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Original Article

Musculoskeletal pain and deformities in Parkinson's disease

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Abstact: Although musculoskeletal problems are common, there have been few studies focusing on the clinical characteristics of musculoskeletal problems in patients with Parkinson's disease (PD). Our aim was to determine the prevalence and clinical features of musculoskeletal problems and to evaluate their relationship with clinical characteristics. One hundred and twelve PD patients and 120 age and sex-matched controls were interviewed by a rehabilitation specialist concerning their musculoskeletal problems. The prevalence of musculoskeletal pain was significantly higher in the PD group than in the control group (58.9% vs. 42.5%, P=0.012). Commonly involved body sites were the lower back (46.4%), knee (21.4%), and shoulder (21.4%) in the PD group, and lower back (26.7%), knee (20.0%) and ankle (11.7%) in the control group. The lower back and shoulder were more frequently observed in the PD group than in the control group (P=0.002, P=0.016 respectively). In addition, the frequency of camptocormia, Pisa syndrome, striatal hand and foot deformities ranged from 2.2 to 5.4% in PD patients. Multivariate logistic regression analysis revealed that the presence of musculoskeletal pain was associated with female gender, older age and a higher UPDRS II total score in the PD group, independent of Hoehn & Yahr stage, disease duration and medication. Musculoskeletal problems especially involving lower back and shoulder pain are frequent in PD patients and may seriously affect daily activities. For efficient and comprehensive management, musculoskeletal problems should be evaluated attentively in patients with PD.

Keywords: Parkinson's disease, musculoskeletal pain, deformities, clinical features, daily activities

Introduction

Parkinson's disease (PD) is a chronic and progressive neurodegenerative disease characterized by motor signs such as tremor, rigidity, bradykinesia and postural instability and non-motor symptoms such as pain, fatigue, depression, anxiety, dementia and restless leg syndrome [1, 2]. Interest in the motor features of the disease has, until recent years, been higher than in the non-motor symptoms. Although studies concerning the non-motor symptoms have been gradually increasing, only a minority of studies have focused on the pain experience of PD. As pain is not an objective symptom in PD, it is usually ignored by both clinicians and patients, which causes any untreated pain to get worse. Also, pain has the potential to increase the negative effects of motor features on disability, depression and quality of life (QoL) [3, 4].

Musculoskeletal problems (eg. pain and deformities) are the leading causes of chronic pain and physical disability in the general population. Further, it is more common in PD patients than in the general population [5-7]. Musculoskeletal pain is also the most frequent type of pain experienced in PD [8, 9]. In previous studies, despite a higher incidence of musculoskeletal pain being found in the PD groups compared to controls, the rates of PD patients receiving analgesic treatment were similar to those of the control group [10, 11]. In Broetz's study, while the incidence of lower back pain in the control and PD group were 27% and 74%

Table 1. Demographic and disease characteristics of the PD patients and controls

Variables	PD patients (n=112)	Controls (n=120)	*P* values
Sex (M/%)	68 (60.7%)	73 (60.8%)	0.985*
Age (years)	65.0 (10.4)	64.3 (10.9)	0.587**
Disease duration (years)	5.9 (5.1)		
H&Y	2.3 (0.8)		
Levodopa (mg)	360.7 (260.8)		
UPDRS II	14.4 (6.9)		
UPDRS III	20.3 (9.1)		
Tremor subscores	2.4 (1.8)		
Rigidity subscores	2.2 (1.0)		
Bradykinesia subscores	9.2 (4.3)		
PIGD subscores	4.4 (3.6)		
Dyskinesia (n, %)	43 (38.4%)		
Motor fluctuation (n, %)	34 (30.4%)		

Abbreviations: PD, Parkinson's disease; H&Y, Hoehn & Yahr; UPDRS, Unified Parkinson's Disease Rating Scale; PIGD, Postural instability gait disorders; *Pearson chi-square, **Independent sample *t* test.

respectively, the corresponding pain medication rates were 30% versus 34%, most probably indicating an underestimation of the musculoskeletal pain problem in these patient groups, either by patients or by physicians [11].

