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An Unusual Cause of Cervical Kyphosis

A Case Report

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Abstract:

Background Context: Acute fixed cervical kyphosis may be a rare presentation of conversion disorder, psychogenic dystonia, and potentially as a side effect from typical antipsychotic drugs. Haldol has been associated with acute dystonic reactions. In some cases, rigid deformities ensue. We are reporting a case of a fixed cervical kyphosis after the use of Haldol.

Purpose : To present a case of a potential acute dystonic reaction temporally associated with Haldol ingestion leading to fixed cervical kyphosis.

Study design : This is a case report.

Methods: A patient diagnosed with bipolar disorder presented to the emergency room several times with severe neck pain and stiffness. The neck appeared fixed in flexion with extensive osteophyte formation over a three month period.

Results: The patient's condition was resolved by a posterior-anterior-posterior surgical approach. It corrected the patient's cervical curvature from 88° to 5°.

Conclusion: Acute dystonic reactions have the potential to apply enough pressure on bone to cause rapid osteophyte formation.

Keywords: dystonic reaction; Haloperidol; fixed cervical kyphosis; osteophyte formation; severe neck pain; chin-to-chest deformity.

Introduction:

Acute dystonia, sporadic or sustained muscle spasms leading to abnormal postures, is a well-known side effect of neuroleptic drugs like Haldol.[1] A typical high-potency antipsychotic, Haldol is reported to cause dystonic reactions in 16% to 94% of patients.[1,2] Usually, the effects of acute dystonia can be reversed by cessation of Haldol, administering an anticholinergic agent IV, a dopamine agonist, or benzodiazepine/diazepam.[1,3] In this case report, we describe an unique case correlating a painful dystonic reaction leading to a fixed cervical kyphosis with rapid formation of anterior osteophytes. An operation was undertaken to correct the deformity.

Case Report:

A 31-year-old female, diagnosed with bipolar disorder and a documented history of intravenous substance abuse, presented to the emergency department with a stiff, painful

neck. Prior to this presentation, she was admitted to a psychiatric hospital and placed on Haldol. After reportedly watching other patients play monopoly within 72 hours of initiation of antipsychotic therapy, the patient experienced neck stiffness and abnormal forward neck flexion. Medical staff at the facility immediately ceased administration of Haldol and placed her on Cogentin, an anticholinergic drug. However, her symptoms persisted, prompting her transfer to the ER 28 days after the start of Haldol administration. She denied headache, numbness, or weakness. The patient had had Haldol during other inpatient stays on the psych unit. However, it had been over one year since her last inpatient psych admission. She showed no signs of radicular symptoms or spinal cord involvement. She reported radiation of neck pain bilaterally to her upper extremities. Her neck was flexed forward and was not passively correctable (Figure 1A). Unfortunately, no cervical spine xrays were taken prior her ER presentation. She was placed on Methadone and Klonopin for pain. At this time, neurology was consulted and suggested a diagnosis of acute dystonia as a complication of Haldol treatment.

Three months later, the patient was referred to our outpatient clinic for treatment of neck pain. Upon imaging of her cervical spine, appearance of osteophytes on the anterior margins of vertebral bodies and kyphosis were noted (Figure 1B). The kyphosis was not passively correctible. The patient underwent manual correction in the clinic on two occasions prior to surgery with no improvement in alignment. The patient complained of severe pain during the last attempted correction. Halo traction was discussed with the patient, but she did not feel she would be able to tolerate it given the discomfort noted during manual correction. Surgical treatment to correct the spinal abnormality was

determined necessary and posterior-anterior-posterior fusion of vertebral bodies C3 to C7 was performed.

A posterior approach was chosen first in order to allow osteotomies to be performed through the facet joints at three levels. Facet osteotomies were chosen in order to facilitate active correction of the kyphosis during the anterior approach. The facets were surprisingly stiff, but no obvious was noted.

Active correction of kyphosis was attempted prior to the osteotomies after general endotracheal anesthesia was induced with little improvement. After posterior elements had been exposed and the facet osteotomies were done, another attempt at correction was done with some improvement in the kyphosis. This facilitated the second stage of surgery which was an anterior approach. The anterior approach involved multiple discectomies, which allowed significant correction of the deformity.

After the surgery, the patient showed complete reversal of the fixed neck flexion. Preoperatively, the Cobb's angle, measured from the superior surface of C3 to the inferior surface of C7, of her cervical spine was 88°. After surgery it become 5° past normal cervical lordosis (Figure 2). Results from pre and post-operative Neck Disability Index (NDI) quality of life survey indicate statistically significant improvement after surgery (Table 1). Patient was seen 14 months after surgery where she refused follow up xray, but was evaluated clinically and found to have stable sagittal balance.

