



Surgical treatment of spinal disorders in Parkinson's disease

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Received: 9 October 2017 / Accepted: 24 January 2018 / Published online: 3 February 2018
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Abstract

Purpose Most patients suffering from Parkinson's disease (PD) exhibit alterations in the posture, which can in several cases give rise to spine deformities, both in the sagittal and the coronal plane. In addition, degenerative disorders of the spine frequently associated to PD, such as spinal stenosis and sagittal instability, can further impact the quality of life of the patient. In recent years, spine surgery has been increasingly performed, with mixed results. The aim of this narrative review is to analyze the spinal disorders associated to PD, and the current evidence about their surgical treatment.

Methods Narrative review.

Results Camptocormia, i.e., a pronounced flexible forward bending of the trunk with 7% prevalence, is the most reported sagittal disorder of the spine. Pisa syndrome and scoliosis are both common and frequently associated. Disorders to the spinopelvic alignment were not widely investigated, but a tendency toward a lower ability of PD patients to compensate the sagittal malalignment with respect to non-PD elderly subjects with imbalance seems to emerge. Spine surgery in PD patients showed high rates of complications and re-operations.

Conclusions Disorders of the posture and spinal alignment, both in the sagittal and in the coronal planes, are common in PD patients, and have a major impact on the quality of life. Outcomes of spine surgery are generally not satisfactory, likely mostly due to muscle dystonia and poor bone quality. Knowledge in this field needs to be consolidated by further clinical and basic science studies.

Graphical abstract These slides can be retrieved under Electronic Supplementary Material.

Key points

1. Parkinson's Disease sagittal deformity
2. Surgical treatment of sagittal deformity
3. Complications

[Citation]

Name	Description	plane	Rigid/flexible
Camptocormia	Marked flexion originating from thoracolumbar spine (minimum 45°) resolved by supine position	sagittal	flexible
Antecollis	forward flexion of neck and head	sagittal	flexible/rigid
Pisa syndrome	lateral flexion of at least 10° which can be resolved completely by passive mobilization or lying supine	coronal	flexible
Global kyphosis	Increased kyphosis in all the spinal segments, with active compensation in the pelvis, that is retroverted	sagittal	rigid
Pelvic kyphosis	bend forward the whole trunk, including the pelvis, not retroverted	sagittal	flexible/rigid
Degenerative scoliosis	at least one spinal curve with axial rotation of vertebrae, not corrected in contralateral bending or in the supine position	coronal	rigid

[Citation]

Take Home Messages

1. Parkinson's Disease sagittal deformity correction is possible in selected cases
2. Early and late complications are common, because of the general impairment of these patients and because of the biomechanical situation of their spine
3. Despite high failure rates, surgery is in most cases the only chance to live an acceptable life for these unlucky, suffering patients.

[Citation]

Keywords Parkinson's disease · Camptocormia · Spine deformities · Osteoporosis · Degenerative · Scoliosis

Electronic supplementary material The online version of this article (<https://doi.org/10.1007/s00586-018-5499-y>) contains supplementary material, which is available to authorized users.

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Introduction

Most patients suffering from Parkinson's disease (PD) exhibit alterations of the posture, which can in several cases give rise to spine deformities, both in the sagittal and the coronal plane [1]. The stooped posture featuring flexion of hips and knees as well as to some degree of the trunk and

neck is indeed one of the most recognizable signs of PD. In addition, PD is frequently associated to degenerative disorders of the spine, such as spinal stenosis and sagittal instability, which can further impact the quality of life of the patient. Osteoporosis is also a typical feature of PD patients [2, 3], and the poor bone quality negatively affects spinal pathology and leads to an increased risk of fractures, and therefore, axial pain. Together with neurological impairment and subsequent poor gait control, these features realize a vicious cycle in which spine surgery is often needed, but with high failure and revision rates.

In recent years, spine surgery has been increasingly performed on PD patients with the aim of treating the spine-related disorders and restoring a correct spinal alignment, with mixed results. The aim of this narrative review is to analyze the spinal disorders associated to PD, and the current evidence about their surgical treatment. Although several patients exhibit deformities and postural abnormalities in both the sagittal and in the coronal plane, disorders in the two anatomical planes were addressed in separate paragraphs. A special focus was kept on the pelvis and the spinopelvic alignment, which gained attention in recent years [4]. Finally, the available studies about the surgical treatment of spinal disorders associated to PD have been reviewed, with emphasis on the reported complications and on their risk factors.

