Cervical Fracture with Diffuse Idiopathic Skeletal Hyperostosis and Respiratory Insufficiency: A Case Report

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Diffuse idiopathic skeletal hyperostosis (DISH) reportedly increases the risk of fracture due to low-energy trauma^{1,2)}. Countries with an increasing aging population have an increased incidence of cervical trauma in elderly patients, mostly occurring in the setting of low-energy trauma^{3,4)}.

We report a rare case of cervical fracture with DISH and subsequent respiratory insufficiency managed by emergency anterior cervical surgery.

An 87-year-old man was brought in by an ambulance and transferred to our hospital 4 h after a fall on the floor. He lost consciousness for 2 h following the fall and woke up with muscle weakness. His past medical and surgical histories included hypertension, atrial arrhythmia, and cerebellar infarction with residual mild paralysis in his right upper and lower limbs. The patient was taking oral rivaroxaban. Plain radiography and computed tomography (CT) revealed ossification of the posterior longitudinal ligament at C3-5, fusion due to the ossification of the anterior longitudinal ligament (OALL) at C5-7, fusion due to DISH at C5-7 and T2-12 (Fig. 1), and a fracture involving the OALL at the C6/7 intervertebral disc level. Magnetic resonance imaging (MRI) slices showed a large retropharyngeal hematoma extending to the mediastinum. Moreover, cervical spinal canal stenosis and myelomalacia at the C3/4 and C4/5 levels were observed, which showed the spinal cord injury (Fig. 2). At that point, muscle strength was fully recovered, although he reported severe paresthesia in both hands.

Dyspnea and respiratory insufficiency occurred 6 h after the presentation. Tracheal intubation was performed, and the patient was ventilated. Arterial active bleeding was still observed after 10 coiling attempts by transcatheter angiography (Fig. 3). Therefore, we decided to perform emergency anterior cervical surgery (Fig. 4).

A large hematoma extending from the retropharyngeal re-



Figure 1. Plain computed tomography scan sagittal slice.

Discontinuity at the ossification of the anterior lingitudinal ligament at the C6/7 intervertebral disc level was detected (arrow).

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Figure 2. Plaain cervical magnetic resonance (MR) images. A: Sagittal T2-weighted image showing massive retropharyngeal hematoma and high-intensity change in the spinal cord at the C4/5 level. B: Sagittal T2-weighted fat-suppressed image showing high-intensity change in the C6/7 intervertebral disc. C: Axial T2-weighted image showing severe canal stenosis at the C4/5 intervertebral disc level and high signal change in the spinal cord.

gion to the mediastinum was removed, and arterial hemorrhage from the inferior thyroid artery was detected and ligated. Rupture of the OALL was observed and considered the cause of the arterial hemorrhage. Anterior cervical discectomy and fusion (ACDF) were performed under microscopic view. A 10 Fr Blake silicon drainage tube was placed in the retropharyngeal space and removed two days after surgery. The postoperative course was uneventful. As severe paresthesia in both hands remained, a second surgery was performed 23 days after the initial presentation. Decompression was performed at C3-6 and posterior fusion at C3-5 using lateral mass screws to prevent the future enlargement of the segmental OPLL, particularly at C4 and C5 levels because they were considered mobile segments. Twelve months postoperatively, the patient could walk with a cane for 200 m; no functional disturbances remained, and there was no implant failure/loosening.

There are several reports in the literature of arterial injury due to cervical spinal injury, which is a life-threatening condition (Table 1)^{5.6}. A recent systematic review showed that

the majority of the traumatic retropharyngeal hematoma with respiratory symptoms was in geriatric patients with falls or traffic accidents whose symptoms developed within 24 h of blunt trauma, and surgical treatment was performed in 23% of patients⁷. Enhanced CT, angiography, and arterial embolization are the most commonly used techniques to diagnose and manage retropharyngeal hematomas due to arterial injury^{7.9}.

To the best of our knowledge, this is the first documented case of a cervical fracture with DISH and subsequent respiratory insufficiency treated with emergency anterior cervical surgery due to arterial bleeding.

Previous studies have shown that cervical fractures in DISH usually occur at disc levels susceptible to low-energy trauma^{4,10)}. These characteristics were also found in this case.