Discussion:

Haldol use in younger patients (ages 20 – 29) has long been associated with development of dystonic reactions including tonic contractions of the neck muscles.[2] Boyer et al reported 15 of 16 patients between the ages of 19 and 32 developed dystonia when given Haldol alone. [2] Haldol is a D2 receptor antagonist which blocks cerebral dopamine receptors, especially those in the nigrostriatal pathway.[6] The blockade of D2 dopamine receptors leads to increased cholinergic output from the striatum causing tonic contractions observed in dystonia.[7] Furthermore, pharmacokinetic aspects of Halodol metabolism offer a partial explanation for acute dystonia.[1,8] It was reported that one of the metabolites of Haldol is haloperiodol pyridinium (HP^+), a compound similar in structure to 1-methyl-4-phenylpyridium (MPP^+). MPP^+ is a neurotoxic metabolite of 1-methyl-4-phenyl-1,2,3,6-tetrahydropyridine (MPTP) which produces Parkinsonian-like symptoms of dystonia in humans.[8] In animal models, HP^+ produces similar effects to MPP^+.[8] Future research on this mechanism *in vivo* in humans may provide reasons for the many side effects of Haldol usage.

Osteophyte formation following fixed cervical kyphosis has not been reported as a result of acute dystonic contractions. Formation of osteophytes has been associated with chronic degeneration of intervertebral discs.[9] This phenomenon, based on Wolff's law, can be explained by observations that vertebrae in kyphotic regions of the cervical spine bear 6 to 10 fold increased stress compared to those in lordosis.[10] Within one to two weeks, increased stress on bone leads to bone remodeling and the formation of osteophytes.[9] Increased mechanical force exerts pressure on interstitial fluid flow activating osteocytes—the primary mechanoreceptors that regulate bone metabolism.[11]

As observed in our patient, the appearance of osteophytes within three months is indicative of a severe load on her cervical spine caused by the acute dystonia.

Other possible diagnoses for our patient's condition included: dropped head syndrome (DHS), myopathic head drop, idiopathic cervical dystonia (CD), and psychogenic dystonia. [12-14] As our patient suffered from acute neck pain and fixed cervical kyphosis, DHS and myopathic head drop were excluded. [12,13] Neither CD nor psychogenic dystonia could be excluded as potential reasons for the development of our patient's fixed cervical kyphosis. CD is the most frequent form of focal dystonia localized to the neck muscles as a result of tonic contraction. Cervical arthropathy is a common sequela of CD. [14] Patients usually have inciting trauma, genetic predispositions, and impaired basal ganglia function as in Parkinson's Disease. CD remains a potential cause for our patient's current presentation. Psychogenic dystonia may occur as a somatoform or conversion disorder. Features supporting psychogenic dystonia as a diagnosis include inconsistency of symptoms interfering with work but not with activities of daily living, symptoms not typical of an organically caused dystonia, abnormal postures that fade when distracted, and a prior history of somatoform disorders. [15] Our patient reported a significant decrease in quality of life and ability to care for herself in her prior condition. Growth of osteophytes is consistent with constant symptomatology placing immense stress on the vertebrae, and not a selective display of symptoms.

As with any pathology, physical therapy is the preferred option.[13] In our patient's case, her neck hung forward in a fixed position, rendering physical therapy inapplicable.

Surgical management of the patient's condition proved the most effective way to restore our patient's upright head position.

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Figure Legends:

Figure 1. Cervical radiographs are shown from onset of dystonic reaction (A) and four months later (B). Cobb's angles, measured from the superior surface of C3 to the inferior surface of C7, are reported; arrows show the osteophytes.

Figure 2. Cervical radiograph following surgical correction is shown. Patient's Cobb's angle was corrected from 88° to 5°.

Table 1. Quality of Life Score

	NDI (%) [a]
2 months Pre-Operative Quality of Life	84
3 months Post-Operative Quality of Life	62
Difference	22
MCID [b]	17.3

[a] NDI, Neck Disability Index. Higher score reflects greater disability. Mean NDI differences pre and post-operatively for anterior and posterior neck surgery have been reported as 19.16 ± 22.5 and 9.9 ± 16.8, respectively.[4]

[b] Minimal clinically important difference in NDI scores with a 95% confidence interval in patients who have undergone anterior discectomy and fusion surgery.[5]