Epidural hematoma in patients with ankylosing spondylitis requiring surgical stabilization: a single-institution retrospective review with literature analysis

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OBJECTIVE Ankylosing spondylitis (AS) is a chronic inflammatory disease affecting the sacroiliac joints and axial spine that is closely linked with human leukocyte antigen–B27. There appears to be an increased frequency of associated epidural hematomas in spine fractures in patients with AS. The objective was to review the incidence within the literature and a single-institution experience of the occurrence of epidural hematoma in the context of patients with AS requiring spine surgery.

METHODS Deep 6 AI software was used to search the entire database of patients at a single level I trauma center (since the advent of the institution's modern electronic health record system) to look at all patients with AS who underwent spinal surgery and who had a diagnosis of epidural hematoma. Additionally, a systemic literature review was performed of all papers evaluating the incidence of epidural hematoma in patients with spine fractures.

RESULTS A single-institution, retrospective review of records from 2009 to 2020 yielded a total of 164 patients with AS who underwent spine surgery. Of those patients, 17 (10.4%) had epidural hematomas on imaging, with the majority requiring surgical decompression. These spine fractures occurred close to the cervicothoracic or thoracolumbar junction. The patients ranged in age from 51 to 88 years, and there were 14 males and 3 females in the cohort. Eight patients were administered an antiplatelet and/or anticoagulant agent, and the rest were not. All patients required surgical stabilization, with 64.7% of patients also requiring decompressive laminectomies for evacuation of the hematoma and spinal cord decompression. Only 1 death was reported in the series. There was a tendency toward neurological improvement after surgical intervention.

CONCLUSIONS AS has been a well-described pathologic process that leads to an increased risk of three-column injury in spine fracture, with an increased incidence of symptomatic epidural hematoma compared with patients without AS. Early recognition of this entity is important to ensure that appropriate surgical management includes addressing compression of the neural elements in addition to surgical stabilization.

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KEYWORDS ankylosing spondylitis; spine surgery; spine fusion; extradural hematoma; EDH; spinal epidural hematoma

ANKYLOSING spondylitis (AS) is a chronic inflammatory disease of the sacroiliac and axial joints with multiple extraspinal manifestations that is closely linked with the presence of human leukocyte antigen–B27 (HLA-B27).¹ Syndesmophyte formation with decreased motion and spinal deformity can lead to significant dis-

ability. While exercise and continued motion remain the mainstay of treatment, recent pharmacological advancements and the use of nonsteroidal antiinflammatory drugs (NSAIDs), antitumor necrosis factor agents, and infliximab have significantly improved functional outcomes.² More men than women are affected, and the incidence

ABBREVIATIONS AS = ankylosing spondylitis; EDH = epidural hematoma.

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ranges from 0.5 to 14 per 100,000 members of the population.³

Spine Fractures in AS

Patients with AS, as in other chronic inflammatory rheumatic diseases, are at higher risk of low bone mass, which predisposes them to fractures.⁴ A recent review identified that low-frequency impacts in patients with AS, such as ground-level falls or trivial traumas, can lead to a significant compromise of spinal stability requiring CT and MRI for evaluation and often necessitating surgical stabilization.⁵ Chronic inflammation, bone remodeling, and osseous resorption lead to osteoporosis and a more rigid spine that predisposes patients to increased fractures that can often be detected on CT; however, occult fractures can be missed and need to be evaluated on MRI.⁶

Spinal EDH and AS

One of the earliest descriptions of surgical intervention required in a patient with AS found to have an epidural hematoma (EDH), after a ground-level fall during a generalized tonic-clonic seizure, was in 1979.⁷ A recent radiographic study looking at patients with urgent and emergency MRI to evaluate spine trauma identified a rate of 68% of patients with AS who had EDHs.⁸ While the relationship between spinal EDHs in patients with AS and traumatic spine fractures has been described in previous publications,^{9–12} there has certainly been a paucity of literature describing this important relationship that can have dire neurological consequences if not recognized early.¹³ To further evaluate and describe the incidence of this relationship, we have reviewed our institutional experience and the published literature.

Methods

Retrospective Cohort Analysis

Using Deep 6 AI software, we performed an artificial intelligence–driven search (see below for detailed explanation) analyzing a complete database of patients at our level I trauma and comprehensive stroke medical center since the incorporation of Epic (Epic Systems Corp.) electronic health record software in 2009. Using search terms to find all patients diagnosed with AS, we found 10,000 patients, of whom 164 were verified to have spine fractures that were managed with surgical intervention. We then queried the records of those patients for a diagnosis of EDH and found the 17 patients included in our study; 10.4% of patients (17/164) with AS with spine fractures also had a spinal EDH. We then critically analyzed and performed retrospective chart reviews of all patients within this cohort.

