Introduction

Fractures of the clavicle are one of the most common bony injuries of childhood, accounting for 15% of the fractures in this population.\(^\text{[1]}\) The majority of these occur in the midshaft region and approximately half are displaced fractures.\(^\text{[1]}\) Nonunion of the clavicle is a recognized complication in the adult population, with reported rates of between 1% overall and 15% in displaced fractures.\(^\text{[2]}\) It is an extremely rare complication in children, with seven published cases in the English literature. We describe a case of a posttraumatic clavicle nonunion in a 13-year-old boy which caused significant dilatation of the external jugular vein (EJV) as a result of a traumatic arteriovenous fistula.

Case Presentation

A 13-year-old boy was referred to clinic with a prominent dilatation on the right side of his neck that had been gradually enlarging since he sustained a displaced fracture of the middle third of his right clavicle 6 months previously. This was treated conservatively by the referring hospital [Figure 1a]. The patient was uninhibited in daily activities, but complained of ongoing pain at the fracture site and was concerned about his cosmetic appearance. His past medical history consisted of a congenital atrial septal defect with partial anomalous pulmonary venous drainage, which was surgically corrected at the age of 18 months. On examination, there was a nonpulsatile dilation in the right anterior triangle of his neck overlying the clavicle which was tender on palpation. There was no other visible deformity and he had full range of motion of the ipsilateral shoulder.

An ultrasound and computed tomography (CT) scan were arranged, which revealed a hypertrophic angulated nonunion of the clavicle fracture. The surrounding callus formation was shown to be compressing the EJV, impeding its drainage, and causing the striking distension measuring 2 cm in diameter. The anatomy of the carotid and subclavian arteries appeared normal.

Following multidisciplinary discussion, a decision was made to operatively reduce and fix the clavicle nonunion. Intraoperatively, gross callus formation was found at the
fracture site. However, an arteriovenous fistula was also discovered in a vascular cavity in the callus between a branch of the subclavian artery and the EJV. The fistula was excised and adjacent veins ligated and closed by vascular surgeon before the clavicle fracture was reduced and fixed with a six-hole contoured plate without the use of bone graft.

Radiographs taken 1 week postoperatively confirmed satisfactory position of the clavicle fixation. At 1-month follow-up, the EJV was less prominent and there was no residual pain around the shoulder. At 6-months follow-up, the patient was asymptomatic with full range of movement of the ipsilateral shoulder. Radiographs and CT scans showed union of the fracture site with good alignment of the clavicle [Figure 1b]. A CT venogram revealed an enlarged right EJV with a stenosed lower end and tortuous collaterals to the right internal jugular vein and subclavian vein, but no residual arteriovenous fistula [Figure 1c].

Discussion

Complications associated with clavicle fractures include malunion, nonunion, thoracic outlet syndrome, vascular injury, and brachial plexus injury. Fracture displacement is associated with an increased risk of nonunion in adults,[2] although this does not seem to be a risk factor in the pediatric population.[1]

Table 1: Pediatric posttraumatic clavicle nonunions in literature

| Author                  | Age (years) | Sex | Mechanism               | Fracture site                        | Time to diagnosis of nonunion | Treatment                                      | Outcome                                      |
|-------------------------|-------------|-----|-------------------------|---------------------------------------|-------------------------------|-----------------------------------------------|----------------------------------------------|
| Nogi[7]                 | 12          | M   | Fall from chair         | Junction distal and middle third right clavicle | 6 months                     | Excision of distal clavicle and stabilization of proximal clavicle | Asymptomatic with full activity at 12 months |
| Caterini[8]             | 7           | F   | Fall onto right shoulder | Middle third right clavicle           | 14 months                    | Kirschner wire stabilization with bone grafting from tibial metaphyseal        | Pain free, full range of movement at 12 months |
| Spapens[9]              | 8           | F   | Fall off bicycle        | Midshaft right clavicle              | 12 months                    | Compression plate fixation with bone grafting from iliac crest               | Asymptomatic with full activity at 10 months |
| Wilkins and Johnston[9] | 7           | M   | Severe Fall             | Not described                        | 72 months                    | Screw with bone graft                | Full unlimited movement                    |
|                         | 13          | M   | Severe Fall             | Not described                        | 10 months                    | None                              | Nonunion, unlimited activity               |
| Jain[10]                | 7           | F   | Fall                    | Refracture of junction middle and proximal third right clavicle | 14 months                    | Compression plate fixation          | Full range of movement at 6 months         |
|                         | 9           | F   | Not described           | Midshaft right clavicle              | 23 months                    | Plate fixation                     | Full range of movement at 6 months         |

M = Male, F = Female
Vascular complications as a result of clavicle fractures are rare, but are recognized as either an immediate complication due to transection of the vessel by the displaced fracture or as a late complication, secondary to compression from abundant callus formation. These include subclavian artery or vein compression, thrombosis, or pseudoaneurysm. There are several reports of upper extremity deep venous thrombosis following clavicle fractures. We are not aware of any other reported case in English literature that describes the formation of a traumatic arteriovenous fistula and symptomatic compression of the EJV following clavicle fracture. There are no known associations between congenital cardiac anomalies and the later development of arteriovenous fistulas.

Complications following pediatric clavicle fractures are uncommon and healing usually occurs within 4-6 weeks. A study by Calder and colleagues recommended that children with isolated fractures of the clavicle with no complications at initial follow-up may be safely discharged from further follow-up. Posttraumatic nonunions of clavicle fractures in children are extremely rare, with only isolated reports in the literature. All cases presented with hypertrophic nonunion between 6 and 72 months following injury and all but one case were treated operatively. Compression plate fixation was the most common procedure, and half of the procedures also involved bone grafting of the fracture site. Union was only achieved through surgery, although the functional outcome was satisfactory in all cases.

This is the first reported case of nonunion of a clavicle fracture causing symptomatic EJV compression and a traumatic arteriovenous fistula formation. This required operative fixation of the fracture and ligation of the fistula to achieve a satisfactory functional outcome for the patient. Although current literature advises against routine follow-up for isolated pediatric fractures, the authors believe that follow-up for displaced pediatric clavicle fractures is good practice. Open reduction and internal fixation reliably restores clavicle length and angulation, with low complication rates and good radiological and clinical outcomes for pediatric clavicle nonunions.

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