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REVIEW

Frozen shoulder or shoulder stiffness from Parkinson disease?

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Abstract

Purpose To understand the role of camptocormia (increased kyphosis) and postural alteration in Parkinson's disease in the development of shoulder pathology, with a special concern for adhesive capsulitis and shoulder stiffness.

Methods A preliminary online search was carried out, with combination of keywords including "Parkinson," "Shoulder stiffness," "Frozen shoulder," "Adhesive capsulitis," "Postural alteration," "Camptocormia". The retrieved papers were screened by title and abstract and those considered relevant to the aim of the review were read in full text and included. Relevant information were extracted and reported into text.

Results Due to a severe impairment of posture, patients affected by PD show an increased thoracic kyphosis (camptocormia) and decreased mobility of the trunk that can yield a humeroacromial impingement syndrome and capsulitis, resulting in inflammation of the bursa, shoulder pain and reduction of movement. Furthermore, kinematic of the shoulder is allowed by the combined movement of the humerus, the scapula, the clavicle, the thoracic wall and thoracic spine. The thoracic spine and wall mobility are severely impaired in the parkinsonian patient, thus limiting the shoulder motion.

Conclusion The postural alteration observed in PD is the primum movens for shoulder pathology, since anterior tilt of the scapula, which occurs with the increment of thoracic kyphosis, yields to a subacromial impingement. A closed loop is then created, as the rigidity of the shoulder causes further alteration in the posture, which worsens the impingement syndrome and so on.

Keywords Parkinson's disease · Shoulder stiffness · Adhesive capsulitis · Postural alteration · Frozen shoulder · Kinematic · Shoulder

Introduction

Adhesive capsulitis of the shoulder, also known as frozen shoulder, is a disabling pathology which affects one or both shoulders, yielding to pain and stiffness, with a severe impairment of the range of motion (ROM). Estimated cumulative incidence is 2.4 per 1000 people per year [30], with highest incidence in the female sex, between fifth and sixth decade of life [25], its impact on working activities is relevant indeed. It has been reported that frozen shoulder is strictly correlated with some systemic pathologies, especially with hypothyroidism and diabetes [27]. It has been reported that incidence of capsulitis in patients affected by diabetes varies among 10% and 36%, and that responsiveness to therapy is impaired when compared to nondiabetic patients [9, 25]. In addition to idiopathic capsulitis causing frozen shoulder, several cases of secondary frozen shoulder have been reported. The secondary pathology is mostly associated with trauma of the shoulder or with tendinopathy or tears of the rotator cuff. In the literature, several papers report also cases of frozen shoulder, subsequently to arthroscopic repair of the rotator cuff [13, 24], causing post-operative shoulder stiffness. Parkinson's disease (PD) is considered as the second most common neurodegenerative disorder of the elderly population, affecting up to 2% of the population over 65 years [8]. It affects both sex, and

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all ages, although is prevalent in older males. It is a disorder of central nervous system, affecting especially basal ganglia and pathways involved in control of movement and balance. Depletion of dopaminergic connection in the substantia nigra of the midbrain is the main pathogenic event, causing loss of function of these pathways [14]. The patient affected by Parkinson's disease commonly shows rigidity and resting tremor, bradykinesia, micrography, a mimic face expression [14]. Furthermore, a severe impairment of posture is retrieved in these patients, with an increased thoracic kyphosis and decreased mobility of the trunk on all planes. It has been described the classic "stooped" appearance of the patient affected by PD, in which hips and knees are mildly flexed and shoulders are in a rounding shape. However, more severe alteration can occur, especially in advanced phases of the disease progression. Among these, scoliosis and Pisa syndrome affect posture on coronal plane, while camptocormia and antecollis affect posture on sagittal plane [10]. The authors questioned about the possibility that postural modification occurring in PD may affect the kinematic of upper trunk and scapulo-thoracic cingulum, leading to impaired shoulder mobility and consequentially to frozen shoulder. The primary endpoint of this review of the literature is to investigate the role of postural alteration in Parkinson's disease in the development of shoulder pathology, with a special concern for the shoulder stiffness. The null hypothesis is that camptocormia and anterior scapular tilt may induce a subacromial impingement which leads chronically to shoulder inflammation and capsulitis, resulting in stiffness.

