

TARDIVE DYSTONIA AND DIFFUSE IDIOPATHIC SKELETAL HYPEROSTOSIS

H F K Chiu, J Leung, K W Li

ABSTRACT

Tardive dystonia is a late side effect of long term neuroleptic treatment. A Chinese woman with tardive dystonia in the form of retrocollis for ten years showed radiological evidence of diffuse idiopathic skeletal hyperostosis (DISH) in the cervical spine. This association has not been previously reported in the literature. It is postulated that the retrocollis led to an abnormal stress of the cervical spine where excessive bone formation occurred. Hence tardive dystonia may act as a risk factor for DISH in patients who have an ossifying diathesis.

Keywords: Tardive dystonia, diffuse idiopathic skeletal hyperostosis, Chinese

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INTRODUCTION

Tardive dystonia is a late side effect of long term neuroleptic treatment⁽¹⁾. The clinical manifestations include torticollis, retrocollis, blepharospasm, opisthotonus and dystonia of limbs and trunk. Diagnostic criteria includes a history of neuroleptic treatment, presence of chronic dystonia, absence of family history of dystonia and exclusion of secondary causes of dystonia⁽²⁾.

Diffuse idiopathic skeletal hyperostosis (DISH) is a well-recognized condition affecting middle-aged and elderly people⁽³⁾. DISH may occur concomitantly with ankylosing spondylitis⁽⁴⁾ and rheumatoid arthritis⁽⁵⁾. Its association with tardive dystonia has not been reported.

CASE REPORT

Madam A is a 45-year-old Chinese woman with history of schizophrenia for 20 years. Since then she was put on various neuroleptics including chlorpromazine, trifluoperazine and thioridazine. However, she had frequent relapses of her psychosis over the years, necessitating long term maintenance of neuroleptics. She developed retrocollis 10 years ago which had become persistent. She frequently bumped into objects when she walked as she could hardly look ahead of her because of the retrocollis. She had neck pain for the past 10 years. Otherwise, her past health was good and there was no family history of mental illness or movement disorders.

Mental examination revealed no active psychotic features. On physical examination, she had a marked retrocollis (Fig 1) and there was limited range of motion of the neck but no neurological deficit. Laboratory investigations including complete blood picture, renal function test, liver function test, thyroid function test, calcium, phosphate, caeruloplasmin and blood copper level were all normal. Slit lamp examination of the eyes was also normal. The clinical picture was compatible with the diagnosis of tardive dystonia.

Fig 1. – Patient with marked retrocollis



Radiographs of the cervical spine (Fig 2) showed flowing anterior ossification of 5 contiguous vertebrae, preservation of the disc height and absence of apophysial joint bony ankylosis and sacroiliac joint erosion or sclerosis.

DISCUSSION

The diagnostic criteria of DISH suggested by Resnick⁽⁶⁾ include:

1. The presence of flowing calcification and ossification along the anterolateral aspect of at least four contiguous vertebral bodies.
2. The presence of relative preservation of intervertebral disc height in the involved vertebral segment and the absence of extensive radiographic changes of "degenerative" disc disease.

Department of Psychiatry
Prince of Wales Hospital
Shatin
Hong Kong

H F K Chiu, MRCPsych (UK)
Lecturer

Department of Diagnostic Radiology
Prince of Wales Hospital

K W Li, FRCR (UK)
Medical Officer

J Leung, MRCPsych (UK)
Psychiatrist

Correspondence to : Dr H Chiu

3. The absence of apophyseal joint bony ankylosis and sacroiliac joint erosion, sclerosis or intra-articular osseous fusion.

Fig 2. – Radiograph of the cervical spine



The radiographic features of this case fit into these criteria. Although DISH is reported to be a common condition in various studies, its prevalence in Chinese is uncertain.

Tardive dystonia is a relatively rare condition and so far only about 170 cases have been reported in the literature⁽¹⁾. The occurrence of DISH in this patient with tardive dystonia may be due to coincidence. Unfortunately an X-ray was not done before the dystonia to let us attribute more conclusively that the cervical vertebral changes were due to the tardive dystonia. Another possibility is that the persistent retrocollis led to an abnormal stress of the cervical spine. In patients with an ossifying diathesis, excessive bone formation might occur in these areas of stress. This is in accordance with the view of Resnick that DISH may represent an ossifying diathesis that causes excessive bone formation at skeletal sites subject to normal or abnormal stresses⁽⁶⁾. Further evidence of a bone-forming tendency is the propensity of these patients to develop ossification after surgery or in response to skeletal alterations accompanying co-existent diseases like rheumatoid arthritis⁽⁶⁾. Hence tardive dystonia may act as a risk factor in patients who have an ossifying diathesis. Further studies in patients with dystonia would be worthwhile to explore this hypothesis.

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