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Postoperative hematoma in cervical spondylosis patient complicated with Huntington's disease: Case report and literature review

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Abstract

Hematoma is a life-threatening complication of anterior surgery in cervical spondylosis patients. Herein, we report a cervical spondylosis patient complicated with Huntington's disease, who developed unexpected neck hematoma after anterior cervical discectomy and fusion (ACDF) surgical treatment. During the debridement, we found no noticeable vessel lesions and concluded that the occurrence of postoperative hematoma might be due to the drainage displacement caused by excessive uncontrolled movements of the neck after the operation. The patient recovered well, and further literature review suggests that chorea secondary to Huntington's disease likely increases mechanical stress on the cervical spine, indicating an internal relationship between degenerative cervical spondylosis and Huntington's disease. Cervical spondylotic patients complicated with Huntington's disease can be treated with surgical intervention but need to be immobilized and under close observation.

Keywords

Cervical spondylosis, Huntington's disease, complications, hematoma, case report

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Introduction

Cervical spondylosis is a chronic and progressive cervical spine degenerative disease that causes spinal cord or nerve root compression and is usually manifested as neck pain, radiculopathy, myelopathy, or a combination of two or more of these symptoms.[1] A retrospective study on the global burden of low back pain and neck pain estimated that there would be more than millions of people worldwide who suffered neck pain symptoms for more than 3 months,[2] and the number was still increasing distinctly. As one of the main causes of neck pain, cervical spondylosis has imposed a significant impact on global health.[3] Huntington's disease (HD), on the other hand, is also one of the degenerative neurological disorders[4] and is much different from cervical spondylosis in symptoms. However, it has been reported that cervical spondylolisthesis and increased signal intensity of the spinal cord were observed in the cervical magnetic resonance imaging (MRI) of the patient with HD, which may pose a correlation between these two diseases.[5] Nevertheless, due to the lack of such case report,[6,7] here we present a rare case of cervical spondylosis complicated with HD who treated with surgical treatment and happened to have postoperative hematoma with no obvious vessel lesions, and the experience of treating this patient may be instructive in further similar cases to come in the future. This work has been reported in line with the Surgical CAse REport Guideline.

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Case presentation

A 41-year-old woman who worked as a farmer in the suburban area of Shanghai was admitted to our hospital complaining about discomfort in her neck and shoulders combined with occasional pain and numbness in both upper extremities for 1 year, and deteriorated quickly for the last 2 months. The patient had involuntary excessive motion in her head and both upper extremities since her childhood. She was diagnosed with HD later in her adolescence and did not receive proper treatment until administration. Since this illness, the patient had maintained a normal mental state and appetite, and her weight had not changed significantly. There was no problem with her defecation as well as sleep. Her personal history and family history were not remarkable.

Physical examination showed gait and balance disturbances. The patient had generalized chorea including the head, neck, and extremities, though her cognitive ability seemed to be average (Figure 1). Muscle hypertension was found in the neck, upper, and lower extremities. The muscle strength of both upper limbs was slightly lowered (between grades 4 and 5, documented as grade 5), left-hand muscle strength was grade 4, right-hand was grade 5, and bilateral lower limb muscle strength was grade 5. The patient had hyperreflexia of bilateral knee reflex and Achilles tendon reflex. Babinski signs were positive on both sides, while Hoffmann signs were weakly positive on both sides. Lasegue sign was negative bilaterally. The Japanese Orthopedic Association (JOA) score was 11, visual analog scale (VAS) of neck pain was 6 (Figure 1).

The patient underwent cervical MRI examination and found disk herniations and stenosis at C3/4, C4/5, C5/6, and C6/7 levels, with increased spinal cord signal intensity from the C3/4 to C5/6 level on the T2-weighted sagittal MRI view (Figure 1). Cervical X-ray showed degenerative features in multiple segments, hyperplasia of Luschka joints, and formation of vertebral osteophytes (Figure 2).

