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Acute Parkinsonism in Young Adult Following Streptococcal Infection

Shahedah Koya Kutty, Ummu Afeera Zainulabid

[1]Neurology Clinical Specialist, International Islamic University of Malaysia, Kuantan, Malaysia; [2]Internal Medicine Specialist & Medical Lecturer, Department of Internal Medicine, Kulliyyah of Medicine, International Islamic University of Malaysia, Kuantan, Malaysia

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Abstract

Acute parkinsonism following streptococcal infection is rarely reported. To date, there is paucity of literature of post streptococcal immune mediated acute parkinsonism and dyskinesia among adult. We report a case of 18 years old woman, who presented with acute parkinsonism after streptococcal tonsillitis. The patient who was previously well presented with a two-month history of intermittent fever, associated with sore throat and sudden onset of behaviour changes. She has been found to be quieter and respond slowly to questions and commands in the preceding one week prior to admission. Features of secondary parkinsonism was noted during examination. Her blood counts, viral screenings and anti-NMDAR were unremarkable. Her lumbar puncture revealed an opening pressure of 10 cm CSF with normal constituents. The ASO titre was raised up to 400IU/ml. Electroencephalogram (EEG) shows mild encephalopathy changes and MRI brain shows normal finding. She was initially treated initially for meningoencephalitis with intravenous acyclovir 500mg q8h and ceftriaxone 2g q12h, concomitant with intravenous methylprednisolone 1g daily for 3 days. Intravenous immunoglobulin was started on her in view of poor response to the above treatment, which later resulted in significant and rapid clinical improvement. Upon clinic review at 3 months, her tremor, bradykinesia, and rigidity resolved. Her medication was able to be weaned off and she was referred to the ENT team for further evaluation for tonsillectomy. This case illustrates the importance of recognising post-streptococcal infection as aetiology of acute parkinsonism in young adults, to avoid treatment delay. As illustrated in our case, the prognosis was excellent with prompt immunomodulatory therapy. [*Bangladesh Journal of Infectious Diseases, December 2020;7(2):110-112*]

Keywords: Parkinsonism; PANDAS; encephalitis lethargica; akinetic rigidity; post-infectious immune mediated parkinsonism

Correspondence: Dr. Ummu Afeera Zainulabid, Internal Medicine Specialist & Medical Lecturer, Department of Internal Medicine, Kulliyyah of Medicine, International Islamic University of Malaysia, Kuantan, Malaysia, **Email:** ummuafeera@iium.edu.my
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Introduction

It has been recognised that a variety of dyskinetic extrapyramidal movement disorders may occur following post streptococcal infection in children, however it is rarely reported in adult. The spectrum of post streptococcal movement disorders extends beyond the common type of chorea and tic and includes the presence of myoclonus, dystonia and tremor. We report our experience of post-streptococcal infection in an 18-year-old young lady, which manifested as acute parkinsonism and dyskinetic movement disorders post tonsillitis with evidence of streptococcal infection. To date, there is paucity of literature of post streptococcal related acute parkinsonism and dyskinesia among adult. This case may guide clinicians to consider immune-mediated response post streptococcal infection as one of the aetiologies of sudden-onset parkinsonism in an otherwise healthy young adult and prompt the necessary treatment.

Case Presentation

This patient presented to Emergency Department with acute behavioural changes which occurred for the past 1 week as she become quieter than her norm, with slowness in answering questions, and reduced facial expressions. She also complained of having insomnia that makes her distress. She was also found to have abnormal movements involving the face, lips and jaws, bilateral upper limbs and lower limbs. Further history, patient had intermittent on off fever associated with relapse remission sore throat. She has past medical history of recurrent tonsillitis for past 10 years. There is no confusion, no hallucination and no delusion. There was no history of similar episodes in the past, any seizures, urinary incontinence, head injury, previous psychiatric illness or intellectual disability. Developmental and family histories is unremarkable. Patient was afebrile and tachycardic with heart rate of 100 beats per minute on presentation. Her clinical examination revealed abnormal movement i.e. orofacial dyskinesia, oromandibular dyskinesia as she protruding her tongue involuntary and unable to retract in, truncal dystonia with arching of back and twitching of bilateral upper limb and paddling movement of lower limbs. Glucometer reading was normal. A comprehensive neurological examination revealed an alert young woman with a Glasgow Coma Scale of 15/15. Her Mini Mental State Examination (MMSE) was 30/30. Specific neurological revealed evident of axial rigidity, reduce blinking, right hand tremor, bradykinesia, cogwheel rigidity more prominent on right side, with normal power. There was marked hypertonia of lower limb with no clonus. Reflex were brisk over the knee bilaterally and 2+ over the ankle. Cranial nerve I-XII was intact with no cerebellar signs. Other systemic review was unremarkable. Throat was not injected neither swollen. Initial blood investigation revealed white cell counts of $10.38x10^9$ /L (normal value: 4-10), haemoglobin 11.8g/L (normal value: 10-15) and platelet count of 363 $x10^9$ /L (normal value: 150-400). The Antistreptolysin O titre (ASOT) was raised up to 400IU/ml (normal value: < 160 IU/ml). Renal function, liver function and coagulation profile were normal. We proceeded with a lumbar puncture which had an opening pressure of 10cm H^2O (normal). Her cerebrospinal fluid analysis showed normal cell counts, biochemistry and negative culture results. Throat swab culture and sensitivity shows normal throat floral. Tumour marker, NMDAR result, and connective tissue screening were negative. Electroencephalography (EEG) shows mild diffuse generalised cerebral disturbance by virtue of excessive theta activity. MRI brain shows no significant abnormality. As part of the work up, a CT scan of the thorax, abdomen and pelvis was carried out to look for underlying malignancy which has negative finding. In the ward patient was empirically treated for meningoencephalitis and completed Ceftriaxone and Acyclovir for a week. She was intolerant to amantadine and bromocriptine which later discontinued due to increase of stiffness. Her symptoms of acute parkinsonism were better controlled with tab Clonazepam 0.5mg OD, 25/100 mg of Carbidopa/Levodopa OD three times a day. Patient was started initially with methylprednisolone however there is minimal improvement and treatment escalated to intravenous immunoglobulin which showed marked resolution of symptoms. Upon the clinic follow up at 3 months, her tremor, bradykinesia, and rigidity resolved. Her medication was able to wean off and she was referred to ENT team for further evaluation of the needs of tonsillectomy.