Methods

A comprehensive search of the literature was carried out between March and May 2018, though electronic search of databases including PubMed-Medline, Cochrane Collaboration and Google Scholar. Keywords and combination of these, utilized for the search included: "Parkinson", "Shoulder stiffness", "Frozen shoulder", "Adhesive capsulitis", "Postural alteration", "Camptocormia", "Kyphosis". Paper retrieved by online search were firstly screened by title and subsequently, the abstract was read in full, for those paper considered relevant for inclusion. Those paper which dealt with postural modification in PD, a possible relationship between PD and shoulder pathology, were retrieved in full text and read. Furthermore, bibliography of each included paper was screened, to retrieve any further relevant publication. Given the heterogeneity of the retrieved paper, the authors were prevented from summarize comparable outcomes and data in tables. A descriptive and comprehensive analysis of data was reported into text.

Results

Pathophysiology of shoulder stiffness

The natural history for shoulder stiffness is a topic for debate in scientific literature. It has been proposed a three-phase pathogenesis which advocates the spontaneous recovery [9]. The first phase is characterized by increasing pain during movement, without significant loss of motion. It typically lasts 2–9 months. In phase two, lasting 4–12 months, the shoulder ROM become progressively limited, pain occurs mostly at end range. The third and last stage can last up to 4 years and consists of resolution with persisting pain at end range, which soothes only with complete resolution and full-ROM recovery [9]. However, spontaneous remittance of this kind of pathology is not always achieved and persistent impairment requires invasive treatment. In a recent review of the literature [32], it has been reported that self-limiting capsulitis theory can be strictly applied to partial rotator cuff tears, but idiopathic disease has a few chances of spontaneous resolution. In the work by Wong et al. [32], it was demonstrated that no evidence is present concerning the progression form stiffness to recovery phases and that no resolution can be achieved in absence of supervised treatment. From a pathogenic point of view, inflammation is the main event which characterizes capsulitis. Although causes of inflammation are easily retrieved for secondary capsulitis, including tendon inflammation, trauma, subacromial impingement and bursitis, for primary disease it is not so clear how an inflammatory process begins [32]. As a result of this, capsular scarring and retraction occurs, yielding to stiffness of the joint for severe reduction of capsule laxity. Histologically, it has been demonstrated that along with mast cells, T and B cells, the presence of myofibroblasts characterizes the affected capsule structure, and these cells seem to be responsible for contraction and retraction of the tissue [15].

Postural changes in Parkinson's disease

Camptocormia is the term which is commonly used to describe an accentuated and particularly severe "stooped" position, with an accentuated thoracic kyphosis. Its prevalence among PD affected patients ranges between 3 and 17.6% [1], thus representing a significant problem for this population. It has been hardly defined, although several authors proposed a 45° turaco-lumbar flexion as a threshold for diagnosis of camptocormia [1, 2]. This alteration is often reversible, with the possibility for the patient to stay up straight if asked or lay down on a semi-rigid

surface. However, not in all cases camptocormia is reversible, with axial rigidity showed by neurological examination [3, 21]. Concerning joint movement, PD usually yields an elastic rigidity of muscles, caused by induced spasticity, as a result of lacking of inhibition pathways. This alteration is easily individuated throughout physical examination of motor function of the limbs. It is defined passive rigidity, as it is present when a passive motion of a joint is made, and contraposes to the active rigidity, which occurs during active motion. Elbow rigidity is paradigmatic in PD, retrieved in most of the patients affected [28].

