp sufferers also report movement difficulties during other manual activities.<sup>2 3 11</sup> Such secondary motor problems have not been evaluated in detail in musicians, and reports focusing on the issue were based on small samples.

Musicians achieve an exquisite level of motor performance, which is specific to the manual task they usually exercise, in agreement with reports on the neurophysiological basis of motor control of the hand.<sup>12–18</sup> The reported percentages of secondary motor problems in affected musicians are usually low,<sup>7–9 19–21</sup> but the heterogeneity of existing results does not allow one to draw satisfactory conclusions. Thus the question arises as to whether the per cent estimates reported previously are reliable, and what other factors may cause a bias towards the often used task specific characterisation of the disorder.

We assessed the secondary motor problems reported by 101 musicians suffering from focal dystonia. The degree of specificity of the symptoms they reported, the type of secondary tasks being affected, as well as the characteristics they shared with the main affected tasks were inspected. Based on this, we discuss a possible explanation for the spreading of symptoms,

and some aspects of relevance to prevent secondary motor effects.

## PATIENTS AND METHODS

We analysed the clinical histories of all patients treated at the Institut de Fisiologia i Medicina de l'Art in Terrassa, Spain, over the past 5 years. All patients were informed about the purposes of the assessment and agreed to participate in the study. The institutional review board approved the present report.

Apart from their clinical history, the affected musicians were selected based on neurological examination and detailed observation of instrumental playing manoeuvres. Patients exhibiting other neurological pathologies, such as Parkinson disease or nerve entrapments, were not included. We screened with particular emphasis on information directly related to the performed activity. In addition, we carefully assessed activities related to the experienced symptoms other than the main affected task. During clinical assessments, we devoted special attention to characterise when and under which circumstances the reported symptoms appeared. We also noted the following: their characteristics, the degree to which they interfered with the affected task, their evolution and the part of the body affected. The data previously recorded were updated whenever successive visits of the same patient took place. The patients evaluated their severity of symptoms in any of the affected activities using a subjective rating scale ranging from 0 to 100%. A score of 0% meant that the assessed activity was totally affected thus fully preventing its performance.

Following Jedynek *et al*,<sup>11</sup> patients were classified as suffering from simple musician's cramp when focal dystonia affected only one task, from complex musician's cramp when symptoms affected more than one activity or musical instrument and from progressive cramps when focal dystonia evolved from the simple to the complex form.<sup>11</sup>

**Table 1** Relevant demographics of the 101 assessed cases, including main instrument, affected hand and sex

Main instrument	n	Right hand affected	Left hand affected	Both hands affected	Males	Females
Classical guitar	28	23	4	1	25	3
Piano	20	16	3	1	15	5
Flamenco guitar	16	14	1	1	16	0
Violin	6	1	5	0	4	2
Trumpet	5	0*	0*	0*	5	0
Electric guitar	3	3	0	3	3	0
Saxophone	3	1	1	0	3	0
Clarinet	2	2	0	0	1	1
Flute	2	1	0	0	1	1
Horn	2	0*	0*	0*	2	0
Oboe	2	1	0	0	2	0
Percussion	2	1	1	0	2	0
Tuba	2	0*	0*	0*	2	0
Others	8	6	2	0	8	0
Total	101	69	17	6	89	12

0\*, focal dystonia of the lips.

Two instruments were considered to be similar when similar hand positions were used to play them. Six instrumental families were defined: (a) *keyboard* (piano, organ, accordion and bandoneon); (b) *plucked strings* (classical guitar, electric guitar, acoustic guitar, flamenco guitar and electric bass); (c) *strings* (violin, viola and violoncello); (d) *wood winds* (saxophone, clarinet, flute, recorders, tiple and oboe); (e) *brass* (trumpet, horn, trombone and tuba); and (f) *percussion*.

Statistical analyses were produced with ANOVAs, two tailed *t* tests, and  $\chi^2$  tests with continuity correction. Significance level was  $p < 0.05$ ; *p* values of multiple, single average comparisons were adjusted using a simple Bonferroni correction.