Deep 6 AI Search Platform

According to the company and the explanation of their software, the Deep 6 AI search platform uses a variety of tools and techniques to perform its precision matching rather than relying on a single algorithm. First, clinical concepts are parsed from a wide range of clinical data sources that are both structured and unstructured. These

sources are updated nightly to offer the most recent results. Next, 100 different clinical ontologies are combined to accurately recognize clinical concepts from unstructured or semistructured data sources, such as progress or consultation notes. A set of annotators then parses all source data, and labels and stores clinical concepts in a graph database. This turns different clinical notes or fields into a multi-dimensional vector that can represent a patient and then become a node in the network. Other machine learning routines help disambiguate concepts, detect negations and hypotheticals, and reconcile different concepts and rank results based on a multifactor confidence score. The user can then write a query that searches against indexed patient graphs and retrieves results across millions of patients in a few seconds. The way the queries are constructed determines the precision and recall, and the Deep 6 AI platform favors recalls that allow for queries to return any matching patient according to the criteria of the original query, even if a single concept is selected, such as “ankylosing spondylitis” in our search. The patient evidence timeline makes it simple and quick to validate those matched patients by presenting the source data in a single place for easy review. Finally, validation then informs further filtering for higher-precision queries using additional filters (e.g., time and demographic) and Boolean functions.

Literature Review

We used the PubMed database to locate articles that identified patients with AS and EDH. We then used the keywords “ankylosing spondylitis” AND “epidural hematoma,” which yielded 55 results. Including only English-language papers, the results were narrowed to 49. We also excluded papers in which traumatic etiologies with spinal fractures were not the cause of the EDH. One paper was excluded because it was unclear whether the case reported¹⁴ was previously discussed in the case series by the same lead author years prior.¹⁵ Two separate authors (G.H. and S.A.U.) independently reviewed each of the papers, and a third author (T.T.K.) reviewed these articles to come to the consensus that 28 articles met the inclusion criteria of being clinical articles that reviewed demographics and outcomes in patients with AS who sustained spinal fractures and were found to have a spinal EDH.

Results

Patient Demographics and Results

Our retrospective review of patients with AS requiring spine surgery between 2009 and 2020 revealed 164 patients. Of these patients, 17 had an EDH and required surgical stabilization. There were 14 males and 3 females in our cohort with an age range from 51 to 88 years (average 69.7 years). The predisposing traumatic mechanisms included ground-level falls in 11 patients (Figs. 1 and 2), motor vehicle collision in 3 patients, fall from a height in 1 patient, and unknown etiology in 2 patients. Patient characteristics and demographics are listed in Table 1.

All patients in our cohort required surgical stabilization. Eleven patients (64.7%) required decompressive laminectomies with evacuation of an EDH, and 1 patient had extensive decompressive laminectomies from C7 to T11

FIG. 1. An 80-year-old female with AS sustained a ground-level fall and was found to have a C5–6 fracture dislocation with a C5–T1 EDH. The patient underwent a C5–7 posterior spinal fusion with laminectomies and evacuation of the EDH. Preoperative T2-weighted sagittal (A) and axial (B) cervical spine MR images demonstrating a C5–T1 EDH. Preoperative sagittal CT scans of the cervical spine (C and D) demonstrating a C5–6 fracture dislocation. Postoperative T2-weighted sagittal (E) and axial (F) cervical spine MR images demonstrating decompression and a C5–6 cord signal change. Postoperative axial (G) and sagittal (H) CT scans of the cervical spine, demonstrating wide laminectomies for decompression and C5–7 posterior spinal instrumentation.

at an outside institution, years prior to presentation, for a traumatic EDH (Fig. 3). Eight patients were administered antiplatelet and/or anticoagulant agents, with the majority being on a regimen of 81 mg of aspirin.

The mean blood loss during surgery was 585 ml (range 70–3000 ml), and the mean surgery duration was 279 minutes (range 119–502 minutes). In 47% of the surgeries performed, transfusion with one or more blood products or derivatives was used, including packed red blood

FIG. 2. A 54-year-old male with AS sustained a ground-level fall resulting in severe C3 spinal stenosis, a C6–7 fracture dislocation, and a C5–7 EDH. The patient underwent a C6–7 anterior cervical discectomy and fusion (ACDF), C3–T3 posterior spinal instrumented fusion, and C5–7 laminectomies with evacuation of the EDH. Preoperative T2-weighted sagittal (A) and axial (B) cervical spine MR images demonstrating large prevertebral swelling and a C6–7 fracture dislocation with associated EDH and severe stenosis. Preoperative sagittal (C) CT scan of the cervical spine demonstrating a C6–7 fracture dislocation. Postoperative cervical spine anteroposterior (D) and lateral (E) radiographs and a postoperative sagittal (F) CT scan of the cervical spine demonstrating posterior hardware from C3 to T3 and a C6–7 ACDF (G). Postoperative axial CT scan of the cervical spine (H) demonstrating wide laminectomies for decompression at C6.

cells, fresh frozen plasma, and platelets. The total hospital length of stay was a mean of 20.5 days (range 2–144 days), and if the longest hospital stay outlier was excluded, the mean hospital length of stay was 12.8 days.

