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Iatrogenic thoracic outlet syndrome caused by revision surgery for multiple subacute fixation failures of a clavicle fracture: A case report

John G. Skedros, MD[a,b,*], Bryce B. Hill, BS[a], Todd C. Pitts, BS[a]

[a]*Utah Bone and Joint Center, Salt Lake City, UT*
[b]*University of Utah Department of Orthopaedic Surgery, Salt Lake City, UT*

During revision surgical fixation of clavicle fractures and fracture nonunions, compression of the prominent neurovascular structures of the thoracic outlet can be an important potential complication. In this report, we describe the case of a manual laborer who had iatrogenic compression of the thoracic outlet of his dominant arm by fracture callus during revision surgery after multiple subacute fixation failures. The surgeon initially failed to recognize that the anatomic alignment of the clavicle fracture during osteosynthesis had resulted in thoracic outlet syndrome (TOS), as definitively established by venogram 14 days postoperatively. This case illustrates how the constellation of signs and symptoms can be misleading when this problem occurs in a subacute setting.

A number case reports have described iatrogenic TOS in the setting of a midshaft clavicle fracture associated with nonunions, malunions, and delayed diagnosis. This report is significant because it addresses a case of iatrogenic TOS accompanied by (1) multiple subacute failures of surgical fixation before final/definitive fixation, all within 60 days of the fracture, (2) description of the patient's clinical findings and diagnostic studies that ultimately led to a venogram and revision surgery, and (3) quantification of patient function and satisfaction for nearly 5 years to definitively establish whether decompression surgery for TOS would be required.

Case report

On April 7, 2004, a healthy 38-year-old, right hand-dominant man sustained closed fractures of the right midshaft clavicle and second rib after falling off an all-terrain recreational vehicle. He worked as a part-time electrician and a manual laborer on a cattle ranch. Eight days later, the treating general-practice orthopedic surgeon performed open reduction with internal fixation using a reconstruction plate and screws (5 unicortical and 1 bicortical). Noncompliance with lifting restrictions caused failure by bending of the plate and pullout of the medial screws at 4 weeks after the operation (Figure 1, *A*). Revision fixation was performed by the same surgeon on June 3, 2004, 6 weeks after the index operation, using a reconstruction plate with 6 bicortical screws and autogenous bone graft. Three weeks later, the fixation failed when the patient had been lifting (Figure 1, *B*).

Our first examination on June 29, 2004, which was 52 days after initial fracture and 26 days after the revision surgery, revealed slightly reduced manual grip strength and finger abduction (4+/5). A mild positive Tinel's sign was present at the right cubital tunnel, but there were no sensory deficits and no obvious evidence of proximal venous obstruction.

Revision surgery (the third operation) was performed on July 2, 2004, 29 days after the previous surgical procedure and 55 days after the initial fracture. All hardware was removed and fibrous tissue was excised from the fracture site. Iliac cortical-cancellous grafts were placed across the fracture site. A 10-hole reconstruction plate was applied to

*Reprint requests: John G. Skedros, MD, Utah Bone & Joint Center, 5323 S Woodrow St, Ste. 202, Salt Lake City, UT 84107.
E-mail address: jskedros@utahboneandjoint.com (J.G. Skedros).

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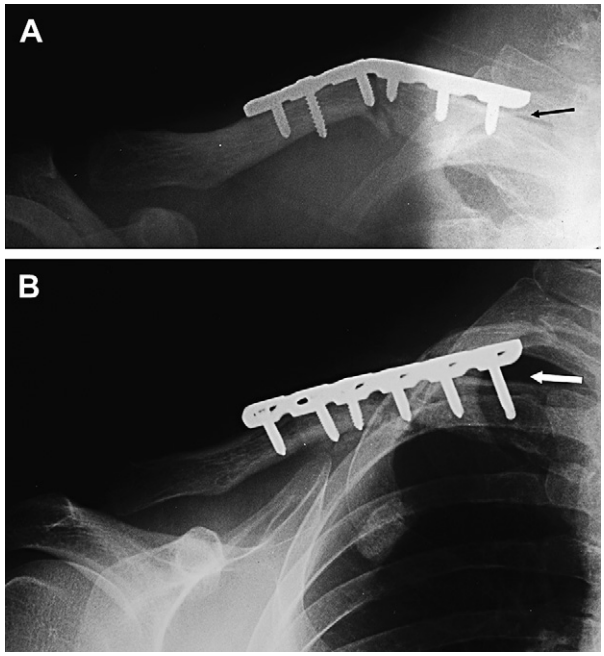