Methods

A review of the available literature about spinal deformities in PD patients was performed in PubMed (<http://www.ncbi.nlm.nih.gov/>). Keywords used included “Parkinson’s disease”, “Parkinson” in combination “scoliosis”, “sagittal imbalance”, “deformity”, “spinopelvic”, “camptocormia”, “deformity correction”, “Cobb angle”, “coronal”, “complications”. The retrieved papers, as well as their lists of references, were reviewed in search for potential added knowledge about the topic. The selected studies were analyzed in a non-systematic way, and a narrative review of the literature was conducted based on them.

Results and discussion

Clinical evaluation of PD

Prior to the review of the spinal disorders associated to PD, a brief description of the clinical variables and scores used for the assessment of the severity of the disease and which will be mentioned in the following paragraphs is here reported. The Hoehn and Yahr scale [5] classifies the progression of the severity of the symptoms of PD in five stages, ranging

from unilateral involvement with minimal or no functional disability (stage 1) to bed or wheelchair confinement (stage 5). Nowadays, the most commonly used scale is the Unified Parkinson’s Disease Rating Scale (UPDRS), which is composed by several sections (evaluation of behavior and mood; self-evaluation of daily activities such as speech, handwriting, hygiene, etc.; evaluation of the motor functions; complications related to the therapy; stage of severity following the aforementioned Hoehn and Yahr scale; evaluation of the dependency on others during daily activities following the Schwab and England scale [6]). A modified version of the UPDRS, aimed to improve its consistency, is also widely used [7]. Although being included as an item of the UPDRS scale, the Hoehn and Yahr score is still frequently explicitly reported in research papers.

In addition to age and sex and the severity of the PD symptoms, other clinically relevant variables usually taken into account in scientific papers are the duration of the symptoms, i.e., the time period between the diagnosis of PD and the collection of the data, the possible presence of back pain which is assessed by means of its conventional grading systems and the possible treatment with L-DOPA and/or deep brain stimulation. Besides, the risk of fall is sometimes evaluated and quantified by means of specific scales, such as that by Tinetti and colleagues [8].

Sagittal alignment

A stooped posture is a characteristic sign of PD, and is observable in all patients [1]. This posture generally features a small or moderate flexion of hip and knee joints, and forward bending of the trunk. In approximately 7% [9] of the PD patients, this bending reaches a high magnitude, which is conventionally named as camptocormia [9, 10] (Fig. 1). In comparison with sagittal disorders of elderly subjects not suffering from PD, camptocormia is generally characterized by a substantial flexibility of the curve, which can be corrected mostly or fully if the patient lies supine. A standardized criterion was proposed for the diagnosis of camptocormia, consisting in a marked flexion originating in the thoracolumbar spine (minimum 45°) which can be almost completely resolved in the supine position [1]. Nevertheless, available studies about camptocormia generally suffer from a low comparability due to the different, non-standardized diagnostic criteria employed. On average, PD patients with camptocormia had higher age [10] and more severe symptoms [11–13] if compared to patients with limited sagittal spinal curvature. In general, camptocormia does not respond well to treatment with L-DOPA [11, 12]. In a case–control study on PD patients with camptocormia and age-matched patients also suffering from PD but not exhibiting the sagittal deformity [12], the authors concluded that PD with camptocormia shows specific characteristics such



Fig. 1 Lateral clinical picture of a camptocormic Parkinson's disease patient. The sagittal malalignment is compensated by active knee flexion and pelvic retroversion

as axial rigidity, disorders and instability during gait, limited response to L-DOPA also for the axial symptoms, which cannot be observed in the control group and may be due to a significantly higher neurological dysfunction, although other authors hypothesized that it might simply reflect a more severe parkinsonian phenotype or a longer duration of the symptoms [1].

Recently, researchers analyzed the spine sagittal alignment of PD patients by means of radiological techniques and expressed the forward bending in terms of orthopedic quantities such as the sagittal vertical axis (SVA) [14, 15]. The authors found that SVA significantly increased with progressing Hoehn and Yahr stage and UPDRS score, and that the lumbar spine was in a kyphotic alignment in the majority of the patients [14]. The thoracic kyphosis was also found to be significantly higher than in an age-matched cohort of asymptomatic subjects, and increasing with the severity of the symptoms [14]. High SVA was associated with older age and with the female sex [15].