Despite the prevalence of musculoskeletal problems in this patient group and the resulting chronic pain which can affect the patient's QoL as well as increase the socio-economic burden of PD [5-9, 12], there is, to our knowledge, only one controlled study focusing solely on musculoskeletal problems and its clinical features [10]. The purpose of this study is to identify the prevalence and clinical features of musculoskeletal pain and examine its relationship with clinical characteristics in patients with PD.

Materials and methods

One hundred and twelve patients with PD consecutively admitted to the movement disorders outpatient clinic from January 2012 to July 2012 were interviewed about their musculoskeletal problems. All patients satisfied the United Kingdom Parkinson's Disease Society Brain Bank Criteria [13] and have been on dopaminergic therapy with levodopa and/or dopamine agonists for at least 3 months.

Patients with non-musculoskeletal pain (n=50) such as neuropathic, radicular, dystonic, central pain or with more than one pain according to the Ford classification [14], and patients with speech (n=3) or cognitive abnormalities (n=8), who could not explain their past medical history nor their existing medical condition, were excluded. One hundred and twenty age and sex-matched controls who agreed to the study were enrolled consecutively by non-relative caregivers or spouses. None of them had a family history of PD or neuropathic radicular pain. All aspects of the study were carried out with the subjects having an adequate understanding of the study. Written informed consent was received from all subjects and the study was approved by the institutional ethics committee.

A rehabilitation specialist interviewed all the participants using a structured format and reviewed their medical records which included the demographic factors (gender, age), disease characteristics (duration of PD), Levodopa daily dose , Hoehn & Yahr (H&Y) stage [15, 16], the Unified Parkinson's Disease Rating Scale (UPDRS) score of part II (daily activities) and part III (motor signs) [17]. After medical recordings, the participants were questioned about their current musculoskeletal problems and examined for any existing deformities. All participants with PD were assessed during ON period.

Musculoskeletal pain was identified according to the Ford classification [14]. Patients and controls who had suffered musculoskeletal pain at least for 3 months were recorded. The areas in which musculoskeletal pain was involved; such as neck, shoulder, wrist, lower back, hip, knee and ankle), laterality of symptoms (for PD patients), pain intensity (visual analogue scale) [18] and the presence of any deformities (striatal hand, striatal foot, camptocormia and PISA syndrome) were recorded.

Descriptive statistics were shown as mean and standard deviations for continuous variables and the number of cases or percentages for nominal variables. Bivariate associations were established using the chi-square test for categorical variables and the independent sample *t*-test for continuous variables. Logistic regres-

Table 2. The prevalence and clinical characteristics of musculoskeletal pain

Variables	PD patients (n=112)	Controls (n=120)	Chi-Square value	*P* values
Existence of chronic MS pain (n, %)	66 (58.9%)	51 (42.5%)	6.255	0.012*
The body parts involved by chronic MS pain (n, %)				
Neck	8 (7.1%)	11 (9.2%)	0.316	0.574*
Shoulder	24 (21.4%)	12 (10.0%)	5.772	0.016*
Wrist/Hands	6 (5.4%)	8 (6.7%)	0.175	0.676*
Low back	52 (46.4%)	32 (26.7%)	9.795	0.002*
Hip	5 (4.5%)	4 (3.3%)	0.199	0.742**
Knee	24 (21.4%)	24 (20.0%)	0.072	0.788*
Ankle/Foot	11 (9.8%)	14 (11.7%)	0.215	0.651*

Abbreviations: PD, Parkinson's disease; MS, musculoskeletal. *Pearson chi-square, **Fisher's exact test.

Table 3. Frequency of deformities in the PD group*

Deformities	PD patients (n=112)
Striatal hand	3 (3.3%)
Striatal foot	2 (2.2%)
Camptocormia	6 (5.4%)
PISA syndrome	6 (5.4%)

*Non in control group.

sion analysis was utilised to establish the risk factors impacting musculoskeletal pain. Firstly, simple logistic regression analysis was used for factors (independent variables) thought to be potentially significant in musculoskeletal pain (dependent variable). Variables that were established to have statistically significant correlation with musculoskeletal pain were included in the multiple logistic regression analysis. A significant level of $P<0.05$ was set for all statistical tests. All analyses were conducted using the Statistical Package for Social Sciences (SPSS) version 21 statistical software.