Figure 1 **(A)** Radiograph shows failure of the first fixation surgery by bending of the plate and pullout of the medial two screws *(arrow)*. **(B)** Radiograph shows failure of the second fixation surgery by pullout of the medial screws *(arrow)*.

the superior aspect of the clavicle and 2 stacked, 6-hole 1/3 tubular metal plates were placed on the anterior surface (Figure 2).

Excessive bleeding (total drain output, 250 mL) occurred during the first 12 hours postoperatively. Unexpected neurologic symptoms were also recognized the next morning, including mild numbness in the ulnar nerve distribution, mild swelling of the right forearm and hand, and mild weakness of right hand grip. During the ensuing 3 hours (12 to 15 hours postoperatively), swelling and the numbness in the ulnar nerve distribution worsened. Manual muscle testing showed reduced strength (4/5) in finger abduction and grip.

The surgeon initially speculated that a hematoma might have caused compression, which seemed to be supported by

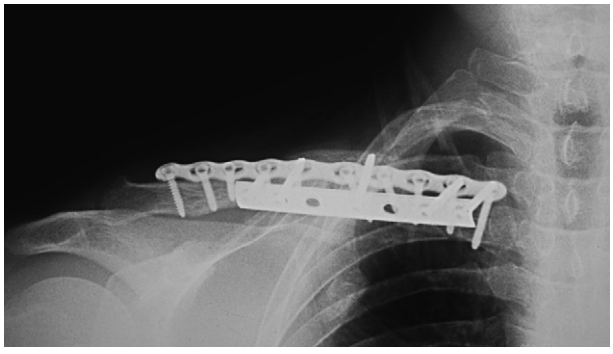

Figure 2 Radiograph of the third surgical fixation shows a 10-hole plate placed on the superior aspect of the clavicle, and 2, stacked, 6-hole plates placed on the anterior aspect of the clavicle. Callus was not obvious on these radiographs.

the large amount of drainage. Urgent workup included Doppler ultrasound imaging that showed no evidence of deep venous thrombosis; however, the study was limited because the metal plates obscured the subclavian vein. A computed tomography angiogram was negative for pulmonary embolus. At 24 hours postoperatively, the patient was taken to the operating room with a vascular surgeon present for consultation. Pulsatile bleeding was observed from a small artery and the associated hematoma was about 40 mL. The surgeons speculated that the hematoma had worsened the preoperative neuritis. Their consensus opinion was to obtain a venogram the next day if symptoms worsened.

Although improvement continued during the next 5 days, by the sixth day, there was worsening weakness, numbness, and swelling in his right hand and forearm. During the following week, the patient also reported intermittent mild cyanosis and coolness of his right hand. A venogram 14 days after our revision surgery showed high-grade stenosis of the subclavian vein (Figure 3, *A* and *B*).

An additional revision surgery was performed by JGS 16 days after our revision surgery. Intraoperative venograms