We encountered a series of transient complications in

TABLE 1. Patient demographics and results

Pt Age (yrs)	Sex	Op Performed	Anticoag/-platelet Agent	Fx Level & Description	Mechanism of Injury	Yr	EBL (ml)	Op Duration (mins)	Complications	BT Required	LOS (days)
80	F	C5–7 lami/PSF/evacuation of EDH	ASA 81 mg	C5–6 Fx disloc w/ C5–T1 EDH	GLF	2008	70	150	DVT requiring IVC filter & anticoag	No	13
84*	F	T6–12 posterolat instrumented fusion	None	T9 Fx disloc w/ EDH	GLF	2009	100	119	Postop wound dehiscence & infection requiring superior hardware removal & wound revision x3	No	32
53	M	C5–6 ACDF evacuation of hematoma, C1–6 decompressive lami for evacuation of EDH & occipital–C7 PSF	None	C5/6 Fx disloc w/ C1–6 EDH	GLF	2009	3000	400	DVT	5 U FFP, 6 U PRBC, 3 U platelets	26
45	M	C4–T3 PSF & C7 & T1 decompressive lami	None	C2 type III odontoid Fx & C7–T1 discitis-osteomyelitis; C7–T1 subluxation & EDH	Unknown; presented w/ worsening back pain & difficulty walking	2010	100	185	C. diff colitis	3 U PRBC, 1 U FFP, 1 U platelets	24
63	M	T10–L3 PSF; T12–L1 decompressive lami & open reduction to T12–L1 Fx disloc	None	T12–L1 hyperextension-distract w/ Fx & EDH	GLF	2013	500	502	None	No	7
72	F	C3–T3 lami for spinal cord decompression w/ evacuation of EDH & C4–T2 PSF	ASA 325 mg	C6/7 Fx disloc & C5–T4 EDH & C3 spinal cord compression due to severe stenosis	GLF	2013	200	229	None	No	2
83	M	T11 laminotomy/T12 lami w/ T9–L2 PSF for lt T11–12 facet Fx	ASA 81 mg	T11–12 Fx disloc	Unknown; presented w/ worsening back pain & difficulty walking	2014	700	317	Postop wound infection	2 U PRBC, 2 U FFP	5
63	M	T8–L1 PSF	None	T9 teardrop & T11 chance Fx	MVC (bus vs tractor trailer)	2016	75	149	None	No	5
84	M	Initially managed in brace; underwent T8–12 (skipped T10) bilat percutaneous PSF w/ close reduction of Fx	ASA 81 mg	T9 & T10 Fxs (3-column at T10)	Head-on MVC w/ self-extrication	2017	100	226	None	No	3
64	M	L4–pelvis PSF	ASA 81 mg	Comminuted sacral Fx causing lumbopelvic dissociation	9-ft fall from ladder	2017	100	229	None	No	8
51	M	T7–12 PSF & T9 & T10 decompressive lami	None	T10–11 Fx distract disloc	GLF	2018	300	154	ARDS/bacteremia	No	144
54	M	C6–7 ACDF w/ partial C7 corpectomy & placement of PEEK graft w/ C3–T3 posterolat instrumented fusion & C5–7 lami w/ evacuation of EDH at C6–7	None	C6–7 Fx disloc	GLF	2018	1550	488	None	3 U PRBC	25

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TABLE 1. Patient demographics and results

Pt	Age (yrs)	Sex	Op Performed	Anticoag/-platelet Agent	Fx Level & Description	Mechanism of Injury	Yr	EBL (ml)	Op Duration (mins)	Complications	BT Required	LOS (days)
88	M		T5–11 posterolat instrumented fusion	ASA 81 mg	T8 extension; Fx w/ EDH	GLF	2020	350	315	Postop hypotension requiring several days of pressors in ICU	1 U PRBC	18
81	M		C2–T3 PSF w/ revision of previous C2–6 PSF; C5, C6, C7, T1 decompressive lami w/ evacuation of EDH at C6 & C7	None	C6–7 3-column Fx w/ C6–7 EDH	GLF	2020	1400	496	None	2 U PRBC	6
82	M		T9–S1 revision PSF (previous L3–S1 fusion); T11 & T12 complete lami for evacuation of EDH	None	T12 hyperextension; Fx disloc	GLF	2020	200	247	Worsened respiratory secretions & difficulty breathing led to cardiac arrest; pt died	1 U PRBC	19
82	M		T10–L3 PSF, T11, T12, L1 & L2 decompressive lami w/ evacuation of EDH	Warfarin	T12 3-column Fx	GLF	2020	800	331	AFIB w/ RVR requiring amiodarone drops	2 U FFP	5
56	M		T6–12 posterolat instrumented fusion	ASA 81 mg & Eliquis 5 mg 2x/day	T8–9 Fx distract	MVC	2020	400	201	AKI, resolved w/ intravenous hydration	None	6