The patient was diagnosed as cervical spondylosis complicated with HD, and received anterior cervical discectomy and fusion (ACDF) under general anesthesia. The protruded intervertebral disk tissue was removed, as well as the hypertrophic Luschka joints and the osteophytes at the posterior edge of the vertebral body in order to enlarge the nerve root canal at the C4-C7 level. A hemovac drainage tube was placed in the prevertebral place. The intraoperative bleeding was about 50 ml, and the patient was returned to the ward under the protection of a cervical brace.

About 2 h after the operation, the patient repeatedly showed involuntary movement in the neck and two hands and occasional muscle spasms in the neck. After a while, the patient started to complain about dyspnea, and the oxygen saturation started to drop from 99% to 94% without oxygen inhalation. Twenty minutes later, subcutaneous congestion around the incision could be seen, and tissue tension was high around the neck, and the patient complained of feeling neck swelling (Figure 3). After aspiration using syringes, more than 50 ml of red liquid with tiny blood clots was drawn out, indicating a possible postoperative cervical hematoma, and an emergency surgical debridement was immediately arranged. The oxygen saturation of the patient was reduced to 84% during transfer, and immediate wound opening was done right into the operation room to relieve the pressure. During surgical exploration, a mess of dark red blood clots as well as exudation were seen under the platysma muscle (Figure 3). The intradermal length of the drainage tube was reduced to about 2 cm long, and the deep fascia sutures were compromised. We found muscle oozing blood obviously, and the chronic blood exudation in the deep prevertebral space, but no obvious rupture of vessels inside the wound. After hemostasis, two hemovac drainage tubes were

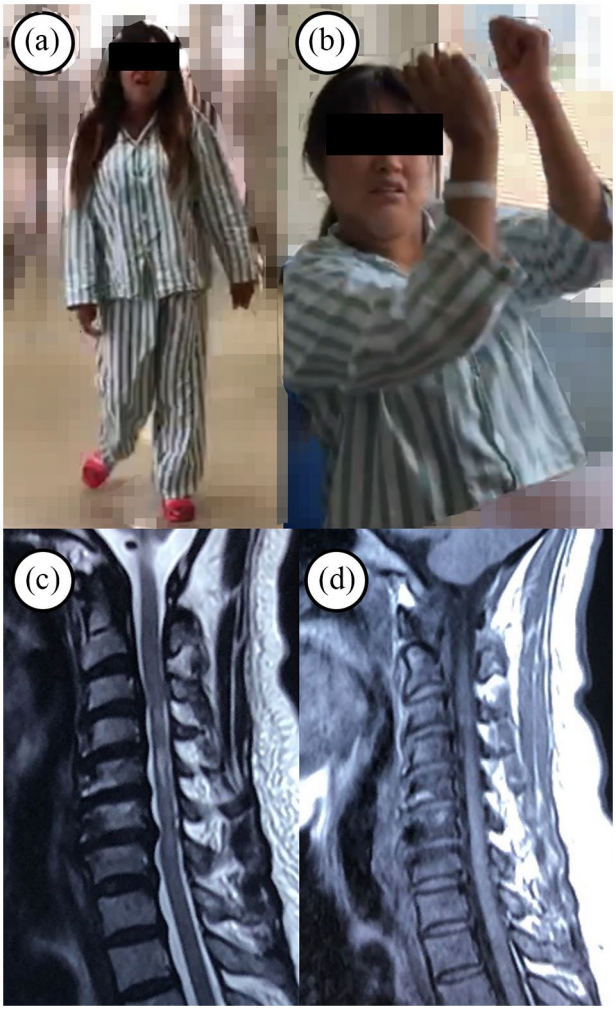

Figure 1. Gait and gesture images and MRI of the patient. (a) Gait and balance disturbances can be observed in this patient. (b) Involuntary and excessive movements of the upper extremities were observed. (c) T2-weighted sagittal MRI view of cervical spine. Disk protrusions were observed in C3 to C7 levels, and high signal intensities were observed in the spinal cord. (d) T1-weighted sagittal MRI view of the cervical spine.