The role of posture on shoulder movement

Kinematic of the shoulder must be always considered in a more complex view than simple scapula-humeral motion, where arm elevation and full range of motion of the arm are allowed by the combined movement of the humerus, the scapula, the clavicle, the thoracic wall and thoracic spine [7, 18]. More specifically, the single arm elevation is the result of scapulo-humeral elevation, scapular posterior tilt and extension of thoracic spine. It has been reported that thorax extension gives a fundamental contribution, as full elevation of both arms requires at least 15° of spine extension [5], while unilateral arm elevation requires at least 9° extension. Furthermore, the monolateral arm elevation yields to thoracic lateral flexion to the contralateral side. It was already described in last '80 s [5, 16] that reduced thoracic motion or increased kyphosis of the thoracic spine negatively influenced the range of movement of the superior limb, during elevation. This is due to the altered rest position of the scapula on the costal wall, which results more abducted, with increased anterior tilt [6, 22], limiting rotational potential of the clavicle during shoulder motion. It has been demonstrated through several tools, including radiography [11], physical examination [12, 19] and digital methods [17], that increased anterior tilt with reduction in the rotation of the scapula during arm elevation puts the acromion in anterior position, with consequent impingement. The result of such a complex mechanism is the development of subacromial impingement [5, 18]. In conclusion, increased kyphosis can yield a humeroacromial impingement syndrome, since anterior tilting of the scapula occurs along with kyphosis itself, resulting in inflammation of the bursa, shoulder pain and reduction of movement. This observation is also corroborated by results of studies investigated effects of postural rehabilitation, in which is shown that improvement in thoracic posture leads to an improvement in shoulder elevation [23, 31].

Shoulder pathology in parkinsonian patients

A study carried out in 2008, by Koh et al. [20], studied ultrasound (US) evaluation of shoulder in patients affected by PD. Findings of the study showed that almost 70% of the patients (22 out of 33) had rotator cuff tendon tears, most commonly at the supraspinatus tendon, and 9 of these had concomitant adhesive capsulitis with symptoms of frozen shoulder. Interestingly, the mean duration of the PD was significantly longer in the group of patients with tendon tears, when compared to those without [20]. Subsequently, a study by Yucel and Kusbeci [33], evaluated findings at magnetic resonance imaging (MRI) in patients affected by PD. They showed that changes in acromioclavicular joints were observed in patients with mild PD, along with high frequency of subcoracoid effusion. Those findings were not retrieved in the control group. Furthermore, full-thickness tears of the supraspinatus were observed in groups with mild and severe PD, while no rotator cuff tear was present in the controls [33]. In addition to these, a more relevant finding was observed: the acromiohumeral distance, a measurement of the width of the subacromial space, was significantly reduced in patients with mild and severe PD, when compared to healthy controls [33]. In 1989, two studies investigated for the first time, the role of shoulder pathology in Parkinson's disease [4, 26]. According to the results showed by Riley et al. [26], frozen shoulder was present in a minority (12.7%) of patients affected by PD with a main duration of 7 years. From a temporal analysis, it was reported that the majority of the cases of frozen shoulder had an onset between 2 years before diagnosis of PD and 3 years after. Moreover, in 16 out of 19 cases, the onset symptoms of PD were at the upper limb, ipsilateral to frozen shoulder [26]. Akinesia of the superior limb was the symptom of presentation in the majority of the cases of frozen shoulder [26].

Discussion

Patients affected by PD show an increased trend in developing modification of scapula-humeral kinematic and rotator cuff tears, as well as adhesive capsulitis [20, 33]. We believe that postural alteration observed in PD is the *primum movens* for development of shoulder pathology, since anterior tilt of the scapula, which occur with the increasing of thoracic kyphosis, yields to a subacromial impingement. The progressive narrowing of the subacromial space, which has been described by previous studies [33], is the main mechanical factor responsible for rotator cuff tendinopathy, since shoulder movement in the context of an acromiohumeral impingement unavoidably leads to rotator cuff shearing on the inferior edge of the acromion. In short term, the shear stress produces a tendinopathy of the rotator cuff, that can