ACDF = anterior cervical discectomy and fusion; AFIB w/ RVR = atrial fibrillation with rapid ventricular response; AKI = acute kidney injury; anticoag = anticoagulation; ARDS = acute respiratory distress syndrome; ASA = aspirin; BT = blood transfusion; C, diff = *Clostridium difficile*; disloc = dislocation; distract = distraction; DVT = deep vein thrombosis; FFP = fresh frozen plasma; Fx = fracture; GLF = ground-level fall; IVC = inferior vena cava; lami = laminectomy; LOS = length of stay; MVC = motor vehicle collision; PEEK = polyetheretherketone; PRBC = packed red blood cell; PSF = posterior spinal fusion; pt = patient.

* Baseline neurological injury from an EDH causing paraparesis from a fall in a city bus in September 2008 after C7–T11 decompressive laminectomies.

FIG. 3. An 84-year-old female sustained a ground-level fall and presented with T9 fracture dislocation with an associated EDH. The patient underwent T6–12 posterior spinal instrumented fusion. She had previous neurological deficit from a fall on a city bus 1 year prior to presentation and underwent C7–T11 decompressive laminectomies with evacuation of an extensive EDH at that time. Preoperative sagittal (A) and axial (C) CT scans and T2-weighted sagittal (B) and axial (D) MR images of the cervicothoracic spine demonstrating extensive C7–T11 laminectomies and wide decompression with a T9 fracture dislocation and EDH. Postoperative anteroposterior (E) and lateral (F) radiographs demonstrating T6–12 posterior spinal instrumented fusion.

our patients, with only 1 death during hospitalization. Two patients developed deep venous thrombosis that required anticoagulation. The patient with the longest hospital length of stay developed acute respiratory distress syndrome and bacteremia, while the single death in our series occurred later in their hospitalization when the patient developed respiratory failure and subsequent cardiac arrest. Two patients developed postoperative wound infections requiring revision and washout. One patient developed acute

kidney injury that resolved with intravenous hydration; and 2 patients required hemodynamic monitoring in the ICU, 1 for hypotension requiring pressors and 1 for amiodarone infusion to treat atrial fibrillation with rapid ventricular response. Finally, 1 patient developed *Clostridium difficile* colitis during hospitalization. In total, 10 patients (58.8%) had an in-hospital complication. There were no patients who had worsening neurological dysfunction, and there was a tendency toward neurological improvement prior to discharge (Table 2).

Literature Review Results and Findings

Throughout the review of the literature, various mechanisms of trauma were noted, and oftentimes progressive neurological deficit was noted to be associated with the discovery of an EDH concomitant with the spinal fracture. The results of our literature review are summarized in Table 3.^{7,8,10,12,15–38}

Discussion

Mechanism of Injury

Recorded mechanisms of injury within our cohort and in the literature review yielded a broad range of mechanisms but were often low-force impacts such as a ground-level fall. The earliest reported case in the English-language literature was in a patient who sustained generalized tonic-clonic seizures and tetraparesis who was found to have an extensive EDH from C6 to T8.⁷ Eleven of our patients (64.7%) sustained ground-level falls which caused traumatic fractures and spinal EDH. High-impact mechanisms were also noted, including motor vehicle collisions and fall from a height. These mechanisms were again noted in our literature review.^{19,31}

Intervention, Diagnosis, and Outcome

In our series, we reported that 64.7%, or close to two-thirds, of patients required decompressive laminectomies to evacuate the EDH. A recent series of patients with AS demonstrated a 29.3% incidence of spinal EDH, while the incidence was found to be 19.9% in patients with and without spinal cord injury in another series.³⁹ Other studies also reported a high incidence of EDH after trauma in patients with AS.^{38,40} Most of our patients were diagnosed via MRI or on surgical exploration when neurological deficits were present and we were unable to perform MRI of the area of interest within the spinal axis. Two patients presented without gross neurological deficit and subsequently worsened, while 1 patient had a neurological deficit that improved with surgical immobilization and was found to have an extensive EDH that was evacuated.