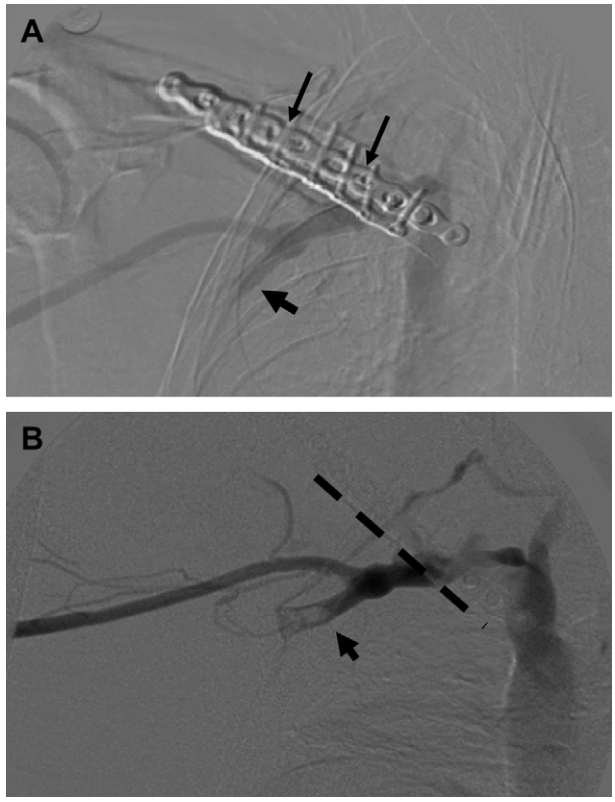

Figure 3 **(A)** The image contrast in this intraoperative venogram has been adjusted to enhance visualization of the stenosed subclavian vein *(large arrow)* and the patent cephalic vein *(at left)*. The metal plates are discernible *(2 smaller arrows)*. **(B)** A subtraction image with increased enhancement of the vessels shows the subclavian stenosis *(large arrow)*. The metal plates are not discernible in this image. The edge of the anterior plate is indicated with the *dashed line*.

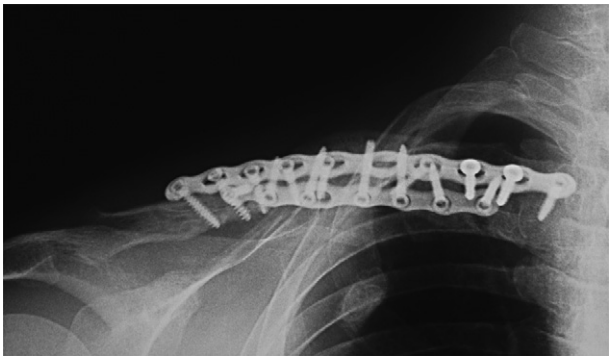

Figure 4 Radiographs of the final fixation construct during the fourth surgery. The clavicle was bowed upward to decompress the thoracic outlet. The resulting 2.5-cm gap in the clavicle was spanned with intercalary and on-lay iliac crest bone grafts.

showed spontaneous resolution of the stenosis after removal of the hardware and bone grafts. The subclavian vein also remained fully patent when revision fixation was completed with a new plate that was bowed upward, gapping open the fracture site 2.5 cm. Intercalary and on-lay bone grafts were applied across the gap (Figure 4).

Postoperative follow-up from final surgery

Follow-up data are reported for 4, 7, 15, 24, 36, and 39 months after the final revision fixation surgery. Information for 48- and 54-months (4.5 years) was obtained by telephone or written surveys. Table I lists various outcome measurements: (1) grip strength (Baseline, Hydraulic Hand Dynamometer, FEI, Irvington, NY, (2) pain on a 10-cm visual analog scale, (3) American Society of Shoulder and Elbow Surgeons (ASES) score,[15] (4) Western Ontario Rotator Cuff (WORC) score,[13] (5) Simple Shoulder Test,[9] (6) Disabilities of the Arm, Shoulder, and Hand (DASH) questionnaire,[11,24] and (7) Short Form 36-Item Health Survey.[3]

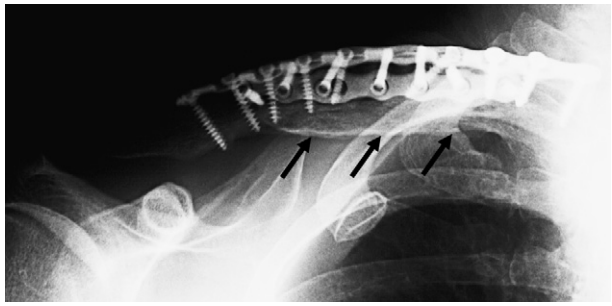

Figure 5 Radiograph shows results at 40 months after the final fixation surgery and just before metal removal. Hypertrophic callus has increased the final diameter of the midshaft clavicle to nearly 3 times greater than that of the patient's left clavicle. The *arrows* indicate the lower edge of the healed bone.

