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ANATOMIC VARIATIONS

Clavicle duplication following physeal injury

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Abstract
Clavicle duplication is a rare entity with limited number of cases reported. Congenital origin and trauma related mechanisms are the main hypotheses to explain this anatomical variation. In skeletally immature patient, trauma may cause physeal-metaphyseal injury in the distal clavicle rather than acromioclavicular strain. The distal epiphysis remains in continuity with acromion and has an intact periosteal sleeve. The periosteal sleeve is extremely osteogenic, and may lead to new bone formation between epiphysis and displaced metaphysis. This remodeling potential and the intact acromioclavicular joint allow the children to be followed by closed reduction. However, there can be new bone formation between epiphysis and displaced metaphysis, resulting in clavicle duplication. Herein, we present the radiographic and computed tomography findings of a post-traumatic duplication of the clavicle in a 5-year-old boy.

Keywords Clavicle duplication · Bifurcation · Computed tomography · Clavicle · Fracture

Introduction

Clavicle fractures constitute around 2.6–4% of all fractures, with lateral end fractures comprising 21–28% of all clavicle fractures [11]. The clavicle is unique since its ossification commences early and completes around 21–25 years of age. Traumatic injuries of the distal clavicle prior to completion of ossification differ since the bone is weaker than the acromioclavicular ligaments. Following trauma, distal end may remain in its anatomical position by intact acromioclavicular ligament and fracture may occur at physeal–metaphyseal junction [7, 9]. In addition, the periosteal sleeve is osteogenic and has great capacity for remodeling and may fill any gap between the periosteum and metaphysis [12]. This may cause shoulder deformities in children. One of these rare deformities is duplication of the clavicle [4, 8, 15].

Clavicle duplication is a rare entity with limited number of cases reported. Clavicle duplication may occur in isolation or in association with acromioclavicular joint duplication. Congenital origin and trauma related mechanisms are the main hypotheses to explain this anatomical variation [9, 12, 14]. Congenital origin theories are based on supernumerary ossification center or displacement and fragmentation of the ossification centers [12]. Trauma theory has been supported by a limited number of clavicle duplication following distal clavicular physeal injuries [8, 9, 15]. Herein, in support of the trauma theory, we present the radiographic and computed tomography findings of a post-traumatic duplication of the clavicle in a pediatric case.

Case report

A 5-year-old boy with a diagnosis of hemolytic uremic syndrome was under regular follow-up in our institution. The patient presented to the emergency room after a motor vehicle accident. Plain radiographs of right shoulder showed fracture of lateral end of clavicle with intact position of lateral end with regards to acromion. The medial fragment was displaced caudally (Fig. 1). There was no increase in coracoclavicular distance. Patient was treated with an arm sling. Four months following trauma, the patient presented with tenderness over the sternal region. Imaging showed that the fracture has healed with formation of a Y-shaped clavicle (Fig. 1c). Computed tomography revealed that the distal epiphysis had maintained its position with respect to acromion with intact acromioclavicular joint. There was new

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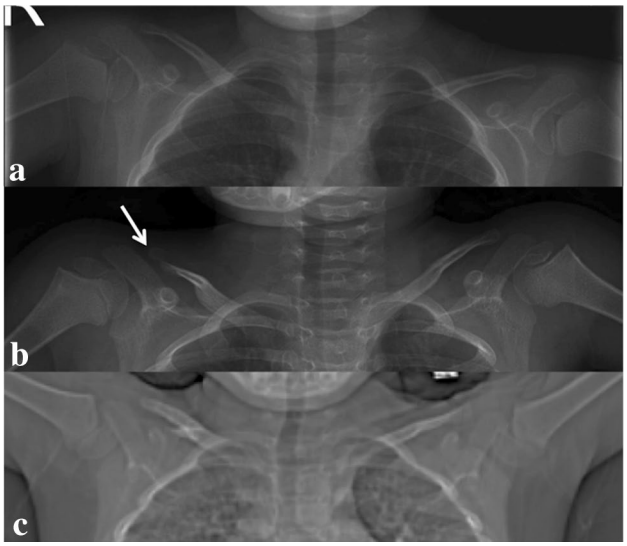

Fig. 1 A 5-year-old boy with a diagnosis of hemolytic uremic syndrome. **a** PA X-ray prior to motor vehicle accident does not reveal any abnormality in both clavicles. **b** X-ray of the right shoulder immediately after trauma reveals displaced fracture of distal clavicle (arrow). (Group IIb as based on the classification proposed by Nenopoulos et al.). **c** Computed tomography topogram 4 months later shows duplication of the clavicle

thick bone formation towards middle 1/3 of clavicle. The intact proximal segment of the clavicle was displaced inferoposteriorly (Figs. 2, 3). Volume rendering was carried out to depict the anatomy more clearly (Figs. 2, 3).

Patient had no complaints with regards to shoulder movement; however, there was a palpable lump over the shoulder girdle.