Discussion

Post streptococcal secondary parkinsonism rarely reported in adolescent. Our patient fulfilled the criteria of parkinsonism as she portrayed to have bradykinesia, tremor and rigidity[1]. The only positive result in her case is marked raised of ASOT titre. There is no history of taking drugs that can lead to secondary parkinsonism. MRI brain done is reported as normal.

With a mixed features or parkinsonism and movement disorder, it raised a suspicious of related autoimmune antibodies in her case. She didn't improve much with initial antibiotic treatment and methylprednisolone. However, a positive remarkable recovery was noted after initiation of immunomodulatory agent of intravenous immunoglobulin. The recovery features of the patient symptoms made it is unlikely to be contributed by a genetic or degenerative cause.

We initially attribute her features could be part or variant of Paediatric Autoimmune Neuropsychiatric Disorders associated with streptococcal infection, termed PANDAS which commonly described in paediatric age group. PANDAS was rarely reported in adolescent and it was first described by Swedo et al in 1998.[2] The interesting features of our patient is the unusual age of late adolescent as most literature documented the mean age of onset for this disease was 6.3 years[2]. Moreover, she didn't fulfil full criteria of PANDAS as she didn't have the features of tics or obsessive-compulsive features. In addition, role of immunomodulatory therapy in PANDAS remained controversial.

This case also raised a possibility diagnosis of encephalitis lethargica (EL) with a features of sleep disturbance with association of acute parkinsonism, despite there is no full-blown encephalitis seen nor ocular features characteristic of EL. The disease which was first described in 1916 by Von Economo captured a specific symptoms of sleep disturbance, lethargy, extrapyramidal symptoms together with neuropsychiatric disorders. [3] One case report published in 2010 showed a remarkable positivity improvement in an EL-like patient who treated with an early administration of intravenous immunoglobulin[4].

The exact role of treatment with immune-modifying therapies in post infectious immune mediated consequence of streptococcal infection remained judicious to case by case basis. It is suggested that immunoglobulin contains anti-idiotypic antibodies, absorbs complement and prevents it from binding to tissue, decreases immune globulin synthesis, block receptors, inhibits lymphocyte proliferation, increases suppressor cells, and inhibits binding of autoantibodies to antigen[5]. In this case, our patient showed marked positivity of response to treatment after given intravenous immunoglobulin.

Conclusion

The case illustrates post infectious streptococcal acute parkinsonism in adolescent. It also highlights the important of diagnosing this condition in adolescent despite of its rarity. Symptoms of recurrent sore throat or tonsillitis with later manifested as parkinsonism should raise the possibility of post infectious immune mediated consequence of streptococcal infection. Our patient represents an unusual age during her first presentation which many of the cases were described in paediatric age group. Further larger studies are needed to establish standard treatment approach for this disease.

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References

1. Postuma RB, Berg D, Stern M, et al. MDS clinical diagnostic criteria for Parkinson's disease. *Mov Disord*. 2015;30(12):1591-1601
2. Swedo SE, Leonard HL, Garvey M, et al. Pediatric autoimmune neuropsychiatric disorders associated with streptococcal infections: clinical description of the first 50 cases. Am J Psychiatry. 1998;155(February):264-271
3. Dale RC, Church AJ, Surtees RAH, et al. Encephalitis lethargica syndrome: 20 New cases and evidence of basal ganglia autoimmunity. Brain. 2004;127(1):21-33
4. Forensic AJ, Pathol M. Encephalitis Lethargica With Quick Response to Immunoglobulin. 2010;31(4):1-3.
5. Bayry J, Thirion M, Misra N, et al. Mechanisms of action of intravenous immunoglobulin in autoimmune and inflammatory diseases. Neurol Sci. 2003;24(SUPPL. 4):217-221