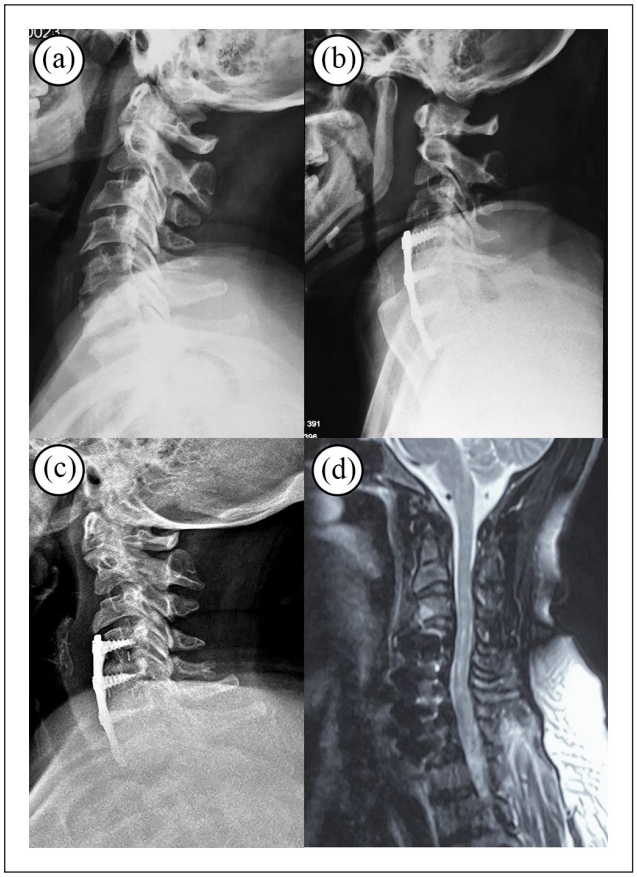

Figure 2. (a) Preoperative cervical X-ray image. (b) Postoperative cervical X-ray image (3 days after operation). (c) Cervical X-ray 6 months after the operation. (d) T2-weighted sagittal MRI image six months after the operation.

placed at the superficial muscular layer and prevertebral space respectively, and the patient returned to the ward under the protection of a neck brace. After 6 months, the JOA score recovered to 16, and the X-ray showed well-placed instrumentation (Figure 2). MRI showed no compression of the cervical spinal cord, and the increased spinal cord signal on the T2-weighted image reduced to some extent (Figure 2). Until the last follow-up, the patient is satisfied with the treatment and feels significant improvement after the surgery (Figure 3).

Discussion

Relation between Huntington's disease and cervical spondylosis

HD is the most common monogenic degenerative disease in developed countries, and it is also the most common form of hereditary dementia.[4] The manifestations are mainly progressive motor, cognitive, and mental symptoms.[4] The clinical and pathological manifestations of HD were closely related to neuropathy. However, current research mainly focused on the pathological changes of basal ganglia, with little attention paid to HD's effects on the spinal cord. Studies showed spinal cord atrophy appeared in the early stage of HD, suggesting that neurodegenerative changes in the brain and spinal cord occurred several years before the onset of the disease. But up till now, no significant correlation has been found between cervical spinal cord atrophy and changes in motor ability (Table 1).[8]

HD and cervical spondylosis may be interrelated. On one hand, involuntary motor disorders such as athletic cerebral