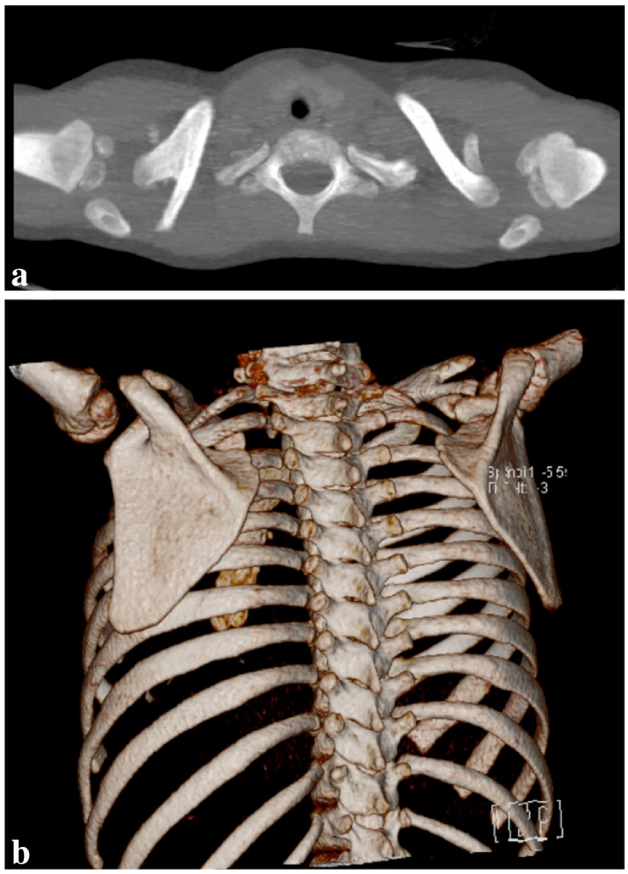

Fig. 2 Axial maximum intensity projection image from superior (**a**) and volume rendering image from a slightly posteromedial view (**b**) obtained from computed tomography performed for 4 months following trauma, reveal that the intact distal epiphysis maintained its position with acromion and there was new bone formation towards middle 1/3 of clavicle body. There was no duplication in the acromioclavicular joint

Discussion

In the literature, there is a limited number of cases reporting duplication of the distal clavicle. Different terms like bifurcation, bifid clavicle, os subclaviculare, Y-shaped and duplication have been used to describe this finding [1, 3, 10, 12, 13, 16,]. Though more recent literature mainly uses the term duplication, bifid or Y-shaped clavicle better suits the variation. Clavicle duplication should not be confused with the more common coracoclavicular articulation or calcification of the coracoclavicular ligament [14]. Duplication of the clavicle may be in isolation or occur as part of an acromioclavicular joint duplication. Clavicle duplications usually have a Y-shape with duplications on the lateral side and fusion medially [6, 9, 10, 13–15]. Only one end of the duplication may remain in continuity with acromion, or acromion can articulate with both lateral ends of the clavicle. In our case, the duplicated part with distal clavicular physis remained in continuity with acromion; however, the proximal clavicular segment was displaced inferoposteriorly and did not articulate with the acromion. There can also be duplication of the clavicle where duplicated clavicle is inferiorly located with a medial free end and lateral attachment to the coracoid process [3]. Additionally, the two clavicular parts may be fused laterally with both articulating with the acromion and medial end of duplicated part articulating with coracoid process [12].

Two hypotheses, namely congenital origin and trauma causing distal physeal injury, have been proposed to explain the clavicle duplication [9, 12, 14]. Embryologically, the clavicle is the first bone to ossify by intramembranous ossification during the 5th–6th weeks of gestation. Ossification starts in the mid part of the membranous bone by two separate ossification centers (medial and lateral) [2]. The two centers unite to form the S-shaped clavicle. Developmental anomalies in these centers may result in aplasia or hypoplasia of that segment of the clavicle.

