



Acute Arterial Thoracic Outlet Syndrome after Intramedullary Fixation for Nonunion of a Midshaft Clavicular Fracture

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In this case report, we describe a rare clinical situation of acute arterial thoracic outlet syndrome (TOS) after osteosynthesis, using an intramedullary Knowles pin, to treat nonunion of a midshaft clavicular fracture. Existing evidence was reviewed to appraise the relationship between TOS and nonunion of clavicular fractures, including the timing of the development of this complication, as well as to identify the relationship between surgical technique and choice of implant and the occurrence of TOS. Perioperative measures used to prevent TOS were identified.

Key words: Clavicular nonunion, thoracic outlet syndrome, subclavian arterial injury

INTRODUCTION

Clavicular fractures account for approximately 2.6% of all fractures, with the majority being midshaft fractures.¹ A recent meta-analysis reported a nonunion rate as high as 14.5% for displaced midshaft fractures treated nonoperatively.² Moreover, a specific association between thoracic outlet syndrome (TOS) and clavicular nonunion has been reported.³⁻⁵ On the other hand, osteosynthesis for clavicular fractures is a reliable treatment, with rates of midshaft nonunion as low as 1.4%.² Although clavicular osteosynthesis decreases the rate of nonunion, TOS remains a possible postoperative complication. In this case report, we describe a rare clinical occurrence of acute arterial TOS after osteosynthesis for the clavicular nonunion. This complication was noteworthy as it required further therapeutic management.

CASE REPORT

Informed consent was obtained from the patient after the nature of the procedure had been explained. The patient was a 34-year-old man who had sustained a fracture of the left clavicle in a motor vehicle accident [Figure 1a]. The fracture was treated with osteosynthesis using a locking plate [Figure 1b]. Since the patient was a young and active laborer, a clinical decision was

made to remove the plate 1 year after fixation to avoid local irritation from prominent hardware [Figure 1c]. Following plate removal, the patient developed a progressively painful shoulder. Follow-up radiographs revealed a nonunion at the fracture site with displacement of the fragments [Figure 2a]. A revision osteosynthesis was performed using an open procedure without fluoroscopy guidance, and a 3.2 mm Knowles pin (Zimmer) was used for fixation.

Postoperatively, the patient developed pain, soreness, and numbness in his left arm with gross observable local swelling over his left shoulder and left upper limb congestion and a decreased left radial pulse and asymmetrical blood pressure between bilateral limbs. Postoperative radiographs revealed malpositioning of the Knowles pin [Figure 2b], with intimal dissection of the left subclavian artery and high-grade stenosis⁶ evident on computed tomography angiography [Figure 3]. Findings were indicative of an acute left subclavian arterial injury combined with TOS, and a cardiovascular consultation was arranged. Orfarin (2.5 mg orally/day) was prescribed, and the signs and symptoms gradually resolved over 1 week. Anticoagulant therapy continued for 1 month. Four months after the TOS diagnosis, radiographic evidence of bone union was visible, and the Knowles pin was removed [Figure 4a and b].

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Acute arterial thoracic outlet syndrome

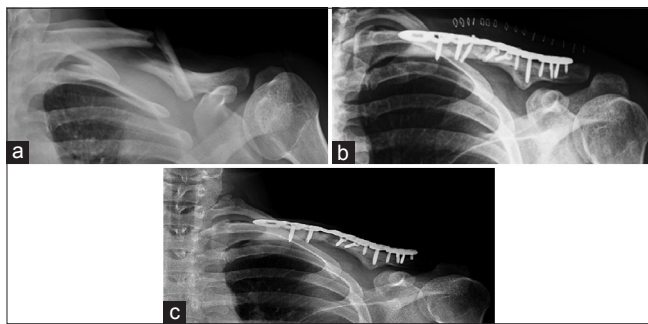


Figure 1: Preoperative anterior-posterior radiograph showing the comminuted midshaft fracture of the left clavicle, (a) Postoperative anterior-posterior radiograph showing the anatomical reduction of the left clavicle fracture with fixation using a plate and screws, (b) Preoperative anterior-posterior radiograph before the plate removal, indicating potential nonunion of the fracture site. A cortical screw in the proximal fragment was removed previously under local anesthesia owing to loosening and skin impingement (c)

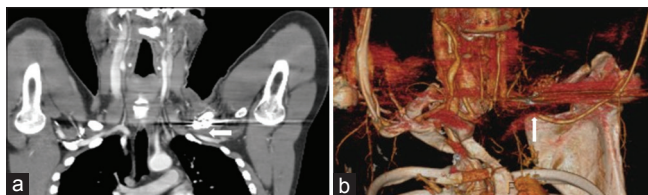


Figure 3: Computed tomography arteriography showing a short segment intimal dissection of the left subclavian artery with high-grade stenosis (white arrow); blood flow through the stenosis is visible, (a) Three-dimensional reconstruction of the computed tomography arteriography image, showing segmental nonvisualization of the left subclavian artery (white arrow) (b)

DISCUSSION

TOS is a disorder caused by compression of the brachial plexus and/or subclavian vessels between the clavicle and first rib, an anatomical region known as the thoracic outlet or an acute compartment syndrome occurred within the thoracic outlet.⁷ The diagnosis of TOS is based on a physician's judgment after a meticulous history taking and physical examinations.⁷⁻⁹ The vast majority of TOS cases are neurogenic (>95%), with an arterial cause identified in 1% of cases. Despite the anatomical proximity of the subclavian vessels to the clavicle, vascular complications with clavicular fractures are rare occurrences, with TOS reported as an acute or late complication due to malposition of the clavicle fracture fragment nonunion, malunion, or hypertrophic callus formation.^{10,11}

There is limited evidence of iatrogenic causes of TOS after clavicular osteosynthesis, with reported late-onset of TOS attributed to misalignment of the fracture, excessive callus, nonunion and screw impingement after plating.^{8,12,13} In our case, acute arterial TOS developed following revision osteosynthesis using an intramedullary Knowles pin and caused acute limb ischemia requiring immediate thrombolysis.⁹ Based on this, we searched for previously described preventative methods.