There are two possibilities for the mechanism of damage to the artery: first, the edge of the fractured bone could have directly damaged the artery and second, sclerotic changes to the artery may have caused the rupture. Additionally, preexisting history of hypertension and the use of oral rivarox-



Figure 3. Angiography of left subclavian attery and left common carotid artery, and transcatheter arterial embolization of inferior thyroid artery. A: Before coiling and B: After coiling; both arrows show active bleeding. Residual arterial bleeding was suspected after coiling.

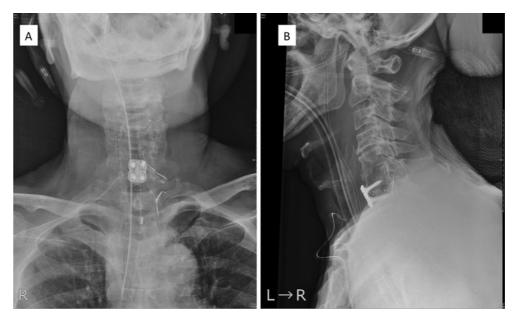


Figure 4. Postoperative X-ray images of C6/7 anterior discectomy and fusion. A: Anteroposterior X-ray and B: Lateral X-ray.

aban are potential risk factors for abnormal bleeding and difficulties in blood coagulation⁴.

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Ethical Approval: None. (As this is a case report without identifiers.)

Informed Consent: The authors certify that they obtained all appropriate patient consent forms. The patient has given his consent to include the images and other relevant clinical data in this report. The patient understands that his name and initials will not be published, and due efforts will be made to conceal his identity; he also understands that anonymity cannot be guaranteed.

Author	Journal	Year	No. of cases	Age	Sex	Fracture	Lev- el	Cause	Time from trauma	Risk factors	Procedure	Findings
Smith JP	J Trauma	1988	-	77	ц							
O'Donnell JJ	Eur	d 1997	1	19	M	C1/2 fracture dislocation	C1/2	Motorcycle accident				Difficulty in intubation
Sandooram D	J Laryngol Otol	2000	-		Μ					Anticoagulant therapy		
Kette F	Eur J Emerg Med 2000	d 2000	1		Μ			Minor frontal wound				
Dullivard C	Eur Argh Otorhinolaryngol	2005 I	0								Tracheostomy, per-oral drainage, and nasogastic tube feeding	
Anagnostrara A	A Emerg Radiol	2005	-	58	Μ	None		Motor vehicle accident		Methylprednisolone		
Freeman BJC		2005	-	31	Μ	Occipital condylar fractures		Motorcycle accident			Emergency endotracheal intubation	
Rizk NN	Laryngoscope	2010	1	55	ц	None		Uncontrolled hypertension and cervical spine arthritis			Transoral and transcervical incisions and drainage	
Wronka KS	Ann R Coll Surg 2011 Engl	3 2011	-			Odontoid fx	C2	Fall			Tracheal intubation	
Iizuka S	J Emerg Med	2012	1	30	ц	None		Motor vehicle collision	4 h		Angiography	Active bleeding from the right thyrocervical artery rupture
Kudo S	Am J Emerg Med 2017	d 2017	1	83	ц	C4/5 dislocation	C5	Motorcycle accident			Angiography and endovascular embolization	Active bleeding from the right vertebral artery rupture and hemorrhagic shock
Iida A	Int J Surg Case 2020 Rep	2020	-	79	Μ	None	З	Fall from height	3.5 h	Warfarin	Active bleeding from the right thyrocervical trunk of ascending cervical artery and embolization	
Yu S	Korean J Neurotrauma	2020	-	55	Μ	None	C6/7	Bicycle accident	4 h		Percutaneous aspiration of hematoma, urokinase administration, and coiling	Active bleeding from right superior thyroid artery posterior branch
Baek JH	Medicine	2020	-	49	Σ	None	C	Minor car accident	2 h		Observation	
Tanaka I	Cureus	2022	-	52	Σ	None	C	Fall while walking	3 days		Tracheostomy	
Lee J	Ther Clin Risk Manag	2019	62								Retropharyngeal space thickness at C2 is associated with difficult direct laryngoscopy	
Tsao YL	Emerg Med Int 2021	2021	68	13-94			-	Falls (54.4%) and traffic 95.2% accidents (35.3%) within 24 h	95.2% within 24 h		Conservative (63.2%), surgical (23.5%), transarterial embolization (8.8%), and died in 12 cases	
Shiba D	Eur J Trauma	2022	24	69 ;	,					27% Anticoagulant/	16/24 tracheal intubation and	

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