The fracture was solidly united by 3 months postoperatively, and by 7 months, the patient's affected hand exceeded the grip strength of the opposite (left, nondominant; Table I). At 9 months, he reported an episode of transient (several hours) weakness in grip strength with heavy lifting. One additional month of only light lifting was recommended, and these symptoms did not recur.

Long-term follow-up

By 18 months, the only activity that caused significant symptoms was hammering nails into fence posts, which caused fatigue in his hand and mild weakness with grip. Because of this he inquired if decompression surgery would provide definitive relief of these symptoms.

Table I Outcome analysis from 4 to 48 months after final fracture fixation surgery

Outcome	Months after operation						
	4	7	15	24	36	39	48*
Grip strength in lbs†							
Left	143	125	125	135	145	-	-
Right	114	129	138	140	149	-	-
Active forward flexion	120	-	165	180	180	180	180
Typical VAS score, 10-cm (best = 0)	-	-	1.5	1.4	0.3	1.3	0.1
ASES score (best = 100)	-	-	84.2	88.0	98.5	93.5	99.0
WORC score (best = 0), No. (%)	-	-	866 (59)	642 (69)	219 (89)	191 (91)	37 (98)
Simple Shoulder Test (no. of yes responses/ no. of questions)‡	-	-	7/12	11/12	11/12	12/12	12/12
DASH score (best = 0)	-	-	-	19.2	3.3	5.8	0.8
Short Form 36-Item Survey	-	-	92.5	92.5	92.3	98.1	92.5

* 8 months after metal removal. Hardware was removed 40 months after final fixation surgery; rib resection was not done and no bone was removed near thoracic outlet.

† Right side is affected. One lb = 0.45 kg.

‡ Yes responses correlate with better shoulder function than no responses.

Two vascular surgeons, who were consulted for independent opinions, initiated vascular and electrodiagnostic work in October 2005. Nerve conduction and electromyographic analyses revealed decreased ulnar sensory action potentials. Other electrodiagnostic abnormalities seen with neurologic TOS were not detected, however, including no evidence of prolongation of the ulnar F-wave, decrease in compound muscle action potential, or electromyographic abnormalities in muscles supplied by the lower trunk of the brachial plexus. Doppler segmental pressures and waveforms, and pulse volume recordings showed similar findings in the right and left (unaffected) sides. Doppler ultrasound imaging showed no evidence of venous thrombosis. Digital photoplethysmography revealed normal waveform amplitude and contour in all five digits of both hands at rest, and pulsatility of the radial artery was normal at 0°, 90°, and 180° of arm abduction. However, with military and Adson's maneuvers, the flow in the right upper extremity was diminished but was unchanged in the left (unaffected) upper extremity. These latter findings were consistent with some arterial stenosis, but a right upper extremity venogram did not reveal focal narrowing or outflow obstruction of the subclavian vein with 0°, 90°, and 180° of shoulder abduction.

Although there was evidence of mild arterial TOS, it was concluded that his symptoms were primarily attributable to a mild brachial plexopathy. Observation was continued for 1 more year, during which the symptoms that were concerning to him further diminished. By then the hardware had become prominent and was removed 40 months after the final fixation surgery (Figure 5). At 8 months after hardware removal (48 months after the final fixation surgery), improvement was seen in all shoulder outcome measures (Table I). In a telephone interview at the final follow-up at 14 months after hardware removal (4.5 years after the final fracture fixation surgery), he reported that the subtle upper extremity fatigue with hammering nails was "tolerable." He was very satisfied with his function.