Results

A total of 112 patients with PD and 120 age and sex-matched controls were included in the study. Among the 112-interviewed PD patients, who had an average age of 65.01±10.35, 68 (60.7%) were men. Seventy-three (60.8%) of the controls were men and the mean age of the controls was 64.25±10.87. There were no significant differences in age and sex when compared with the control group. The average of H&Y stage, UPDRS II and UPDRS III scores of the PD group were 2.29±0.76, 14.43±6.85 and 20.31±9.08 respectively. The clinical characteristics of the PD patients are presented in **Table 1**.

Of the 112 PD patients, 66 patients (58.9%) and 51 out of 120 (42.5%) participants in the control group had had musculoskeletal pain for at least 3 months at the time of the interview. The PD group had a significantly higher rate of musculoskeletal pain compared to the control group (P=0.012). In the PD group the areas of pain were distributed as follows; the lower back (46.4%), knees (21.4%), shoulders (21.4%). In the control group, the most commonly affected parts were; the lower back (26.7%), knees (20.0%), foot-ankle (11.7%). The PD group had a significantly higher rate of lower back and shoulder pain than in the control group (P=0.002, P=0.016 respectively). However, there were no significant differences between the PD and control groups in terms of neck, wrist, hip and foot-ankle pain (P>0.05). There was no difference concerning the severity of musculoskeletal pain assessed by VAS between the PD and control groups (P=0.944). Additionally, 6 (5.4%) patients had camptocormia, 6 (5.4%) patients had PISA syndrome, 3 (3.3%) patients had striatal hand deformity and 2 (2.2%) patients had striatal foot deformities in the PD group. The frequency and characteristics of musculoskeletal pain and deformities are given in **Tables 2** and **3**.

We also surveyed the laterality (the more involved side) of musculoskeletal pain and PD symptoms (left, right or bilateral); among the 66 patients who suffered musculoskeletal pain, 49 (74.2%) patients had pain on the same side and the PD symptoms were more severe.

Table 4. Comparison of clinical characteristics between PD patients with and without musculoskeletal pain: Univariate logistic regression analysis

Variables	β	SE	Oddsratio	95% CI	*P* value
Sex (F)	0.810	0.410	2.248	1.007-5.021	0.048
Age	0.038	0.019	1.038	1.000-1.078	0.051
Diseaseduration	0.002	0.003	1.002	0.995-1.008	0.596
H&Y	0.411	0.267	1.508	0.893-2.545	0.124
Levodopa	-0.001	0.001	0.999	0.998-1.001	0.317
Levodopa (+)	0.066	0.454	1.069	0.439-2.600	0.884
Agonist (+)	0.031	0.467	1.032	0.413-2.579	0.947
UPDRS II	0.115	0.035	1.122	1.048-1.200	0.001
UPDRS III	0.065	0.025	1.067	1.015-1.121	0.011
Tremor subscore	0.119	0.113	1.126	0.903-1.404	0.291
Rigiditysubscore	0.493	0.212	1.637	1.080-2.481	0.020
Bradykinesiasubscore	0.140	0.051	1.150	1.041-1.270	0.006
PIGD subscore	0.156	0.068	1.169	1.024-1.335	0.021
Dyskinesia (+)	1.097	0.424	2.995	1.303-6.881	0.010
Motor fluctuation (+)	0.919	0.450	2.507	1.038-6.056	0.041

β, regression coefficient; SE, standard error; CI, confidence interval; F, female; H&Y, Hoehn & Yahr; UPDRS, unified Parkinson's disease rating scale; PIGD, postural instability and gait disorders.