Several reports in the literature demonstrated varied presentations, time to diagnosis, and outcomes. A recently published case report demonstrated the first instance of robotic guidance for placement of pedicle screws in addition to decompressive laminectomies³⁷ in a patient with AS and a traumatic spine fracture. In a series of 5 patients, Broom and Raycroft¹⁹ described a 74-year-old patient with AS with a C5–6 fracture who developed a subacute C6 complete spinal cord injury, even with halo immobilization, and was found to have an EDH that was decom-

TABLE 2. Neurological function before and after surgical intervention in patients with AS and EDH

Pt Age (yrs)	Sex	Presenting Neurological Exam	Neurological Exam Findings at Discharge
80	F	Initially neuro intact; progressed to C4 complete SCI	C7 ASIA grade C SCI; some movement in lt UE & intrinsic w/ some leg movement; weak triceps on rt, otherwise no movement distally
84*	F	1–2/5 in proximal LEs & 3/5 distally (baseline)	Unchanged
53	M	UEs 2–3/5 & LEs 0/5	Unchanged
45	M	4/5 lt LE & 4+/5 in lt UE	5/5 in all extremities w/ 4+/5 in bilat iliopsoas
63	M	Bilat quadriceps & iliopsoas weakness (4/5) w/ L1 & L2 numbness	Improved strength & sensation; ambulates w/ walker
72	F	Tetraplegia w/ sensation intact; improved to full strength w/ rt UE 0–2/5 after collar placed	Full strength, except rt hand 2/5
83	M	ASIA grade C SCI; 0/5 in EHL/DF/gastrocnemius; 2/5 quadriceps	ASIA grade D SCI; 3/5 DF/PF; 4/5 quadriceps
63	M	Neuro intact	Neuro intact
84	M	Neuro intact	Intact
64	M	Neuro intact	Neuro intact
51	M	T10 ASIA grade A (complete) paraplegia	T10 ASIA grade A
54	M	Neuro intact	Neuro intact
88	M	Neuro intact	Neuro intact
81	M	Lt UE WE/HG/intrinsic 4/5	Lt UE improved to 4+/5
82	M	Neuro intact	Neuro intact
82	M	Initially neuro intact; progressed to 1/5 in LEs w/ L2 sensory level	Improved to 4–5/5 in LE
56	M	Neuro intact	Neuro intact

ASIA = American Spine Injury Association impairment scale; DF = dorsiflexion (ankle); EHL = extensor hallucis longus; HG = hand grip; LE = lower extremity; LLE = left lower extremity; neuro = neurologically; PF = plantar flexion; SCI = spinal cord injury; UE = upper extremity; WE = wrist extension.

* Baseline neurological injury from an EDH causing paraparesis from a fall in a city bus in September 2008 after C7–T11 decompressive laminectomies.

pressed and evacuated, but the patient remained without neurological improvement. In another study, a 91-year-old patient with AS had a delayed diagnosis of a thoracic spine fracture that was initially managed conservatively, but the patient's condition progressed to paraplegia. He was diagnosed with an EDH after neurological deficit and underwent posterior decompression and fusion with eventual neurological improvement.³¹ De Peretti et al.²⁰ described a case of a male patient who was involved in a motor-bike collision with progressive paraplegia and eventually received a diagnosis of an EDH with subsequent evacuation; the patient improved to full neurological recovery. Fitt et al.²⁴ described a 68-year-old male with 24 hours of progressive paraplegia and a minor fall 1 month prior to presentation. The patient was found to have an EDH that was evacuated but with incomplete neurological recovery. Garza-Mercado²³ described a 32-year-old male who fell after being pushed during an argument and experienced progressive paraplegia and bowel and bladder dysfunction; the patient ultimately underwent decompressive laminectomies and evacuation of an EDH, resulting in a delayed, complete neurological recovery. Even in minor trauma with rapid diagnosis and intervention, outcomes can be catastrophic, as seen in a 63-year-old male who presented after a minor fall and was found to have a C3–T2 EDH that was decompressed; despite surgical intervention, the patient subsequently died.³⁰

Most of the patients in our cohort had gradual neurological improvement or neurological preservation after surgical intervention, and no patients had a decline after

surgical intervention. However, of the 5 patients in our cohort who had the worst neurological dysfunction prior to surgery (i.e., tetraplegia or paraplegia), only 1 had a significant neurological improvement postoperatively, indicating that early recognition of a compressive hematoma and decompression is essential to prevention of devastating outcomes.

Anticoagulation and Hematoma

In our series, 8 patients (47%) were receiving antiplatelet and/or anticoagulant therapy. This was also noted in some patients in our literature review. Elgafy et al.²⁹ reported on a 69-year-old male taking the anticoagulant Coumadin who had fallen from a 9-foot height, developed subsequent tetraparesis, and was found to have an EDH and an occult fracture. The patient was treated with decompression without fusion and continued cervical immobilization with improvement in neurological function.