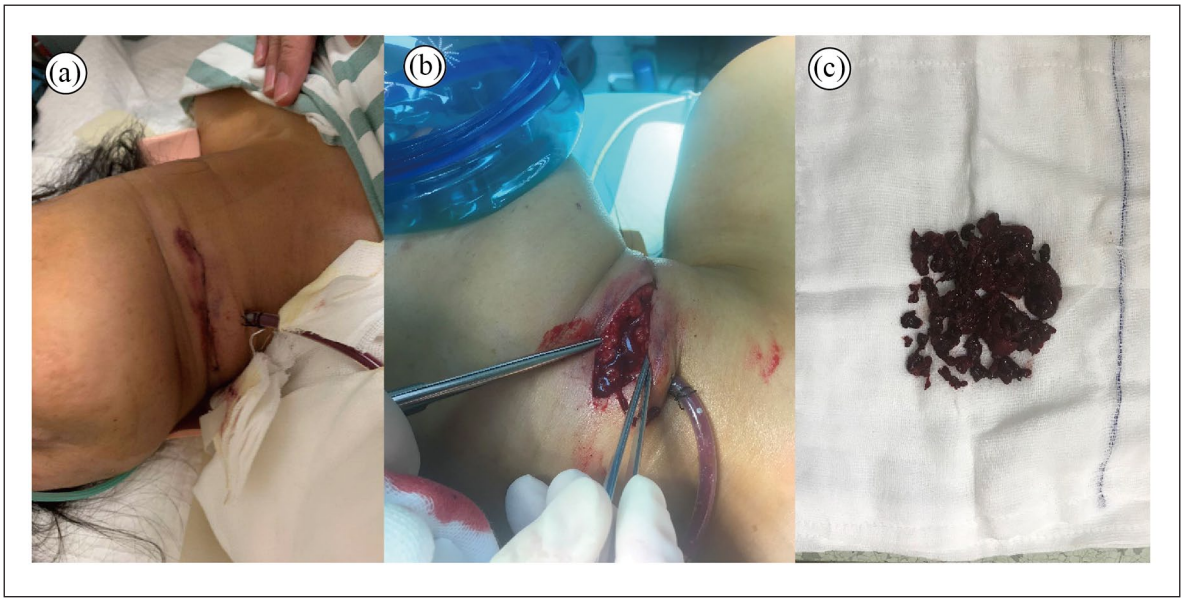

Figure 3. Debridement of postoperative cervical hematoma. (a) Neck swelling and bruise can be observed around the incision postoperatively, which indicates postoperative hematoma. (b) Hematoma can be seen during initial wound opening. (c) A mess of dark red blood clots as well as exudation were removed from the peri-vertebral space.

Table 1. Differentiation of symptoms of cervical spondylosis, HD, and motor neuron disease.

Symptoms of the motor system								
Cervical spondylosis			Huntington disease			Motor neuron disease		
Symptoms	Description	Literature	Symptoms	Description	Literature	Symptoms	Description	Literature
Spasticity and clonus	Spasticity and hyperreflexia are the hallmarks of clinical examination; in the more extreme cases, unsustained or sustained clonus is found.	Takagi et al.[9]	Chorea	Involuntary, excessive movements, which are short-lived and may appear semi-purposeful; spread from distal extremities to more proximal regions of the body. Activation of facial and neck muscles can cause head-turning, eye closure, and tongue protrusion, whereas the involvement of axial muscles can lead to extension and arching of the back.	Ghosh and Tabrizi[4]	Extremity weakness	Have difficulties in writing, turning keys in locks, raising the arm overhead, or tripping, and gradually resulting in impaired mobility and difficulty with activities of daily living.	Foster and Salajegheh[10]
Muscle weakness and atrophy	Arm or hand weakness, loss of manual dexterity.	Theodore[1]	Dystonia	Caused by sustained muscle contractions and increased muscle tone, leading to abnormal postures (such as torticollis, opisthotonos).	Ghosh and Tabrizi[4]		Muscle atrophy, stiffness, or fasciculations.	Foster and Salajegheh[10]
	The muscle atrophy of the upper limbs associated with CSA is generally classified into two subgroups, proximal-type CSA (with atrophy of the scapular muscles, deltoid, and biceps) and distal-type CSA (with atrophy of the triceps and muscles of the forearm and intrinsic muscles of the hand).	Luo et al.[11]		More common in juvenile HD than adult HD.	Quigley[12]	Respiratory failure	Manifested as dyspnea on exertion, orthopnea, morning headaches, and foggy thinking.	Foster and Salajegheh[10]
Gait	Gait and balance disturbances, especially in the absence of visual cues (Romberg's sign).	Theodore[1]	Bradykinesia, akinesia, and rigidity dominate	Bradykinesia (slowness of movement), akinesia (delay in initiation of movement), and rigidity dominate.	Ghosh and Tabrizi[4]	Bulbar onset	Dysarthria and dysphagia, and related communication problems and weight loss.	Foster and Salajegheh[10]
	Gait difficulty because of weakness or stiffness of legs.	Abbed and Coumans[13]	Gait	Ataxic, easy to fall down.	Ghosh and Tabrizi[4]	Motor syndrome	The motor syndrome can be distal, proximal, or mixed.	Statland et al.[14]
Upper motor neuron signs	Clonus, hyperreflexia, Hoffmann's sign, and Babinski's sign.	Theodore[1]	Myoclonus and tics	Brief, intermittent, stereotyped movements (such as blinks, sniffs) and head jerks, or they can be vocal (such as grunts, snorts).	Ghosh and Tabrizi[4]	Upper motor neuron signs	Brisk reflexes with spread to other regions, Babinski sign, Hoffmann reflex, or increased tone.	Statland et al.[14]
Lower motor neuron signs	Deep tendon reflexes related to that nerve are frequently decreased or absent.	Takagi et al.[9]		Generalized tonic-clonic is the most common seizure type of juvenile HD.	Quigley[12]	Lower motor neuron signs	Flaccid weakness, fasciculations, and diminished reflexes.	Foster and Salajegheh[10]