Discussion

As is typical of iatrogenic TOS, the surgeon initially failed to recognize that the revision osteosynthesis, which restored anatomic alignment of the clavicle, compressed the forming callus onto the thoracic outlet. In view of previous publications on this topic, our case is novel because the TOS occurred within 60 days of the fracture in the setting of

Table II Selected cases with some characteristics or issues similar to our case

Details	Study (first author)						
	Present study	Miller, 1969	Braun, 1979	Koss, 1989	Kitsis, 2003	Stötzer, 2005	Papagelopoulos, 2006
Patients, No.	1	4	2	1	17	1	1
Clavicle fracture	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Iatrogenic TOS	Yes	No	No	No	No	Yes (fixation of comminuted fx)	Yes (fixation of nonunion)
TOS corrected by ~3 months	Yes	Yes	No	No	NK	Yes	Yes
Noncompliance caused fixation failure	Yes	No	NA	NA	NA	No	NA
Rib resection	No	No	Yes (2 patients)	No	NK	No	No
Type of TOS (vascular, neurogenic, or both)	Both	Vascular	1 vascular, 1 neurogenic	Both	2 vascular, 10 neurogenic	Vascular	Both
Time to clear recognition of sxs	12 h after revision fx fixation	1-3 mon after fracture	8-12 mon after primary fx fixation	3.5 y after fx	NK	2 d after primary fx fixation	NA
Duration of sxs	14 d	1-3 mon	~6 mon or more	4 mon recurred 3.5 y later	NK	2 d	24 hr
Callus contributed to TOS	Yes	Yes	Yes	Yes	Yes (11)	No	Yes
Time from fx to surgery for TOS	55 d	3 patients <5 w; 1 patient ~3 mon	~12 mon (1); ~8 mon (1, nonunion)	3.5 y (nonunion)	NK (some nonunions)	< 1 w	~18 mon (nonunion)
Occupation	Rancher	NK	NK	NK	NK	NK	Manual laborer

fx, Fracture; *NA,* not applicable; *NK,* not known; *TOS,* thoracic outlet syndrome; *sxs,* symptoms.

multiple subacute fixation failures. These recurrent injuries and surgeries were associated with nascent callus formation that was not yet detectable on standard radiographs.

Our review of English and German literature (summarized in Tables II and III)[2,4-8,10,12,14,16-22,25] showed that previously reported cases of iatrogenic TOS associated with surgery for clavicle fractures have been (1) associated with fracture nonunions, malunions, or hypertrophic callus, or both, but not in association with multiple subacute fixation failures, or (2) were diagnosed/detected relatively much later or symptoms developed much later in surgical cases, typically after surgical fixations for nonunions, or both.

This patient is also unusual because of the 16-day delay in the revision/decompression surgery with gap plating. The absence of calcified callus on radiographs coupled with what was thought to be a large hematoma steered the orthopedic and vascular surgeons away from the correct diagnosis. Failure to recognize the urgency of a definitive diagnostic radiographic workup (ie, venogram) also contributed to this delay.

Multiple procedures are often required in the surgical management of clavicle fractures associated with brachial plexopathy or TOS, or both. For example, TOS was evident in 12 of the 17 patients with high-energy clavicle fractures reported by Kitsis et al[14] in 2003. They performed 11 decompressions with callus excision, 3 simple neurolysis, 8 fixations for nonunion, 1 osteotomy, and 3 excisions of bone spikes.[14] Two patients required a further decompression procedure, 1 required sequestrectomy for deep sepsis, 2 patients required removal of hardware. Fixation failure occurred in 2 patients after surgery for nonunion, but unlike our patient, these were for single/nonacute fixation failures.

In the context of our patient, an additional notable finding in the Kitsis study[14] is that only 5 of the 14 patients were asymptomatic or had minimal symptoms after these surgeries. In fact, most of the patients continued to experience some degree of residual symptoms: 8 patients had persistent dysesthesia and 3 had persistent cold intolerance. The recommendation for avoiding surgery in our patient was couched in the perspective of these results.

Among the case reports describing complications associated with clavicle fractures identified in our literature review, only a few are cases of iatrogenic TOS having similarities with our patient (Table II). Stötzer's[25] case is the most similar and deals with acute iatrogenic TOS that

Table III Selected literature review of reports describing cases of fracture- and iatrogenic-related thoracic outlet syndrome