Nontraumatic Etiologies

There were patients who did not meet our inclusion criteria due to the lack of spinal fractures. We found a series of cases in the literature in which patients with AS underwent epidural anesthesia and sustained a postoperative EDH requiring urgent decompression. One patient was a 65-year-old male with AS who underwent a laparotomy for abdominal wall reconstruction and had epidural anesthesia with a catheter inserted at the T8–9 level. He was on a preoperative regimen of aspirin for prophylaxis and tinzaparin

TABLE 3. Literature review of cases and case series of patients with AS with spinal fracture and EDH

Authors & Year	Pts*	Symptoms, Fx Level	Hemorrhage Level	Surgical Intervention	Neurological Outcome	Outcome	Complications/Notes
Grisolia et al., 1967 ¹⁶	50, M	Complete motor paralysis, C7 transverse Fx	C4–7	C3–C7 lami & PSF	Partial return of UE strength	Partial motor recovery in UE; sustained paralysis in LE	
Bohlman, 1979 ¹⁷	60, M	Partial sensory loss w/ incomplete motor paralysis	C7–T1	Surgical decompression	Pt died 18 days postop	Pt died	
	NA	Partial sensory loss w/ partial motor paralysis	C7–T1	Surgical decompression	Residual motor weakness on discharge	Recovery w/ no lasting defect	
	NA	Complete sensory loss w/ complete motor paralysis	C3 or C4	Surgical decompression	Pt died 3 days postop	Pt died	
	NA	Partial sensory loss w/ complete motor paralysis	C3 or C4		Pt died 1 day after accident	Pt died	
Agnetti et al., 1979 ⁷	54, M	Severe sensory loss (below C6); complete flaccid tetraparesis; urinary/bowel incontinence	C6–T8	C6–T8 lami & PSF	Severe sensory loss (below C6); complete flaccid tetraparesis	Pt died, anoscopic 1 mo after admission w/ progressive delay	2 chronologically different hemorrhages found during decompression, based on clot aspects
Foo & Rossier, 1982 ¹⁵	72, M	Partial sensory loss (below C4); partial motor paralysis developing to complete motor paralysis (10 days postinjury); C5–6 anterior body Fx	C6	C5–7 lami & PSF	Partial sensory loss (below C4); partial motor paralysis (flaccid below C6); normal DTRs & Babinski bilat	No neurological recovery; pt died 4.5 mos after injury	Concurrent tuberculosis treatment
Hissa et al., 1986 ¹⁸	53, M	Partial sensory loss in BLE; motor weakness in BLE	T2–3	T2–7 lami & PSF	Residual sensory & motor deficits; spastic gait; atonic bladder	Recovery w/ no long-lasting defect	Initially started on high-dose ICS, symptoms improved then declined after taper
Broom & Raycroft, 1988 ¹⁹	74, M	Neck pain; progressive motor weakness in all extremities	C6	Surgical decompression	Quadriplegia at C6 level	Discharged to custodial care 5 mos after injury	Diagnosis delayed until 12 days after initial injury
de Peretti et al., 1993 ²⁰	41, M	Complete sensory loss w/ complete motor paralysis; urinary/bowel incontinence; priapism; T7 vertebral body Fx	T5–8	T6–8 lami, T4–10 PSF	Return to baseline	Recovery w/ no lasting defect	
Ticó et al., 1998 ²¹	71, M	Complete sensory loss w/ complete motor paralysis below T2	T5–10	NA	Partial sensory loss (below T5) w/ partial motor paralysis	NA	Pt died in chronic phase
	56, M	Complete sensory loss w/ complete motor paralysis below C6; C6–7 Fx disloc	T1–12	NA	Complete sensory loss (below C6) w/ complete motor paralysis	NA	Pt died in acute phase
	65, M	Partial sensory loss w/ partial motor weakness below T9	C2–T3	NA	Partial sensory loss (below T9) w/ residual motor weakness	Recovery w/ no lasting defect	

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TABLE 3. Literature review of cases and case series of patients with AS with spinal fracture and EDH