## RESULTS

### Demographics

Patients visited our clinical centre 3.4 times on average (range 1–8, SD 1.3) during an average time period of 9.3 months (range 0–33, SD 6.2). A total of 101 out of 771 assessed cases conformed to the diagnosis of focal dystonia; 89 (88.1%) were males. Of the 101 cases, 72 (71.3%) were professional musicians. Sixty-one (61.1%) of the sufferers played only one instrument while 40 (38.9%) played a second instrument.



**Figure 1** A guitarist showing uncontrolled finger flexion of the middle, ring and little finger while playing the guitar. The ring and the little finger in particular flex uncontrolled (A). One year after the appearance of the first movement abnormalities, similar symptoms appeared during type writing (B). Informed consent was obtained for publication of this figure.

Average age for taking up the instrument was 12.2 years (range 4–40; SD 5.2; males 12.6 (SD 5.3); females 9.7 (SD 3.8);  $t = 1.84$ ;  $p < 0.7$ ). The one factorial ANOVA for the beginning of playing with the factor instrumental family (keyboard/plucked strings/strings/wood winds/brass; percussion was not considered in the analysis because only two cases were assessed) was significant ( $F = 3.377$ ,  $df = 1,4$ ,  $p < 0.05$ ). The post hoc *t* test for independent samples revealed that keyboard players started earlier compared with the plucked strings ( $t = 3.407$ ,  $p = 0.01$ ; keyboard 9.65 (SD 4.5); plucked strings 13.3 (SD 4); strings 9.3 (SD 2.8); wood winds 12.5 (SD 3.4); brass 14.8 (SD 10.9)).

In the majority of cases (57.5%), the second instrument belonged to the keyboard family, in 7 (17.5%) to the wood winds, in 4 cases (10%) to the plucked strings, in 4 more cases (10%) to the strings and in 2 cases (5%) to the brass instruments

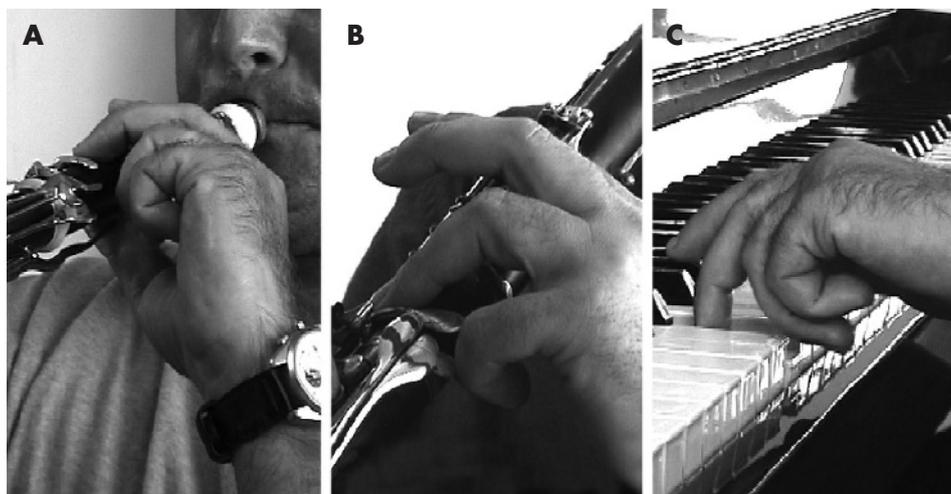
Male guitarists (classical = 25 plus flamenco = 16), corresponding to 93.2% of guitarists and to 40.6% of all sufferers, and male pianists (15 cases), corresponding to 75% of pianists and to 19.8% of all sufferers, were clearly over represented (among the four cases who were females, two played the violin, one the clarinet and the other the flute) (table 1). The mean age of the onset of dystonic symptoms depending on sex was 29.3 years (range 18–56, SD 0.8; males 29.6 (SD 7.9); females 27.5 (SD 10.1); differences not significant).

