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Pisa syndrome, Bonsai syndrome and Dropped Head Syndrome in Parkinson's disease: Treatment with Botulinum Toxin Injection

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ABSTRACT

Lateral axial dystonia (LAD) has been described in patients with Parkinson's disease (PD) as Pisa syndrome (PS). In patients with PS, the trunk is pulled by the paraspinal muscles to the lateralized side. In these cases, botulinum toxin (BTX) should be injected into the paraspinous muscles of the lateralized side. But, in some cases of LAD, the enlarged paraspinal muscles push contralaterally. When, this happen BTX should be injected into the paraspinous muscles opposite the lateralized side. BTX was injected into the paraspinous muscles of the lateralized side in PS, opposite of the lateralized side in BS and into the sternocleidomastoid muscles or scalene muscles in dropped head syndrome (DHS). We experienced three cases of PS, two cases of BS and four cases of DHS. Seven cases out of eight were improved. Treatment with BTX is useful for PS, BS and DHS in PD. When treating LAD in PD, PS or Bonsai syndrome (BS) should first be identified.

Keywords

Parkinson's disease, Dystonia, Pisa syndrome, Bonsai syndrome, Dropped head syndrome.

Introduction

Lateral axial dystonia (LAD) has been described in patients with Parkinson's disease (PD) as Pisa syndrome (PS) [1]. In patients with PS, the trunk is pulled by the paraspinous muscles to the lateralized side [2]. In these cases, botulinum toxin (BTX) should be injected into the paraspinous muscles of the lateralized side [3]. However, in some cases of LAD, the enlarged paraspinous muscles push contralaterally. When this happens, BTX should be injected into the paraspinous muscles opposite the lateralized side [4].

The leaning tower of Pisa is pulled by gravity, but Bonsai trees are tightened by wires. Similarly, in Bonsai syndrome (BS), LAD is a result of tension created by enlarged contralateral paraspinous muscles. In cases with dropped head syndrome (DHS), BTX should be injected into the sternocleidomastoid muscles or scalene muscles.

Case Report

BTX was injected into the paraspinous muscles of the lateralized Neurol Res Surg, 2018

side in PS, opposite of the lateralized side in BS and into the sternocleidomastoid muscles or scalene muscles in DHS. We experienced three cases of PS, two cases of BS and four cases of DHS. Seven cases out of eight were improved (Table 1).

Case four- 55 year-old female, house wife

From the end of 30's, only her left shoes became worn out. She was diagnosed as Parkinson's disease when she was 44, L-dopa + carbidopa was administered. But for the side effects, her prescription was changed to cabergoline 3mg. Her trunk was gradually tilt to the right, she came our hospital for the second opinion. On the first visit, the volume of her left paraspinous muscles was enlarged and press upon right and front. Lidocaine was injected to her right paraspinous muscles, but her trunk tilt was worsened. She did not want more treatment (Figure 1).

Discussion

Cabergoline and pergolide stimulate D1 and D2 receptor, Pramipexole stimulate D2 and D3 receptor. D1 receptor agonist often induced involuntary movement [5]. In this point, there is no denying the reason that "dropped head syndrome" and LAD are induced by Cabergoline [6].

| No. | Sex | Age | Onset | Diagnosis (H & Y) | Symptoms | Caused muscle | Туре | Medication | Injection | Prognosis |
|-----|-----|-----|-----------|----------------------|------------------------------------|---|-----------------|---|--|-----------|
| 1 | F | 72 | 65 | PD (III) | Tilt to the left, Camptocormia | Left paraspinal muscle | Pisa | Pramipexole, Selegiline, L-dopa + Carbidopa | BTX injection to the left paraspinal muscle | Improved |
| 2 | F | 58 | 52 | PD (III) | Tilt to the right, Camptocormia | Right paraspinal muscle | Pisa | Pramipexole, Selegiline, L-dopa + Carbidopa | BTX injection to the right paraspinal muscle | Improved |
| 3 | М | 48 | 25 | PD (III) | Tilt to the right, Camptocormia | Right paraspinal muscle | Pisa | Pramiipexeol, Selegiline, Amantagine, L-dopa + Carbidopa | BTX injection to the right paraspinal muscle | Improved |
| 4 | F | 55 | Late 30's | PD (III) | Tilt to the right, Camptocormia | Left paraspinal muscle | Bonsai | Cabergoline | Lidocaine injection to the right paraspinal muscle | Worsened |
| 5 | М | 77 | 67 | PD (IV) | Tilt to the right, Camptocormia | Left paraspinal muscle | Bonsai | Pergolide, Selegiline, L-dopa + Carbidopa | BTX injection to the left paraspinal muscle | Improved |
| 6 | F | 86 | 80 | PD (IV) | Tilt to the right, Antecollis | Sternocleidomastoid (right dominant) | Dropped head | Trihexyphenidyl, L-dopa + Carbidopa, | BTX injection to the sternocleidomastoid muscle (right dominant) | Improved |
| 7 | F | 80 | 67 | PD (IV) | Antecollis | Scalane | Dropped head | Pramipexole, Selegiline, L-dopa + Carbidopa | BTX injection to the scalane muscles | Improved |
| 8 | М | 93 | 89 | PD (IV) | Antecollis | Scalane | Dropped head | L-dopa + Carbidopa | BTX injection to the scalane muscles | Improved |

Table 1: All cases.

No: number, M: male, F: female, PD: Parkinson's disease, H & Y: Hoehn and Yahr scale, BTX: Botulinum toxin.



Figure 1: Case four (Bonsai syndrome). The enlarged paraspinous muscles push to the right. Bonsai trees are tightened by wires. Similarly, in Bonsai syndrome, lateral axial dystonia is a result of tension created by enlarged contralateral paraspinous muscles.

Regarding the mechanism of onset for dropped head syndrome in Parkinson's disease, imbalances in tonus of the anterior and posterior neck muscles (excessive muscle tonus of the anterior neck muscles compared to the posterior neck muscles) has been suggested. Difference between sensitivity against dopamine receptor in both paraspinal muscles influenced the onset of LAD [7]. In conclusion, treatment with BTX is useful for PS, BS and DHS in PD. When treating LAD in PD, PS or BS should first be identified.

Disclosure

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. The Kawasaki Municipal Tama Hospital (approval number: C7173) approved this study.

Informed consent

Informed consent was obtained from all individual participants included in the study.

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