Table 5. Comparison of clinical characteristics between PD patients with and without musculoskeletal pain:Multivariate logistic regression analysis

Variables	β	SE	Oddsratio	95% CI	*P* value
Sex (F)	1.189	0.519	3.283	1.186-9.084	0.022
Age	0.048	0.023	1.050	1.004-1.097	0.034
UPDRS II	0.156	0.064	1.169	1.032-1.325	0.014
UPDRS III	-0.096	0.100	0.909	0.747-1.106	0.339
Rigiditysubscore	0.459	0.357	1.582	0.786-3.183	0.198
Bradykinesiasubscore	0.185	0.146	1.204	0.904-1.603	0.205
PIGD subscore	-0.144	0.161	0.866	0.631-1.186	0.369
Dyskinesia (+)	0.041	0.610	1.042	0.315-3.446	0.946
Motor fluctuation (+)	0.074	0.640	1.076	0.307-3.775	0.908

β, regression coefficient; SE, standard error; CI, confidence interval; F, female; UPDRS, unified Parkinson's disease rating scale; PIGD, postural instability and gait disorders.

In addition, the univariate analyses showed that PD patients with musculoskeletal pain, compared with PD patients without musculoskeletal pain, were often females, older patients, had higher scores on the UPDRS II and UPDRS III, independent of Hoehn & Yahr stage, disease duration and medication. In addition, they had higher rigidity, bradykinesia, dyskinesia and motor fluctuation sub-scores of UPDRS III (**Table 4**). However, the multivariate logistic regression analyses revealed that the presence of musculoskeletal pain was associated with female gender (OR=3.28; 95% CI 1.18-9.08), older age (OR= 1.05; 95% CI 1.004-1.097) and UPDRS II (OR=1.16; 95% CI 1.03-1.32) total score (**Table 5**).

Discussion

Our results revealed that 58.9% of patients with PD had musculoskeletal pain, which was significantly higher than in the control group and compatible with previous studies in which the frequency of musculoskeletal pain ranged from 30 to 70% in patients with PD [8, 9]. Musculoskeletal pain is common, increases with age and is also more prevalent in females than in males in the general population [5]. Similar to our results, several studies have reported that the prevalence of musculoskeletal problems in PD groups is higher in females than in males and that musculoskeletal pain is significantly associated with increased age [10, 19]. Therefore, we can deduce that musculoskeletal pain, affecting 2/3 of patients with PD and which is more frequent than in the general population, should be evaluated attentively in patients diagnosed with PD.

Our study has shown that the most frequent musculoskeletal problem in the PD group was lower back pain (46.4%) and was found to be significantly higher than in the control group. The prevalence of lower back pain varies between 42.6-74% and it is the most frequent type of musculoskeletal problem seen in patients with PD [11, 20]. Festination, kyphosis and dystonia, which are clinical features of PD,

can induce stress on the ventral portion of the lumbar disc structure causing lumbar disc herniation and may lead to radicular lower back pain [21]. Abnormal muscle tone and a reduction in vertebral flexibility can cause non-radicular low back pain originating from the muscles, soft tissue and skeletal structure in these patients [11]. In addition, long-term kyphosis or long-lasting asymmetric posture, rigidity and osteoporosis can contribute to lower back pain [19]. As it negatively affects ADL and contributes to disability and in order to improve function and prevent further disability for patients with PD, it is critical to evaluate lower back pain and mechanical preventive measures and physical therapy treatment programs must be taken to prevent the occurrence of pain.

Shoulder problems are also common in patients with PD and the prevalence of shoulder pain has been estimated to be 11-80% [22, 23]. Shoulder pain in these patients was associated with the intensity of akinesia and increased incidence of frozen shoulder associated with increased muscle inactivity especially in the early stage of the disease rather than previous shoulder trauma [22, 23]. Remarkably the first PD symptom may be shoulder pain, especially frozen shoulder in 2-8% of patients with PD, which may long precede the clinical diagnosis of PD. In studies evaluating the pathology of shoulder problems, rigidity and long disease duration have been shown to be factors that cause shoulder problems in PD [24, 25]. In our study, shoulder pain was significantly higher in the PD group than in the control group. Shoulder pain imposes serious restrictions on the functions of the upper extremity which is very important for the successful execution of ADL, and thus it is important to question shoulder pain while evaluating patients with PD. In our clinical experience, in order to prevent shoulder problems, an evaluation of simple shoulder range of motion in addition to careful questioning about shoulder pain is critical to provide early diagnosis and treatment.