The ANOVA for differences in age of onset of symptoms across players of the various instrumental families was not significant ( $F = 1.97$ ,  $df = 1,4$ ,  $p = 0.10$ ). The mean time of dystonia evolution was 6.4 years (range 0.5–53, SD 6.8; males 6.6 (SD 7.0); females 5.1 (SD 4.5); NS). The defined instrumental families did not reveal any other significant results. The mean practice time with the main instrument during the year prior to symptom onset was 5.1 h per day (range 1–11, SD 2.1; males 5.1 (SD 2.2); females 4.7 (SD 1.2); NS).

None of the patients presented signs of nerve compressions but electromyography was performed in 67 (66.3%). We found objective signs of lower nerve conduction speeds in five cases (corresponding to 7.5% of the electromyographically assessed cases).

### Secondary motor problems

Fifty-four musicians, corresponding to 53.5% of the total number of affected subjects, reported secondary motor problems in activities other than playing their main instrument. In the remaining 47 (46.5%), the symptoms were specific to the affected instrument (simple musician's cramp). Of the 40



**Figure 2** A professional musician with concomitant tendency to flexion in the ring and little finger while playing (A) his first instrument, the tiple (a Catalan double reed instrument), (B) the clarinet and (C) the piano. Also note the similarity of symptoms between the patient displayed in this figure and the patient in fig 1. Informed consent was obtained for publication of this figure.

musicians who played more than one instrument, 25 (62.5%) reported having some difficulties playing the second instrument, while the remaining 15 (37.5%) did not. In 20 musicians (19.8% of the total number of sufferers, corresponding to 37.0% of those with secondary motor problems), main and secondary motor problems appeared simultaneously (complex musician's cramp). In the other 34 (34% of the total number of sufferers, corresponding to 63.0% of those with secondary motor problems), secondary motor symptoms appeared with delays (progressive musician's cramp). When secondary motor difficulties appeared with a certain delay, the onset interval ranged from 1 month to 12 years (average 24 months for everyday life activities and 24.9 months for the second instrument).

We did not find cases of segmental or generalised evolution. In addition, we did not find any statistically significant differences regarding severity of disturbances while playing the main instrument depending on type of focal dystonia (per cent scores: simple musician's cramp 46.2 (SD 29.9); complex musician's cramp 41.1 (SD 30.6); progressive musician's cramp 51.1 (SD 27.5)).

Only 14 of the 101 musicians assessed reported daily life and the second instrument being affected together. Eight reported that their problems had developed simultaneously, three had suffered problems with the second instrument first and the remaining three reported having problems during their daily life activities first. In spite of the different degree of disturbances according to the type of task being affected, the

fingers and the type of symptoms (flexion, extension, tension, etc) were the same for the main and the secondary affected motor task (see figs 1 and 2). The most affected everyday life activities were: keyboarding of personal computers (25 cases), every action including a precision grip of thumb and index finger (18 cases) and writing (15 cases). Several musicians reported more than one everyday life activity as being affected.

The instrumental family of the affected first instrument accounted for differences in clinical course. Plucked strings more often displayed the simple form of cramps (23 cases, 22.7% of the total amount of patients) while keyboard, wood wind and brass players reported similar but clearly lower percentages of simple cramps (6.9%, 5.9% and 7.9%, respectively). Furthermore, plucked strings and keyboard players both showed similar percentages of progressive cramps (15.8% and 11.8%, respectively) (see table 2).

However, in this regard, we could not find any significant differences between the following: sex, time since onset of music practice, professional degree and number of hours playing per day. Also, age at the time of dystonia onset, time of illness evolution, degree of disturbance while playing the main instrument, time of clinical assessment, alterations of nerve conduction velocity and the beginning of secondary motor symptoms did not reveal any significant difference.

Because our sample included only three left-handed patients, to test left-handedness for its influence on secondary motor problems in musician's cramp evolution would be meaningless in the present report. The secondary affected motor activities as a function of the distribution of symptoms in the body are reported in table 3.

In only three cases did focal hand dystonia appear in both the right and left hand (1, 13 and 84 months after the onset in the main affected hand, respectively). In addition, none of these patients changed the instrument or inverted the playing position in order to overcome their problems.