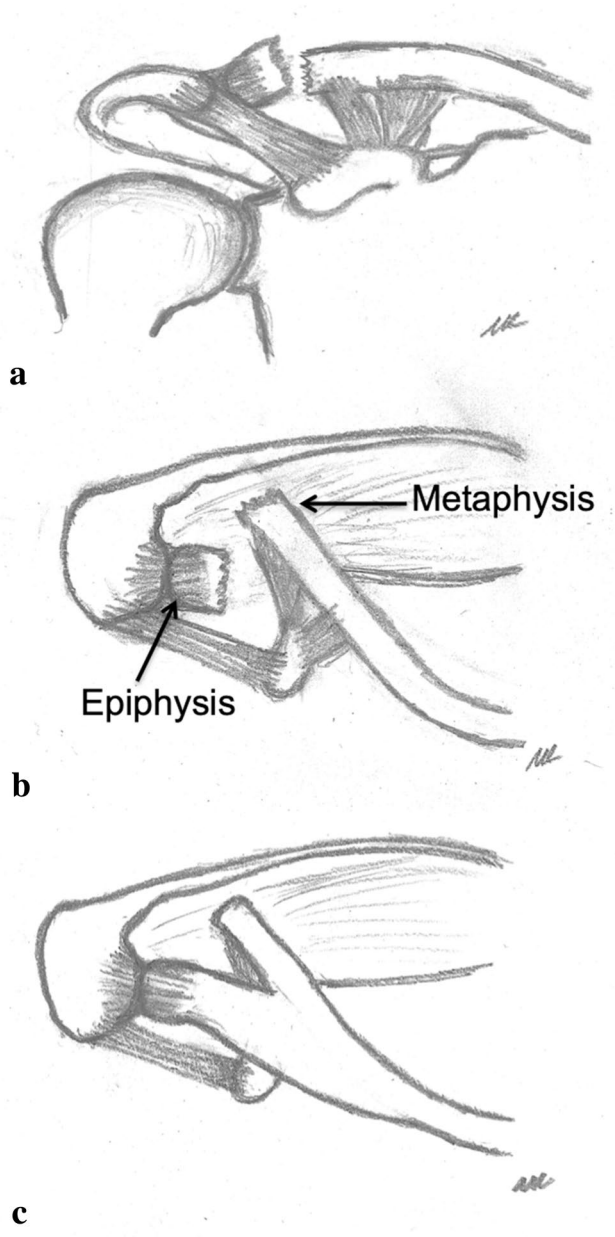

Fig. 3 Schematic drawing from anterior (**a**) and superior (**b**) view of the distal clavicular injury pattern. The epiphysis maintained its continuity with acromion and the proximal metaphysis was displaced inferoposteriorly through a tear in periosteum. The coracoclavicular ligaments remained intact. Schematic drawing from superior view (**c**) shows new bone formation from distal epiphysis toward the midportion of the clavicle whereas the inferoposteriorly displaced proximal metaphysis lost its continuity with acromion resulting in a bifid clavicle

Clavicle duplication by congenital origin may occur either by displacement of these two ossification centers or presence of more than two ossification centers with displacement of the extra ossification center [12]. However, presence of more than two ossification centers has not been reported in these reports and displacement of one of the two ossification centers would result in the main part of the clavicle being hypoplastic or absent. A new theory to explain the congenital origin, namely the fragmentation theory of the ossification center has been proposed to better explain clavicle duplication [12]. In this theory, early in development a small fragment of the ossification center detaches and gives rise to a duplicate clavicle. If the fragment separates out completely, this can result in a completely separate supernumerary clavicle; or if the fragment maintains its connection with main ossification center then the duplicate clavicle may remain fused. Duplication of the clavicle along with acromioclavicular joint duplication supports the congenital origin hypopthesis. Gowland in 1915 and Twigg et al. in 1981 reported duplicated acromioclavicular joints, whereas Sharma in 2003 reported clavicular duplication associated with a coracoid process tripartition [4, 12, 13].

In the initial reports, no link between trauma and clavicle duplication had been ascertained. Ogden was the first to recognize trauma as a causative factor in clavicle duplication and has proposed that the early reports occurred as a result of unrecognized injuries [9]. The case by Rutherfurd in 1921 had lateral clavicle duplication with a single articulation with acromion, though Rutherfurd did not entertain a traumatic origin as the surgically removed duplicate part had "conformity to the normal part" [10]. Later on new case reports and case series of distal clavicle fracture have demonstrated the formation of clavicle duplication in the post-traumatic period [5, 8]. Trauma as a causative factor in clavicle duplication can be related to distal physeal injury [9]. Nenopoulos et al. have classified the distal clavicular injuries in skeletally immature patients to evaluate the long-term follow-up and also to propose an algorithm for treatment [8]. The distal clavicle and epiphysis is a relatively weak structure; however, it is an important attachment site for acromioclavicular and coracoclavicular ligaments. These ligaments attach densely into the perichondrium of the distal clavicular epiphysis and then subsequently blend into the periosteum. This complex has resilience and ligament disruption is less likely in the immature skeleton, compared to the adult where trauma usually causes acromioclavicular separation. Instead, the physeal–metaphyseal junction is the weakest region in children and is the usual site of disruption following trauma [9]. After separation of the distal epiphysis from the metaphysis, the metaphysis may rupture through the periosteal sleeve, leaving the distal epiphysis intact. The periosteal sleeve is extremely osteogenic, and will lead to new bone formation between epiphysis and displaced metaphysis [15]. This remodeling potential and the intact acromioclavicular joint allow the children to be followed by closed reduction and the functional results are usually excellent regardless of the preferred method of treatment [8].

However, this remodeling as demonstrated in our case may lead to a partial duplication of the clavicle.

Clavicle duplication is a rare entity that can be either congenital in origin or related to trauma in the distal physis. In cases of clavicle duplication not associated with acromioclavicular joint duplication, distal physeal injury is a plausible underlying mechanism. As demonstrated in our case, in a skeletally immature patient, trauma may cause physeal-metaphyseal injury in the clavicle rather than acromioclavicular strain. The great osteogenetic capacity of the intact periosteum may lead to new bone formation between epiphysis and displaced metaphysis, resulting in clavicle duplication.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no competing interest.

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