(Continued)

Table 1. (Continued)

Other symptoms								
Cervical spondylosis			Huntington disease			Motor neuron disease		
Symptoms	Description	Literature	Symptoms	Description	Literature	Symptoms	Description	Literature
Sensory symptoms	Sensory symptoms can include numbness, tingling, or burning pain in the dermatomal distribution supplied by that particular nerve. Shooting pain (often originates in the neck and passes through the arm to the fingers in a related manner, to the sensory distribution of the nerve).	Takagi et al.[9]	Cognitive symptoms	Deterioration with respect to tests of visual attention, psychomotor speed, and visuomotor and spatial integration.	Tabrizi et al.[9,15,16]	Sensory sparing	The onset is insidious, and the weakness is painless.	Statland et al.[14]
	Isolated to the neck or may radiate broadly, such as to the shoulders, head, chest, and back. Commonly radiating from the shoulder or upper back to the proximal arm, may also be accompanied by painful neck spasms.	Theodore[1]		The severance of cognitive symptoms ranges from subtle deficits to frank dementia.	Ghosh and Tabrizi[4]	Cognitive impairment	Executive dysfunction-impaired attention, working memory, organization, and planning.	Foster and Salajegheh[10]
	Paresthesia, numbness, or weakness that often (but not always) corresponds to dermatomal distributions of the affected cervical nerve root.	Theodore[1]	Psychiatric symptoms	Depression is the most common condition, and anxiety is the second, neither of which relates to the disease stage.	Ghosh and Tabrizi[4]	Behavioral impairment	Personality changes, obsessions, and disinhibition.	Foster and Salajegheh[10]
	Sensory loss in the hands or feet.	Theodore[1]	Behavioral difficulties	Apathy or lack of initiative, dysphoria, irritability, agitation or anxiety, poor self-care, poor judgment, inflexibility.	Walker[17]			
Incontinence	Patients with cervical myelopathy rarely have incontinence, although urgency, frequency, and hesitancy can be present in moderate and severe cases.	Takagi et al.[9]						

CSA: cervical spondylotic amyotrophy; HD: Huntington's disease.

palsy, torticollis, and Tourette syndrome were reported to cause cervical spondylosis, presenting as radiculopathy and/or myelopathy.[18] In particular, in patients with involuntary dyskinesia, the mechanical stress caused by lateral bending and axial rotation was more likely to cause the degeneration in the C3/4 and C4/5 levels,[18,19] which was also consistent with the case we reported. Therefore, we thought that HD's long-term involuntary activity might lead to a high load on the lower cervical spine, accelerating degeneration and eventually, cervical spondylosis was developed.[20–22] On the other hand, HD could cause dystonia, which increased susceptibility to cervical spondylosis. Dystonia mainly occurred in the later stages of HD, but it had also been reported as an early manifestation of HD.[23] The severity of dystonia was associated with the onset age of HD, with patients of younger onset age often having severer dystonia.[24,25] What's more, neuronal degeneration and neuromuscular junction injury of HD might lead to muscle atrophy,[26] which further reduces musculoskeletal stability and promote the development of cervical spondylosis.