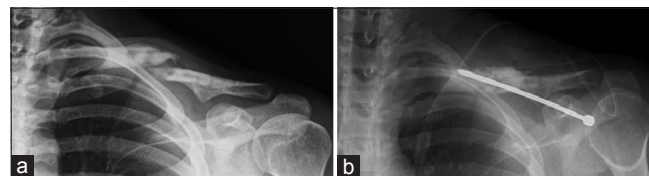


Figure 2: Anterior-posterior radiograph showing an atrophic nonunion of the clavicular fracture with displacement, (a) Postoperative anterior-posterior radiograph of the left clavicle, showing a malpositioning of the Knowles pin through the scapula to the medial clavicle fragment (b)

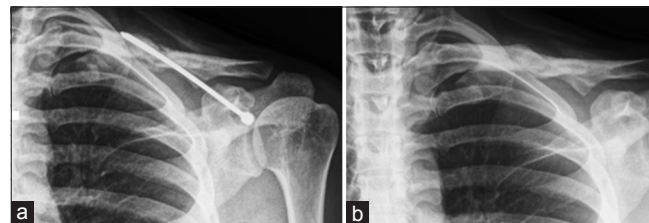


Figure 4: Anterior-posterior radiographs demonstrated bone union before Knowles pin removal (a) and after the procedure (b)

Osteosynthesis for nonunion of a mid-shaft clavicle fracture is associated with a >2-fold increase in the risk of postoperative complication within a month, compared to acute clavicle fractures.¹⁴ Although TOS is generally considered to be an acute or late, limb-threatening postoperative complication,^{8,9,12} TOS has occasionally been reported as an acute complication after osteosynthesis. Jennings *et al.* reported TOS development with occlusion of the mid-subclavian artery confirmed on arteriography with ipsilateral arm in a position of >80° of abduction in a patient who underwent revision surgery for atrophic nonunion of the clavicle be checked for the same side radial artery pulsation.¹³ Based on their experience, the authors suggest that passive intraoperative arm abduction be completed during all clavicular nonunion osteosynthesis operations since this maneuver provokes compression of the thoracic outlet and may allow early detection of vascular TOS. In our case, superficial swelling and congestion of the upper extremity developed within a few hours postoperatively, associated with paresthesia which subsequently progressed. Therefore, performing the arm abduction maneuver intraoperatively might have been beneficial for this patient. The arm abduction maneuver is considerable to be performed in all cases of nonunion or malunion clavicular fractures following operations for these clavicle fractures by osteosynthesis, although the sensitivity and specificity of the maneuver in predicting TOS have not yet been clarified.

Osteosynthesis with plating is the gold standard for the treatment of a nonunion fracture of the clavicle,¹⁵ providing the longer term stability required for effective management of these fractures. Open intramedullary fixation also provides the advantage of using bone grafting. Compared to plate fixation,

intramedullary fixation offers several advantages, including the use of a smaller incision with better cosmesis, less soft tissue stripping, decreased hardware irritation, and easier hardware removal (often under local anesthetic).^{16,17} Lee *et al.* reported a case of clavicular nonunion complicated with venous and neurogenic TOS, in which bone union was achieved by revision osteosynthesis using a Knowles pin and an autogenous bone marrow graft.⁴ Our patient had initially been treated using a precontoured anatomical locking plate. However, the timing of plate removal turned out to be inappropriate because the clavicular fracture failed to achieve union. Consequently, we selected a Knowles pin for intramedullary fixation to treat the clavicle shaft nonunion after plate removal. When performing the retrograde drilling of the distal fragment, it is important to acknowledge that nonunion with fibrous scarring may mislead the axis. Adequate debridement of the fracture site is required and the position of intramedullary fixator should be checked by the fluoroscopy. In our case, TOS resulted from intraoperative malpositioning of the Knowles pin causing vascular injury because these procedures were not performed exactly. Subsequent treatment for arterial TOS, including delivering thrombolytic agents, thrombectomy, and decompression with or without bypass surgery, depends on the severity of the disease.^{9,10}

To our knowledge, this is the first case to describe TOS as a complication of intramedullary fixation with a Knowles pin for nonunion of a clavicular fracture. Based on our experience, we propose the following components be included in the operative treatment of nonunion/malunion of a fracture of the clavicle: Preoperative neurovascular assessment to be used as a baseline reference for postoperative assessment; intraoperative fluoroscopy and physical examination, including the arm abduction maneuver, to provide opportunity for adjustment of the fixation hardware; and careful postoperative monitoring for TOS, including diagnostic testing for any suspicious case. We believe these concepts and methods promote the safety of revision clavicular osteosynthesis in that iatrogenic TOS can be detected and prevented intraoperatively.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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