First author	Year	Patients	TOS associated with fx*	Type of TOS (V, N, C)	TOS associated with surgical fx fixation†	V, N, or C TOS	Rib resection?	Multiple subacute revision surgeries?§
		Total N	No. (%)	(fx cases)	No. (%)	(fixation cases)	(for fx cases only)	
Papagelopoulos	2005	1	0 (0)	NA	1 (100)‡	100% C	No	No
Casbas	2005	13	8 (61.5)‡	37% V, 13% C, 50% aneurysms	0	NA	3/8	No
Stötzer	2005	1	1 (100)	V	1 (100)	100% V	No	No
Davidovic	2003	25	2 (8)‡	100% V	0	NA	2/2	No
Kitsis	2003	17	12 (71)‡	17% V, 83% N	0	NA	No	No
Parry	2000	1	1 (100)‡	C	0	NA	No	No
Leffert	1999	282	12 (4)‡	NK	NK	NK	12/12	No
Connolly	1989	15	15 (100)‡	47% C, 53% NK	0	NA	No	No
Koss	1989	1	1 (100)‡	C	0	NA	No	No
Dannöhl	1988	2	2 (100)‡	50% V, 50% N	0	NA	1/2	No
Jupiter	1987	23	4 (19)‡	100% C	0	NA	2/4	No
		5 additional cases with brachial plexopathy/paresthesias						
Manske	1985	10	3 (30)‡	100% N	0	NA	No	No
Braun	1979	2	1 (50)	100% V	1 (50)‡	100% N	2/2	No
Mulder	1973	7	3 (43)‡	67% C, 33% V	1 (14)‡	100% N	No	No
Enker	1970	1	1 (100)	C	0	NA	No	No
Miller	1969	4	4 (100)	100% N	0	NA	No	No
Howard	1965	14	11 (78.5)‡	18% V, 27% N, 36% C	0	NA	No	No
		1 case with aneurysm; 1 case with arteriovenous fistula						

NA, Not applicable; *NK,* not known or not clear; *TOS,* thoracic outlet syndrome (*V,* vascular, *N,* neurogenic, *C,* combined).

* The fracture (fx) caused or was associated with TOS either initially as the result of a nonunion (most typically) or malunion, but before surgical fixation or reconstruction.

† TOS was associated with surgical fixation of the fracture (fx) or fracture nonunion, even if this caused compression of the thoracic outlet by fracture callus.

‡ Case, or cases, associated with clavicle fracture (fx) nonunion or pseudoarthrosis.

§ Subacute refers to < 90 days after initial clavicle fracture.

developed in a patient after percutaneous intramedullary fixation of a highly comminuted clavicle shaft fracture. Revision surgery within a few days after the initial fracture fixation included the application of a plate, screws, and cerclage wires. The anatomic reduction of the inferiorly displaced fracture fragments provided immediate resolution of symptoms by decompressing the outlet. Unlike our case, the TOS in their patient was not associated with fracture callus or with multiple subacute fixation failures.

Abundant callus can develop relatively soon after a clavicle fracture caused by severe trauma in adults. For example, Miller and Boswick[19] reported 4 adults who had sustained clavicle fractures with subsequent neurologic TOS or brachial plexitis that was attributable to abundant callus.[19] In 3 patients the compression caused by exuberant callus was treated within 5 weeks of the injury; 1 underwent a claviculectomy, and the callus was debulked in the other 2 patients. In the fourth patient, the callus was excised and the clavicle was lengthened by Z-plasty and internally fixed; this was done just beyond 3 months after the injury. In contrast to our patient, these 4 patients did not have subacute fixation failures, and their symptoms were not associated with surgical fixation.

In conclusion, this report emphasizes that surgeons must be vigilant in considering the potential complication of iatrogenic TOS during clavicle fracture fixation, even when the fracture callus has not yet ossified or when surgery is within 60 days of injury, or both. Multiple subacute failures (2 in our case), resulting in multiple open reductions with fixation, can increase the volume of callus formation and further increase the likelihood of iatrogenic TOS during anatomic osteosynthesis. Surgeons should have a low threshold for diagnostic testing (ie, venography) should signs of TOS develop in this context. The use of venography must be judicious, however, because complication rates have been reported to be 1% to 6%, ranging from relatively minor (eg, local swelling or hematoma at the injection site, cellulitis, and minor contrast agent reactions) to severe (eg, pulmonary embolism, and kidney failure or anaphylaxis from contrast agents).[1,23]

Disclaimer

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