Authors & Year	Pts*	Symptoms, Fx Level	Hemorrhage Level	Surgical Intervention	Neurological Outcome	Outcome	Complications/Notes
Wu & Lee, 1998 ²²	44, M	Partial sensory loss (below C5); partial BLE motor weakness progressing to BUE & BLE paralysis; C5–6 transverse Fx	C4–T1	C4–T1 lami	Rapid progressive improvement of sensory & motor function; residual sphincter deficit	Recovery w/ no lasting defect	
Garza-Mercado, 1989 ²³	32, M	Partial sensory loss (below rt T7 & lt T10) w/ complete motor paralysis below C6; loss of anal tone; C7 Fx	C5–T1	C4–T1 lami	Ambulates w/o walker; sphincter tone regained	Recovery w/ no lasting defect	
Fitt et al., 1992 ²⁴	68, M	Partial sensory loss (below T8) w/ partial motor paralysis progressing to complete BLE motor paralysis	T7–8	Surgical decompression	Complete recovery	Recovery w/ no lasting defect	
Rodi et al., 2003 ²⁵	42, M	Neurologically intact; L2 Fx	T5–L1	Surgical decompression	Postop able to move legs; no sensory deficit	Recovery w/ no lasting defect	Hematoma complication from initial anterior stabilization of L2 Fx
Nakstad et al., 2004 ²⁶	53, M	Transvertebral C5–6 Fx	C4–7	Surgical decompression	Complete BLE paralysis	Paresis	
	53, M	Transvertebral C6 Fx	C5–7	Surgical decompression	Return to baseline	Recovery w/ no lasting defect	Initially externally immobilized
	73, M	C7 Fx	C3–T5	Surgical decompression & fusion	Partial BLE paralysis	Paralytic	
	66, M	C7 Fx	C7–T2	Surgical decompression	Return to baseline	Recovery w/ no lasting defect	Initially externally immobilized
Pedrosa et al., 2002 ²⁷	86, F	Central cord syndrome; 3-column C6 injury	C5–T1	NA	NA	Died of cardiac/respiratory complications	
	67, F	Central cord syndrome; urinary incontinence; 3-column C6 injury	C6–T1	NA	NA	Residual paresthesia of fingers in both hands	
	89, M	Complete motor paralysis; 3-column C4 injury	C1–6	NA	NA	Died of cardiac/respiratory complications	
Vives et al., 2008 ²⁸	59, M	Complete sensory loss w/ complete motor paralysis	C3–7	C3–7 lami & PSF	Partial BLE paralysis	Recovery w/ no lasting defect	
Elgafy et al., 2011 ²⁹	69, M	Partial sensory loss (BUE) w/ partial motor weakness (BUE)	C5–T3	C5–T3 lami; C4–T3 PSF	Partial BUE sensory loss; partial BUE paralysis	Complete resolution of muscle weakness; residual numbness in C6 dermatome	Pt on Coumadin for AFIB
Kim & Kim, 2011 ³⁰	63, M	Partial motor paralysis (BLE); C7–T1 Fx	C3–T2	Surgical decompression	Pt died	Pt died	
Aoki et al., 2013 ³¹	91, M	Partial sensory loss (BLE) w/ partial motor weakness (BLE) progressing to complete BLE paralysis	T7–9	T7–9 lami; T7–12 PSF	Improved BLE strength	4/5 muscle strength in BLE standing w/ support; unable to walk	Initial imaging failed to recognize Fx; treated conservatively w/ degrading neurological exam for 74 days postfall

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TABLE 3. Literature review of cases and case series of patients with AS with spinal fracture and EDH

Authors & Year	Pts*	Symptoms, Fx Level	Hemorrhage Level	Surgical Intervention	Neurological Outcome	Outcome	Complications/Notes
Wong & Yu, 2015 ¹²	51, M	Partial sensory loss (lt C5, rt C2) w/ complete motor paralysis; C4 chance Fx	C2–T9	C2–T9 lami; lateral mass plates C3–5	Return to baseline	Recovery w/ no lasting defect	Wound infection (POD 7) causing C4 corpectomy w/ C3–5 fixation & fusion
Pitta et al., 2017 ³²	68, M	Partial sensory loss (BLE) w/ partial motor weakness (BLE); L4–5 chance Fx	L2–S1	L2–S1 lami; L2–pelvis PSF	Improved strength w/ residual weakness in bilat TA	Recovery w/ residual weakness in L4 distribution	L2–3 loss of screw fixation; required additional T10–pelvis PSF
Ramirez Villaescusa et al., 2017 ¹⁰	79, F	Partial sensory loss (BLE) w/ partial motor weakness (BLE); bowel incontinence; T3–4 Fx	C7–L1	T4–7 lt partial lami; T3–9 PSF	NA	Recovery w/ no lasting defect	
Scala et al., 2019 ³³	68, M	Neurologically intact; T12 vertebral body Fx	T11–12	T11–12 lami; T10–11, L2–2 PSF	NA	NA	Developed spinal arachnoiditis ossificans 4 yrs postfixation; needed additional debridement
Tamburrelli et al., 2018 ³⁴	59, M	Displaced L5 Fx	L2–5	Surgical decompression	NA	NA	
Tavolaro et al., 2019 ³⁵	69, M	Partial sensory loss w/ partial motor weakness; C7 displaced Fx	C5–T4	C2–T5 lami; C4–T3 PSF	NA	NA	
Vazan et al., 2019 ³⁶	12 pts w/ associated sEDH	Varied neurological deficits on presentation; 9 w/ type C Fx & 3 w/ type B Fx	Varied	Surgical decompression & fusion	Varied	Varied	
Al Saiegh et al., 2020 ³⁷	64, M	T8–9 distract Fx	T7–11	T7–11 lami & PSF	NA	NA	Fixation achieved w/ aid of robotic surgery
Shah et al., 2021 ³⁸	53, M	NA	T5–6	T5 decompression & T3–7 posterior fusion	NA	NA	
	62, M	NA	C5	C3–T1 PSF	NA	NA	
	82, M	NA	T8–9	T5–11 PSF	NA	NA	
Vierunen et al., 2021 ¹⁸	19 pts w/ associated sEDH	Varied	Varied	Varied	Varied on cases/speed of treatment	Varied on cases/speed of treatment	