Regarding type of cramp, in the group of musicians playing more than one instrument ( $n = 40$ ), 14 (35.0%) had simple musician's cramp, 13 (32.5%) complex musician's cramp and the remaining 13 (32.5%) progressive musician's cramp. Conversely, among the 61 musicians playing only one instrument, 33 (54.1%) had a simple, 7 (11.5%) a complex and 21 (34.8%) a progressive musician's cramp. The  $\chi^2$  test comprising the two above mentioned groups was statistically significant ( $\chi^2 = 7.3$ ,  $df = 2$ ,  $p < 0.05$ ). The largest contribution to this  $\chi^2$

**Table 2** Number of musicians per instrumental family and their association with the three different forms of musician's cramp: simple, complex and progressive

Instrumental family	Cramp form			Total
	Simple	Complex	Progressive	
Plucked strings	23	9	16	48
Keyboard	7	4	12	23
Wood winds	6	2	3	11
Brass	8	1	0	9
Strings	3	2	3	8
Percussion	0	2	0	2
Total	47	20	34	101

**Table 3** Affected activity and affected body part. Only those musicians suffering from the affliction of one hand were considered for  $\chi^2$  statistics because of the very low frequencies of affliction in both hands and lips (see results for further details)

Affected activity	Affected body part				Total
	Right hand	Left hand	Both hands	Lips	
Only 1st instrument	28	7	1	11	47
2nd Instrument	2	7	0	0	9
Everyday activities	23	4	1	1	29
Both activities	13	2	1	0	16
Total	66	20	3	12	101

value was observed for those musicians playing more than one instrument and showing a complex musician's cramp (expected value = 7.92). In 65% of musicians who played more than one instrument (26 of 40 cases), dystonic symptoms also affected other activities, compared with 45.9% (28 of 61) of those musicians who played a single instrument. Nevertheless, these differences were not significant ( $\chi^2 = 3.54$ ,  $df = 1$ ,  $p = 0.06$ ). In 75% of the cases playing only one instrument and presenting secondary motor problems, these appeared with delay (21 cases) compared with 50% of those musicians presenting secondary motor problems and playing a second instrument (13 cases). However, these differences were not significant ( $\chi^2$  with continuity correction = 2.621,  $df = 1$ ,  $p = 0.10$ ). Musicians playing a second instrument similar to their main instrument (11 cases) did not show any tendency to suffer from more problems while playing the second instrument, compared with those playing a second, non-similar instrument (29 cases) but remarkably, they reported symptoms worsening with a higher frequency (72.7%) in comparison with those playing a second but dissimilar instrument (27.6%;  $\chi^2$  with continuity correction = 5.021,  $df = 1$ ,  $p < 0.05$ ). Moreover, reports on degree of deterioration of manual skills while playing the first instrument, a second music instrument or while performing everyday life activities revealed that the first instrument was more affected than the second, and that the less affected activities were everyday life activities (means and range in per cent scores: first instrument 24.2 (range 0–80); second instrument 56.5 (range 0–95); everyday life activities 73.8 (range 10–95), higher values representing lower amounts of deterioration in skills).

## DISCUSSION

In common with others,<sup>7 8 19 20 22 23</sup> we found that male guitarists and pianists were clearly most affected by task dystonia compared with other groups of musicians. In particular, the percentage of male guitarists suffering from musician's cramp was higher in our sample. This difference may be related to our particular sample as there was a higher number of Flamenco guitarists in Spain and these individuals manifest more hand problems. Also, this technique is less practiced among women.<sup>10 24</sup> Strikingly, in this type of guitar technique, vigorous movements of the hands are commonly used suggesting that movement intensity is an important factor in the development of dystonia.