The significance of this case to surgical treatment

It was worth noting that the hematoma after the operation might be closely related to HD in this case. In fact, the incidence of severe complications after ACDF surgery was low. In accordance with a retrospective analysis of a prospective study of 37,261 patients undergoing ACDF, the incidence of postoperative hematoma requiring reoperation was only 0.4%.[27] The operation process of our patient was smooth, and theoretically, the possibility of postoperative hematoma was minimal, but it was contrary to the situation of our case. Noticing the displacement of the drainage tube and the patient's history of HD, we speculated that the hematoma may be due to the increment of the patient's persistent involuntary activity, which involuntary neck movement increased significantly after waking from anesthesia that may cause the displacement of drainage tube, and eventually led to the occurrence of postoperative hematoma.

To date, we found few literature reporting the relationship between HD and the incidence of postoperative hematoma. There was also no supporting literature about whether the pathological mechanism of HD was related to coagulation and wound healing. So our case may add some evidence to the increased risk of complications of surgical patients with HD and may become a reminder to surgeons met with a similar circumstance.

The management and treatment of HD in surgical patients have reference value on clinical practice. Generally, symptoms of HD include unusual movements, cognitive and psychiatric disorders. In the light of the severity and symptoms of HD, drugs should be reasonably selected for symptomatic treatment. Psychiatric medicine might be reasonably used to avoid depression/mania and other mental symptoms after the operation. The involuntary muscle movement should be reduced after the operation to prevent some complications like a hematoma. According to preoperative drug usage, the dosage of muscle relaxants during anesthesia should be adjusted reasonably. Consulting from an experienced neurologist when treating chorea before the surgery would be helpful to develop a comprehensive treatment plan if it is available. On the other hand, if hemostatic agents like SURGIFLO Hemostatic Matrix were available, it can be used before closing the wound to minimize the possibility of postoperative hematoma.

In view of our particular case, for patients with cervical spondylosis complicated with HD, we suggested that we focus on the possible postoperative hematoma and prevent complications through intensive braking and close nursing. Fortunately, this patient achieved good results through surgical treatment, and the patient and her relatives were satisfied with the clinical outcome and praised for our timely interventions. However, the risk of surgical treatment in this case was still unclear due to lack of evidence. Therefore, the treatment choice for HD complicated with cervical spondylosis still needs further study.

Conclusion

Chorea secondary to HD likely increases mechanical stress on the cervical spine. There might be an internal relationship between degenerative cervical spondylosis and HD. Since the cervical spondylotic symptoms could be relieved after surgery, clinicians should be aware of the risk of postoperative hematoma. And patients receiving surgery should be immobilized and under close care after the surgery.

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Author contributions

All authors were involved in patient management, preparation of manuscript, and approving the final version.
ZZ, GW, and CX drafted the work and the original paper. CX made substantial contributions to the conception. MD, XS, and BS provided resources. MQ, YT, GW, and YL revised the work and validated the contents. WY designed the work and was the project administration. BG provided knowledge of neurology to make the paper more professional. All authors read and approved the final manuscript.

Declaration of conflicting interests

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Ethical approval

Ethical approval of this case report was obtained from "the Naval Medical University Ethics Committee Review Board" (Approval No. 2021SL044). This research was conducted in regard to the Declaration of Helsinki, and the case report was documented following the Surgical CAse REport Statement. The informed consent was obtained from the patient.

Informed consent

Written informed consent was obtained from the patient for her anonymized information to be published in this article.

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