BLE = bilateral lower extremity; BUE = bilateral upper extremity; DTR = deep tendon reflex; ICS = corticosteroids; NA = not available; POD = postoperative day; sEDH = spinal EDH; TA = tibialis anterior.

* Data for patients reported on singly are presented as age in years and sex.

for a previous pulmonary embolism, both of which were discontinued 7 days and 24 hours preoperatively, respectively. The patient woke from surgery with dense hemiparesis, and MRI revealed an EDH from T5 to T11 for which the patient underwent decompressive laminectomies and evacuation. He had minimal neurological recovery with sustained paraplegia after intervention.⁴¹ Another case reported in the literature is that of a patient with AS who had complete paraplegia 6 days after attempted spinal anesthesia for a total hip arthroplasty that required decompressive laminectomies for EDH, which resulted in eventual motor recovery.⁴² In a large review of 51 patients who developed spinal EDHs after spinal anesthesia, 5 patients had AS.⁴³

Spinal Subdural Hematoma

Although our literature review and cohort focused on EDHs, another closely related entity is spinal subdural hematoma, which can also cause catastrophic neurological injury when there is a delayed diagnosis or without appropriate intervention. A recent study looking at emergency and urgent spinal MRI identified a rate of 18% of subdural hematoma in traumatic spine fractures.⁸ Esfahani and colleagues⁴⁴ described a 60-year-old male on a regimen of Coumadin who was getting out of bed when he developed back pain and progressive lower-extremity motor and sensory deficits that required surgical evacuation which eventually resulted in complete neurological recovery. Another patient with AS who presented with a 1-week history of an inability to walk was diagnosed with a spinal subdural hematoma and underwent decompression and, although initially he was able to move his toes, his condition progressed to complete paraplegia.⁴⁵

Study Limitations

Our intention in this critical literature appraisal and single-institution retrospective review was to emphasize early recognition and intervention for an underappreciated pathology in patients with AS with spinal fractures: EDH that could cause neurological decline or catastrophic neurological injury. Given that this retrospective review constitutes class III evidence (the lowest class of clinical evidence), as are all the other papers cited herein, caution must be taken to ensure that the data reported in our series could underestimate or overestimate the actual incidence of this pathological entity in this population of patients. Further population-wide studies and reviews could help better approximate the true incidence within the literature. However, class I evidence will likely be unavailable due to the ethical considerations in a study that would prevent intervention or investigation of an underlying and potentially devastating pathologic process.

Conclusions

AS is a relatively rare spinal disorder that leads to a more rigid and fragile spine and predisposes patients to a greater risk of unstable spine fractures that may be occult. Spinal EDHs also have a higher prevalence in this population, and a missed or delayed diagnosis could lead to catastrophic neurological outcomes. Therefore, we present our institutional experience and a literature review to raise

awareness and to encourage early MRI or CT myelography studies, even with a low index of suspicion for these lesions. When neurological deficit is present, urgent surgical exploration with decompression is warranted. Early recognition of this relationship is important to ensure that appropriate surgical management includes addressing compression of the neural elements in addition to surgical stabilization.

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Conception and design: Johnson, Hanna. Acquisition of data: Johnson, Hanna, Uddin, Kim. Analysis and interpretation of data: Johnson, Hanna, Uddin, Drazin, Kim. Drafting the article: Johnson, Hanna, Uddin, Drazin, Kim. Critically revising the article: Johnson, Hanna, Uddin, Trontis, Drazin, Kim. Reviewed submitted version of manuscript: all authors. Statistical analysis: Johnson, Hanna, Ross. Administrative/technical/material support: Johnson, Hanna, Ross. Study supervision: Johnson, Hanna, Ross.

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