



Case report

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Pisa Syndrome and Neurosyphilis: A Case Report in Taiwan

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Abstract

Background: Pisa syndrome, defined as more than 10 degrees tonic lateral flexion in upright spine without any significant associated vertebral rotation resembled the leaning tower of Pisa, is considered to be related to neuroleptics. Unfamiliarity of this disease, many neuro- and orthopedic surgeons can lead to unnecessary diagnostic and interventions.

Method: We report an extremely rare case of neurosyphilis presenting with PISA syndrome in order to assess the clinical presentation and treatment.

Results: A 49-year-old man, who has neurosyphilis and psychotic disorder due to general medical condition, treated by valproic acid 500 mg twice and quetiapine 200 mg once daily for as an unchanged regimen for the past 1 year. Five days after treatment with clozapine for poor controlled psychosis, he was noted leaned to one side. Physical examinations revealed a severe right truncal shift associated with left cervical tilt. We discontinued all medication except adding an anticholinergic drug (2 mg biperiden twice daily). After 7 days, the deformity disappeared with normal body posture. His psychosis flared up and we rechallenged clozapine 25mg daily. He was noticed leaned to one side again.

Conclusions: orthopedic and neurological surgeons visiting patients with abnormal postures of the trunk need to evaluate medications especially with neuropsychiatric disease. Even drugs with a minimum risk of extrapyramidal symptoms, such as clozapine, can cause Pisa syndrome in even short duration and low dosage.

Keywords

Pisa syndrome; Neurosyphilis

Introduction

Pisa syndrome (PS) is defined as more than 10 degrees of tonic lateral flexion in the upright spine without associated vertebral rotation, like the leaning tower of Pisa which is first described by Ekblom et al. in 1972 [1,2]. PS often considered related to longterm treatment with neuroleptics.

We report an extremely rare case of neurosyphilis presenting with PISA syndrome in order to assess the clinical presentation and treatment.

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Case Report

The 49-year-old man has neurosyphilis and psychotic disorder due to general medical condition since the age of 48. He had been admitted three times within a period of 1 years because of exacerbation of mood or psychotic episodes. The main presenting symptoms of each episode were aggression, religious delusion, thought of being controlled, and auditory hallucination. He was treated by valproic acid 500 mg twice and quetiapine 200 mg once daily for as an unchanged regimen for the past 1 year. His psychiatric symptoms were under controlled in this regimen without hospitalization.

However, he was admitted this time for persisted auditory and visual hallucination, physical and verbal aggression. We have titrated quetiapine dosage but muscle rigidity, sialorrhea, tilting to right side were noted. We then shift quetiapine gradually to clozapine 50mg daily for poor controlled psychosis and intolerable side effects.

Five days after treatment with clozapine, he was noticed progressively leaned to one side with a tonic right flexion of the trunk and left flexion of the neck but himself seemed unaware of and unconcerned by his body change. On physical examination, he had no evidence of dynamic (e.g., active spasms, posturing with jerks or tremor, paradoxical muscle groups associated with an abnormal posture or asymmetrical contractions of paraspinal muscles). Neurologic exam was unremarkable for motor and sensory deficits. Under the impression of PISA syndrome, we discontinued all medication except adding an anticholinergic drug (2 mg Biperiden twice daily). After 7 days, the deformity disappeared with normal body posture. His psychosis flared up and we rechallenged clozapine 25mg. He was noticed progressively leaned to one side again.

Discussion

The prevalence rate of PISA syndrome was reported to be 9.3% in women, 6.4% in men [3]. The risk factors were female sex, old age, organic brain disorders and a history of treatment with conventional neuroleptics [4]. The pathophysiology of PS is related to dopaminergic system.

PS has been described in several diseases including Parkinson disease, progressive supranuclear palsy, and Huntington disease [5-7]. PS was also described as a side effect of neuroleptics such as risperidone, quetiapine, ziprasidone, paliperidone, aripiprazole, and cholinesterase inhibitors [8-13]. Clozapine, has lower binding affinity to Dopamine 2 receptors, seems less risk of PS. Furthermore, clozapine is also reported to be a treatment of PS [14].

To our knowledge, this is the first case report of PISA syndrome in neurosyphilis. The patient has the risk factors as organic brain disorders and treated with antipsychotics. PS occurs five days after initiating clozapine treatment in a low dosage of 50mg daily and improved after discontinuation. Relapse of PS after rechallenging clozapine in a lower dosage is also noted.

In summary, orthopedic and neurological surgeons visit patients with abnormal postures of the trunk need to evaluate medications especially with neuropsychiatric disease. Even drugs with a minimum risk of extrapyramidal symptoms, such as clozapine, can cause Pisa syndrome in short duration and low dosage.

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