### Secondary motor problems

With a certain delay, about half of the cases we studied showed spreading of symptoms into other tasks, albeit these secondary motor disturbances were less pronounced. This delay may

explain the fact that characterisations of the disorder tend to stress the task specificity of musician's dystonia.<sup>9 19 20 25 26</sup> Likewise, similar percentages of secondary motor disturbances have been reported for writer's cramp.<sup>2 11 27</sup>

### Movement similarity threshold

Increasing evidence suggests that focal hand dystonia has some relation to the stereotypical repetition of skilled movements,<sup>28</sup> and in musicians, evidence points to a clear relationship between the distribution of the symptoms within the body and the body part preferentially being used for a specific task.<sup>7 8 19 25 26 29 30</sup> The observed secondary motor disturbances support this assumption. In our sample, dystonic symptoms were reported during attempts to play a second musical instrument that was most probably played at a lower degree of dexterity and intensity along with less time for practice. Nevertheless, those playing a second instrument reported more often the earlier development of secondary motor problems in other daily life activities, suggesting a decrease in task specific symptoms. Thus all manual activities containing similar kinematics may show motor disturbances, independently of the context in which these motor behaviours occur. Therefore, an important factor determining the spreading of symptoms might be the degree of similarity between the performed tasks. If so, a "dissimilarity threshold" between movements may exist. This threshold might then be defined by tasks' kinematics and their inherent sensory parameters, suggesting that additional dystonic symptoms may arise whenever a task surpasses such a threshold. Strikingly, of the 11 patients suffering from dystonia of the lips, only one reported other affected activities. In wind players, the configuration of the lips such as those observed during attempts to play, for example, the flute, might not be used in any other everyday life activity. This may result in a low amount of repetition of dystonic positions reducing the spreading of symptoms to other activities. Moreover, those affected musicians playing a second musical instrument similar to their main instrument reported symptoms worsening to a higher proportion, as compared with those playing a second but dissimilar instrument.

For most of the cases analysed, symptoms were very similar regardless of contextually different tasks. Figures 1 and 2 reveal that the angulations of the fingers are similar in all depicted tasks, and the differences are in the wrist and arm rotations necessary to perform these particular tasks. Because we failed to find higher frequencies of symptoms (as opposed to symptom worsening) in those playing a second, similar instrument, it is probable that other factors such as practice intensity or degree of skilfulness interact with instrumental similarity or similarity of hand positions.

### Factors determining evolution of symptoms

Dystonia of the right hand significantly affected daily life activities. Conversely, when the left hand was affected, the problems were more task specific (table 3). These results probably reflect differences in the amount of hand use among right handed people: while the right hand may be used to a greater extent in everyday life, the left hand may experience more use only while playing an instrument.

The instrument being played also appears to affect illness evolution. For example, guitarists were mainly affected in their right hand (table 1), with a predominance of simple musician's cramp (52.2%, table 3). With this instrument, right hand functionality is characterised by minute postural changes. Such small changes may not play an important role in daily activities thus naturally preventing a spreading of symptoms to other domains.

## Clinical relevance

We agree with Jedynek *et al*<sup>11</sup> that the complex form of dystonia is neither a progressive nor a more severe form of simple focal dystonia. In our sample, musicians suffering from complex or progressive cramps reported comparable amounts of manual disturbances during their instrumental activities. It is important to emphasise that in evaluating and treating patients with focal hand conditions, assessment of other aspects of the patients' daily activities may reveal important information. Even if merely speculative, we firmly believe that our data suggest that some common neuronal encoding is responsible for the symptoms observed during different tasks. Similar behaviours may activate similar kinematics and therefore similar neural substrates. Consequently, focal dystonia may be more movement than task specific. Provided that this is confirmed, the specific reduction of activities involving similar movement behaviours (eg, specific afferents) but not the general reduction of afferents from the affected limb,<sup>31</sup> might prove to be of benefit to limit the symptoms to a particular task. Yet the evidence that any sort of limb immobilisation, reduction of activity, training or physiotherapy prevents progression of musician's dystonia is lacking. Therefore, whether new movement specific, voluntary driven therapeutic interventions<sup>13 16 32-36</sup> will profit from such strategies remains to be tested. Nevertheless, reduction of the amount of practice of some motor behaviours may be of importance given that local somaesthetic factors are more selectively involved in focal hand dystonia,<sup>37</sup> the possible role of fatigue in the corruption of motor programmes involved in playing musical instruments<sup>38</sup> and that brain organisation and the fine tuning of neuronal networks may heavily depend on specific behavioural experience.<sup>32 39</sup>

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