Another sagittal disorder associated to PD is the forward flexion of neck and head named antecollis (Fig. 2) [1]. Antecollis contributes to the stooped posture and is observable to



Fig. 2 Lateral clinical picture of a typical antecollis in a Parkinson's disease patient. The patient is unable to look forward without external help

some extent in the majority of PD patients. In severe cases, antecollis can become a fixed deformity of the cervical spine [13], whereas it is more frequently associated with increased axial tone but with passive neck extension still achievable. Moon and coworkers [16] could not find an association between antecollis, measured radiographically as the cervical kyphosis, and the severity of PD symptoms. Nevertheless, increased cervical kyphosis was significantly associated to global sagittal malalignment. The above-mentioned conditions are summarized in Table 1.

Spinopelvic alignment

In the last years, a substantial effort has been devoted to the study of the pelvis anatomy and orientation and to its relation to the sagittal alignment of the spine, in asymptomatic and in particular in degenerative subjects. A few

Table 1 Summary of the most common spinal sagittal and coronal pathological findings in Parkinson's disease patients

Name	Description	Plane	Rigid/flexible
Camptocormia	Marked flexion originating from thoracolumbar spine (minimum 45°) resolved by supine position	Sagittal	Flexible
Antecollis	Forward flexion of neck and head	Sagittal	Flexible/rigid
Pisa syndrome	Lateral flexion of at least 10° which can be resolved completely by passive mobilization or lying supine	Coronal	Flexible
Global kyphosis	Increased kyphosis in all the spinal segments, with active compensation in the pelvis, that is retroverted	Sagittal	Rigid
Pelvic kyphosis	Bend forward the whole trunk, including the pelvis, not retroverted	Sagittal	Flexible/rigid
Degenerative scoliosis	At least one spinal curve with axial rotation of vertebrae, not corrected in contralateral bending or in the supine position	Coronal	Rigid

recent studies analyzed this aspect in patients suffering from PD, and generally found a higher prevalence of spinopelvic disorders with respect to subjects of similar age [14, 15, 17]. Although the available literature is still scarce and no definitive conclusions can be drawn, evidence about a lower ability of PD patients to compensate the sagittal malalignment with respect to elderly non-PD subjects exhibiting imbalance seems to be emerging [14, 15]. In the study mentioned above, Watanabe et al. [14] found that PD patients had more difficulties in activating compensatory mechanisms such as pelvic retroversion, decrease of thoracic kyphosis and knee flexion to limit the imbalancing effect of the loss of lumbar lordosis with respect to an age-matched cohort. PD patients tended to bend forward the whole trunk, including the pelvis, likely due to muscle degeneration in addition to the neurological disorder. This has been observed and classified by Lamartina and Berjano in 2014 as “pelvic kyphosis” in the classification of sagittal imbalance based on compensatory mechanisms [18]. In contrast, Bissolotti and colleagues [17, 19] found that several PD patients showed significant differences from the normality in terms of the ratio between sacral slope and pelvic incidence indicating pelvic retroversion, thus showing a certain ability of these patients to compensate the sagittal imbalance. As a matter of fact, the investigated populations considerably differed among these studies, confirming the need for further research about this relevant topic. Spinopelvic compensation of sagittal malalignment is described in Fig. 3.

Coronal alignment

Alterations in the coronal alignment of the spine are frequently seen in PD patients [1]. Two distinct types of anomalies were observed: the Pisa syndrome, consisting in a marked lateral bending of the trunk, which is generally flexible and can be resolved in the supine position; and scoliosis, which is defined following the consolidated diagnostic criteria commonly used in orthopaedics and radiology based on X-rays images. Doherty et al. [1] proposed a precise

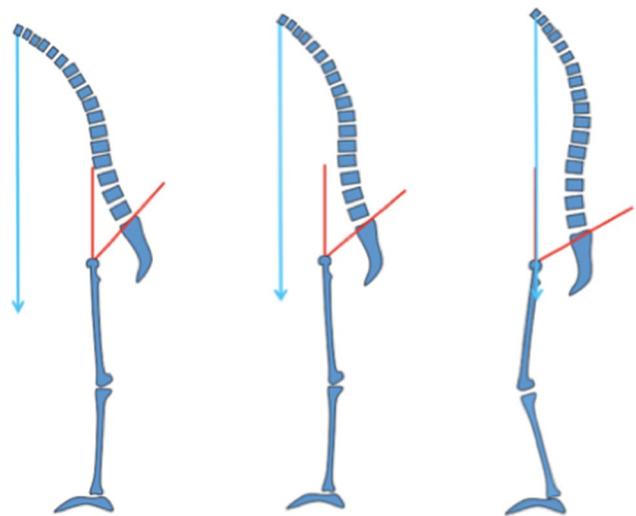


Fig. 3 Schematic drawing of pelvic compensation in sagittal malalignment. The blue line is the sagittal vertical axis (SVA), that should normally fall on the femoral heads. The red angle is the pelvic tilt, that measures pelvis orientation in the sagittal plane, and is a positional parameter. Moving from left to right it is possible to appreciate the progressive restoration of “normal” SVA position due to pelvic retroversion