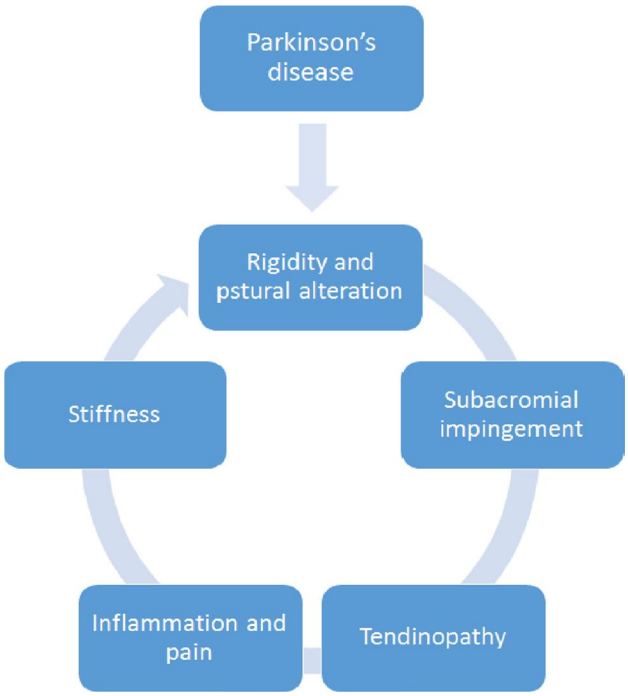

Fig. 1 Main hypothesis for the development of causative loop between postural alteration and shoulder stiffness

be symptomatic or not. In mid to long term, tendinopathy produces inflammation and pain of the shoulder, causing the patient not to mobilize properly the joint. Immobilization increases the inflammation and pain, causes muscular and capsular atrophy, with severe stiffness of the joint. In the context of such an inflamed rotator cuff, it is really common that a partial or a full-thickness tear of the tendons occurs, with a worsening of the pain, inflammation and avoidance of mobility. This clinical pattern characterizes the so-called frozen shoulder. According to our hypothesis, a closed loop is then created, as the rigidity of the shoulder causes further alteration in the posture (e.g., rounded shoulders), which worsens the impingement syndrome and so on (Fig. 1). All the findings reported by included studies lead us to evaluate the possibility that shoulder rigidity, which is commonly found in patients affected by Parkinson's disease, may be the result of a complex biomechanical process involving thoracic posture and scapular angulation. It is not clear, whether akinesia of the shoulder (the most common presentation of parkinsonian patients with frozen shoulder [26]) is a cause of frozen shoulder, or pre-existing alterations of the shoulder can yield someway to limitation of movement. All these findings suggest that there is a possibility that shoulder alteration can be a presentation symptom of Parkinson's disease, although no clinical study had confirmed this yet. In accordance with the study by Riley et al., a short communication by Cleeves and Findley [4] revealed that some patients affected by PD had reported shoulder pain as a presenting symptom of Parkinson's disease. Cleeves and Findley [4] stated that patients had a convincement that shoulder pain was a presenting symptom of PD, though a time of up to 1 year had passed between pain and the onset of other usual symptoms of PD. Similar findings have been also proposed more recently by Stamey et al. [29] in a letter to the editor. A strong limitation in defining this hypothesis is the lack of studies which correlate the postural changes of PD and shoulder pathology. However, in our clinical experience, it has been frequently found that patients presenting to the orthopedic consultant complaining shoulder pain and stiffness, with increased kyphosis and a somewhat impaired motion pattern, were subsequently diagnosed with Parkinson's disease. By a sub-analysis of these cases, it resulted that diagnosis advocated by the orthopedic specialist, concerning shoulder complain, was actually adhesive capsulitis, or frozen shoulder. From this observation and from the analysis of results presented in the literature, we formulated the hypothesis that shoulder pain in patients affected by PD is derived from postural changes, and that early alterations may lead to initial acromiohumeral impingement, with a clinical presentation of frozen shoulder. Main limitation of the present investigation is the lack of systematic data concerning PD and shoulder stiffness. The paper retrieved reported heterogeneous data in term of endpoint and outcome measure. By contrast, according to our knowledge, this is the first paper formulating the present hypothesis.

Conclusion

According to our analysis, there is a concrete scientific base on which our hypothesis can be formulated. The interrelationship between postural alterations observed in Parkinson's disease and shoulder pain, rotator cuff tendinosis and capsulitis, should be further investigated, in order to advocate the posture-shoulder closed loop hypothesis as a characteristic of Parkinson's disease.

Compliance with ethical standards

Conflict of interest The authors have no conflict of interest to disclose.

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