The prevalence of neck, hand-wrist, hip and foot-ankle pain were similar in both groups. It should be kept in mind that additional pain may further increase the risk of disability in the PD patient group, although they are considered to be usual degenerative conditions of articular joints related to disuse and immobility.

In this study, striatal foot and hand deformities were detected in 2.2 and 3.3% of PD patients. While striatal hand deformity is characterized by ulnar deviation of the hand, flexion of the metacarpophalangeal joints, and extension of the interphalangeal joints, striatal foot is defined as extension of the great toe with or without flexion of the remaining toes [26, 27]. Rigidity, dystonia and spasm in the fingers were accused of hand and foot deformities [28, 29]. In addition to obvious cosmetic problems, they can also interfere with ADL; may cause pain, impair walking and contribute to an increased risk of falling. Hand deformities may be misdiagnosed as the hand deformities of rheumatoid arthritis, Dupuytren's contracture or flexor tendon entrapment of the digits. Accordingly, it is imperative that physicians bear in mind the differential diagnosis of these deformities in the clinical evaluation of patients with PD. Although their prevalence has not been studied systematically [26], striatal hand and foot deformities have been reported in 10% of patients with advanced, untreated PD [30], so the prevalence of 2.2 to 3.3% found in our study population, who were on medical treatment, seems reasonable.

Camptocormia in PD is a progressive, irreversible, and most common and most disabling syndrome, associated with basal ganglia and lenticular lesions, and characterized by pronounced forward flexion of the thoracolumbar spine. It increases while standing and walking and disappears in the supine position [26, 27, 31], and is frequently associated with back pain [31]. On the other hand, Pisa syndrome is a form of axial dystonic syndrome rarely described in PD and associated with striatal cholinergic-dopaminergic imbalance. It was first described in patients taking neuroleptic medication, and characterized by tonic lateral flexion accompanied by a slight rotation of the trunk in the sagittal plane [32, 33]. Although Pisa syndrome may be reversible with drug therapy [32, 33], currently, there are no effective therapies for camptocormia. Custom-made corsets and physical therapy modalities have been recommended for camptocormia with limited benefit [31], so there is a need for more research into the effectiveness of the treatments. While there have been very few previous studies looking at the prevalence of these disorders among patients with PD, the findings

of this study have revealed prevalence rates of 5.4% for both deformities.

Another point is that UPDRS part II total score assessing the ADL, was higher in patients with musculoskeletal pain and therefore it is rational to infer that PD patients with musculoskeletal pain might become more dependent on ADL. However, our study was cross-sectional and as correlation does not imply causation, an inverse relationship may exist in that decreased ADL might be the cause of musculoskeletal pain.

The selection of the control group by spouses and caregivers is a limitation of this study. Since PD is a chronic disease, the incidence of musculoskeletal pain may be higher in the control group with chronic diseases. The exclusion of patients with cognitive dysfunction or speech problems and assessment of patients only admitted to outpatient clinics are other limitations, which could have reduced the incidence of musculoskeletal pain in the PD group. Finally the possible effects of mechanical problems caused by scoliosis and osteoporosis on musculoskeletal pain, have not been assessed in this study. Future studies should focus on the mechanical stress/pain relationship to clarify the etiopathogenesis of musculoskeletal problems.

Conclusion

Musculoskeletal problems are common and may seriously affect the ADL in patients with PD. Since PD most frequently affects older adults, both the disease itself and musculoskeletal problems can lead to serious physical disability and can put patients in a vicious cycle of pain and disability. For the efficient and comprehensive management of PD patients the definitive evaluation of musculoskeletal problems is essential.

Disclosure of conflict of interest

None.

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