diagnostic criterion for the Pisa syndrome, which consists of a lateral flexion of at least 10° which can be resolved completely by passive mobilization or lying supine. The Pisa syndrome, therefore, differs from scoliosis, which is a true spine deformity which features at least one curve, exhibiting axial rotation of vertebrae, which is not corrected in contralateral bending or in the supine position. In PD patients is usually a degenerative scoliosis, since PD is a neurodegenerative condition and usually happens in the adulthood or elderly. Aebi described this pathological entity in 2005 [20] and according to this classification, scoliosis in Parkinson's Disease is a secondary degenerative scoliosis (type 3).

Degenerative scoliosis is found more frequently in PD patients rather than in the general elderly population [21]. For example, Baik and colleagues [22] performed a

radiographic analysis of 97 PD patients, and found scoliosis in 32 subjects, whereas the prevalence of scoliosis in old non-PD subjects was found to range between 6 and 30% [22–24]. An association between the direction of scoliosis and the laterality of PD symptoms was hypothesized [21, 25], but contrasting findings were also reported [22, 26]. As a matter of fact, animal studies showed the development of scoliosis in dogs [25] and rodents [27, 28] correlated with the side of the major symptoms, with a tendency toward a contralateral bending with respect to the symptoms. However, the scarce data available about human patients are rather contradictory [29] but tend to show a lack of association between scoliosis direction and laterality of the symptoms [22, 30].

Scoliosis in PD patients usually shows a single curve, is more common in female patients rather than in males and is not respondent to L-DOPA treatment [22, 30]. The Cobb angle of the major curve was shown to be associated with the severity of the symptoms measured with both the Hoehn and Yahr stage as well as the UPDRS scale [30], showing that the progression of PD may also involve the risk of a worsening of the scoliotic curve. Interestingly, the prevalence of scoliosis was associated to the age of the patients but not to the duration of the symptoms [30]. Sagittal misalignment of the spine was not found to be correlated neither with the prevalence of scoliosis [31], nor with its Cobb angle in scoliotic subjects [30], even if both disorders are frequently present in the same subject. Coronal anomalies of PD patients are displayed in Table 1.

Spine surgery in PD patients

Thanks to technical innovation and improvement in patients' pre and postoperatively general care, spine surgery is able to address severe and huge deformities involving the whole spine. Despite this, clinical outcomes of PD patients treated affected by spinal pathology tend to be worse, showing high rates of complications and re-operations. As a matter of fact, the surgical treatment has become only recently a viable option and is still nowadays selected by a minority of patients, resulting in a general scarcity of available data.

Both the sagittal misalignment and scoliosis may be the object of the surgical treatment, as well as other disorders such as spinal stenosis and segmental instability. The surgical treatment of camptocormia was also reported, but only for a few cases [32, 33]. Decompression associated to short fusion is also frequently performed in patients with spinal stenosis and no or minor spinal deformities [34].

Complications

Surgical complications can be divided in early and late complications. Typically, early complications concentrate

in the immediate postoperative period and are related to Parkinson's systemic impairment. A recent publication by Baker and colleagues [35] showed an increased risk of hemorrhagic, cardiac, pulmonary, genitourinary and neurologic complications in PD patients compared to non-PD population undergoing spine surgery. Other well-known early post-surgical complications of PD patients, regardless to the kind of surgery, are: delirium, orthostatic hypotension, venous thromboembolism. Moreover, it has been demonstrated by the same group in a different study, based on the same population, that in PD patients the length of the construct is related to morbidity and mortality [36].

Late complications are the dreadful hardware-related biomechanical complications, responsible for the outstanding revision rates of instrumented fusions in PD patients. Implant loosening, pseudoarthrosis and loss of correction with progressive deformity have been reported [37–39].

The largest multicentric study currently available, which included 48 patients subjected to a long fusion, from the upper thoracic spine to the sacrum or the pelvis in 44 patients and with a median length of 14 vertebrae [39]. This large study is based on the results of seven spinal centres in France, who collected PD patients operated for spinal deformities over almost 30 years. The rarity of this kind of patients is one of the main obstacles in the collection of clean, reliable data.

