

Twenty Year and Still Isolated Form of Restless Arm with Hypometabolism of Brain in Caudate Bilaterally

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Authors' contributions

This work was carried out in collaboration of all authors. Authors KA and SB designed the article and wrote. All authors managed the literature search and wrote the first draft of the manuscript with assistance from author OME, VA and VE. All authors read and approved the final form.

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Case Study

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ABSTRACT

We reported a patient with restless arm but without any other extremity involvement during 20 year period. There was hypometabolism of brain in caudate bilaterally. Interestingly, the restlessness had not progressed to involve other hand or his legs during this long time period. Only restless arm syndrome (RAS) is very rare. In describing the patient, we hope to raise awareness about RAS and avoid misdiagnoses and inappropriate investigations.

Keywords: Restless arm syndrome; FDG PET; hypometabolism; caudate.

1. INTRODUCTION

Restless arm symptoms are characterized as disagreeable sensations in the upper extremities that are relieved by movement [1]. Diagnostic

criterion is the same as restless leg syndrome (RLS) and the symptoms most commonly occur at night. Upper extremity symptoms can develop in a large proportion of the patients with RLS, but are rarely the initial symptom [1-4]. Hereby we

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report a case of restless arm syndrome (RAS) without any leg symptom during 20 year follow-up. Only restless arm syndrome is very rare and our case is the first report with abnormal FDG PET exam and without any leg symptom during 20 years.

2. CASE REPORT

A 61 year old man described restlessness occurring only in the right sided upper extremity and disrupting sleep during all day. Sensation of itching, tingling, and congestion was described too. There was no restlessness in his legs. Polysomnographic profile and its impact on nocturnal sleep had not been investigated but whenever he went to a doctor, different drugs including haloperidol, anti-inflammatory drugs, anti-cholinergic, gabapentin and pregabalin had been prescribed previously. He had been taken any drug mentioned above for very short time and gave up using them because he did not feel well. A careful review of his history revealed a relationship between his symptoms and resting position. Any type of arm movements was decreasing the symptoms. Repeated neurological evaluations were normal except choreo-atetoid like movement which showing restlessness and could be easily controlled by him in right hand. There was not any cognitive problem but it was really worsening his life quality. There was no family history of movement disorders and no history of smoking, alcohol or using excessive caffeine. He had no history of any other disease or medication except the drugs prescribed for this complaint previously. Results of laboratory tests, including serum chemistry, complete blood cell count, thyroid tests, iron studies, and vitamin B₁₂ levels, were normal. Cranial and cervical MRI examinations and EMG examination made to exclude the secondary causes of restless arm were completely normal. PET exam showed that there was hypometabolism in caudate bilaterally (Fig. 1). For his restless arm, he took pramipexole 0.375 mg daily and the discomfort in his right arm completely abated by follow-up in 2 weeks. It was diagnosed as RAS because the symptoms had a circadian pattern and were improved by movement and dopaminergic therapy. He is still taking pramipexole for relaxation.

3. DISCUSSION

In present case, the patient described an urge to move the right arm usually accompanied or caused by uncomfortable and unpleasant sensations in the arms and the urge to move or

unpleasant sensations begin or worsen during periods of rest or inactivity such as lying or sitting. The complaints were partially or totally relieved by movement of arm. The diagnosis was RAS according to consensus essential diagnostic criteria [5]. His symptoms had been started nearly 20 years ago awakening him during sleep and were still present only in the right extremity. Interestingly, the restlessness had not progressed to involve other hand or his legs during this long time period.

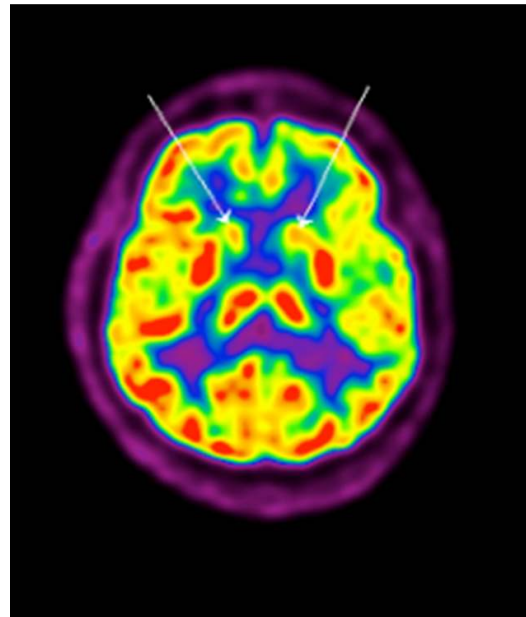


Fig. 1. PET exam showed that there was hypometabolism in caudate bilaterally

In movement disorders, daily used imaging studies such as MRI and CT exams mostly do not show any abnormality. PET investigation is an alternative for differential diagnosis of movement disorders because central dopaminergic and overall brain functional activity are altered to different degrees in idiopathic Parkinson's disease and atypical parkinsonian disorders, periodic limb movement disorder and restless legs syndrome [6,7]. Although there is no certain fact about RAS, previous reports points to central mechanisms involving dopaminergic systems in RLS and some hypothesize an abnormal hyper excitability along the entire spinal cord, especially its lumbosacral and cervical segments as the primary cause of periodic leg or arm movements accompanying RLS [8]. Both FDG and FDOPA PET scans show similar findings and can demonstrate patterns of neuronal dysfunction that are specific to a

particular movement disorder in the patients [6-10]. It is well known that mean caudate and putamen FDG uptake are reduced in the RLS patients compared with control subjects [6,7]. The equal affection of the caudate and the putamen differs, for example, from the dopaminergic dysfunction in Parkinson's disease, which affects the putamen earlier and more severely than the caudate [6,10]. There was some published material showing PET findings of the patients with RLS, but there was not with RAS [6-10]. The FDG-PET study was performed during our follow-up and decreased FDG uptake in caudate nucleus bilaterally similar to findings seen in RLS was shown. This finding was the first report showing caudate involvement in RAS. We think that this is an important finding supporting the relationship between RLS and RAS.

4. CONCLUSION

In summary, we reported a patient with RAS but without any other extremity involvement during 20 year period. In describing the patient, we hope to raise awareness about RAS and avoid misdiagnoses and inappropriate investigations.

CONSENT

Consent and disclosure form from the patient for any figure in this article have been received.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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