The results of this large study showed that 20 of the 48 patients required a revision surgery, for a total of 35 revisions, 28 of which due to pseudoarthrosis and junctional kyphosis. It is interesting to note that, despite the high rate of complications and although only one-third of the patients had a good functional outcome and spinal alignment, 78% of the patients were satisfied or very satisfied. In another retrospective study [34], 23 PD patients suffering from degenerative lumbar instability, degenerative scoliosis, post-traumatic kyphosis and other disorders were surgically treated. Corpectomies were performed in four patients, whereas pedicle subtraction osteotomy in one patient. 52% of the patients had major complications and 33% had revision surgery. The authors concluded that a correct restoration of the sagittal balance is imperative to achieve successful results. Despite the high rate of complications and revisions, 74% of the patients were satisfied or very satisfied with the clinical outcome. Similar findings were found in other retrospective studies [40, 41]. Generally, poor surgical outcomes were also reported [42].

One of the possible causes which have been frequently attributed to the high failure rates is the poor bone quality in PD patients [34, 38, 39, 42]. Koller and coworkers [34] reported that PD patients have brittle and osteopenic bone. In a systematic literature review, osteoporosis was found to have an odds ratio of 2.61 in PD patients in comparison with healthy controls [43]. Evidence of a lower bone mineral

density (BMD) and higher fracture risk was also reported [43]. Factors determining the lower BMD included vitamin D deficiency with secondary hyperparathyroidism and reduced sunlight exposure. L-DOPA might also be involved in the reduced BMD, and the risk could be dependent on the dose [44]. It should be noted that, in addition to the low bone quality, the increased risk of fractures was determined by the high risk of falls due to postural instability, dysfunction of autonomic nervous system and motor disorders. The neurological physiopathology of PD affects patients' stability in several ways. First, postural control is often impaired for the altered muscular tonus of axial muscles, unresponsive to L-DOPA medication. Second, dysautonomic dysfunction is responsible for orthostatic hypotension and reduced proprioception inputs to the brain. Third, PD patients with severe clinical progression show a reduced ability to modify the motor program of gait, becoming at high risk of falls in case of unexpected obstacles or unplanned changes of the planned walk. All these different mechanisms put PD population at high risk of complications when spine surgery is required, while on the other side can be the initiating factor for clinical progression of degenerative spine pathology and for subsequent surgical indication [45, 46].

As a matter of fact, surgical treatment of spinal deformity in PD patients suffers from a high rate of revisions and complications, but seems to provide satisfactory self-reported results in the majority of the patients, this indicating an improvement in the quality of life which makes it a viable option in selected patients. Although there is no statistical evidence of this phenomenon, due to the small number of patients undergoing this special kind of surgery, the qualitative evidence of the study from Bouyer and colleagues [39] is witnessing how miserable should be the clinical condition of these patients, if they are happy to face such a heavy burden of complications and reoperations. The available scoring systems for PD patients are focusing on motor performance and pure neurological features, and it is just by physical examination that the clinician can quantify the everyday impairment of these patients, who are frequently isolated at home, unable to walk, to look at themselves in a mirror, to eat normally for the forward flexion of the trunk and neck. These non-standardized impairment is usually the driving force for surgical indication in such frail and risky patients.

The poor clinical outcome has been related to the natural progression of the pathology [39, 42], and only limited improvements might be foreseen in the next future. However, some risk factors which should be considered to select the patients who may benefit from the surgical treatment have been identified. Based on the literature review, Sarkiss and coworkers [47] indicated that poor outcome was associated to: older age, thoracolumbar kyphosis, osteoarthritis of the hip, and increasing level of camptocormia. Risk factors related to the surgery itself were postoperative SVA greater

than 5 cm, inadequate sacropelvic fixation, and poor level selection.

Conclusions

Disorders of the posture and spinal alignment, both in the sagittal and in the coronal planes, are common in PD patients, and have a major impact on the quality of life. The current armamentarium available to surgeons offers several options for the surgical treatment of spinal disorders in PD patients; nevertheless, poor clinical outcomes related to the high rates of complications and revisions are frequently reported, but on the other hand most of patients are personally satisfied of the surgery, reporting a better quality of life compared to the preoperative period. As a matter of fact, peculiar characteristics of PD such as muscle dystonia and poor bone quality challenge even modern surgical techniques and implants. Knowledge in this field needs, therefore, to be consolidated by further clinical and basic science studies.

Compliance with ethical standards

Conflict of interest All authors declare that they have no conflict of interest.

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