Carpal coalition

A review of current knowledge and report of a single institution's experience with asymptomatic intercarpal fusion

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Abstract

Background Carpal coalition is a relatively common, yet poorly recognized, anomaly that may present independently or in association with syndromic or metabolic disorders. While largely asymptomatic, symptoms may manifest secondary to biomechanical stress at the site of fusion. The purpose of this report is to describe 103 cases of carpal synostosis and provide a review of the etiology, classification, and clinical significance of intercarpal fusion.

Methods Retrospective review of medical and radiographic records was conducted on all patients with identified carpal coalitions, between 2008 and 2012, at our institution. Demographic and historical data regarding trauma, infection, documented joint disease, and prior wrist symptomatology were analyzed for each case, and radiographic evaluation of carpal alignment was performed.

Results A total of 103 cases of carpal coalition, among 85 individuals, were reviewed. All cases presented asymptotically with the highest proportion noted among individuals of African-Caribbean descent. The most frequent variant in our study population was lunate–triquetral coalition, which was present in 92 of the 103 identified cases. None of the

cases evaluated in this study demonstrated clinical or radiographic evidence of carpal mal-alignment or instability.

Conclusion The results of our investigation demonstrate that carpal coalition presented most frequently among individuals of African-Caribbean descent. While the prevalence of this condition has yet to be studied specifically in Hispanic and African-Caribbean populations, our findings may reflect the genetic linkage between these and certain West African populations. To our knowledge, this represents the largest clinical series of carpal coalition in the English literature.

Keywords Carpal coalition · Carpus · Congenital anomaly · Intercarpal fusion

Introduction

Carpal coalitions are relatively common anatomical variants present in approximately 0.1 % of the population and are characterized by the anomalous union of two or more carpal bones [13]. Two primary forms of carpal coalition exist: osseous coalition (synostosis) and non-osseous coalition [1]. In osseous coalition, the carpals are united as a single osseous block; whereas, in non-osseous coalition the affected carpals are united either by cartilage (synchondrosis), fibrous tissue (syndesmosis), or some combination of the two [1]. Although these distinct entities are frequently referred to as complete and incomplete coalitions, respectively [8], osseous and non-osseous may be preferable as they more accurately describe the nature of the union [1].

In general, coalition of the carpus may represent a congenital or an acquired anomaly seen in otherwise healthy individuals. The former most often presents as an isolated entity, however, may occur as part of a syndrome or in conjunction with known metabolic disorders [34]. Isolated fusions typically involve two bones within the same row, while syndromic-associated fusions are quite often multiple

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in nature [34, 38]. Though this condition has been reported in nearly every possible combination [23, 24], carpals located in the ulnar region, occupying the same row, are most frequently involved [13]. Furthermore, intercarpal fusion is genetically transmissible—demonstrating a non-sex linked, Mandelian dominant pattern of inheritance [17].

In the present report, the authors describe 103 cases of asymptomatic carpal synostosis and provide a review of the literature surrounding the etiology, prevalence, classification, and clinical significance of intercarpal fusion. To our knowledge, this represents the largest clinical series of carpal coalition in the English literature.

Materials and Methods

Approval for the study was granted by the University of Miami Institutional Review Board prior to study commencement. Retrospective analysis of medical and radiographic records was conducted on all patients with identified carpal coalitions, during the period from 2008 to 2012. All patients presented to either the Jackson Memorial Hospital emergency department or hand clinic with histories or injuries that warranted radiographic evaluation of the wrist, hand, or distal upper extremity. A total of 91 patients were identified; however, reliable radiographic and historical data was accessible for only 85 patients in this series. When available, radiologic images of the contralateral wrist were reviewed for purposes of comparison.

Radiographs reviewed in this study were most often obtained to evaluate or rule out acute injuries in patients who sustained falls or were involved in motor vehicle accidents, physical altercations, or sports-related trauma. Common presenting injuries included simple, comminuted, and compound fractures of the distal radius and ulna; metacarpal fractures; phalangeal dislocations; and crush injuries of the forearm or hand. In some circumstances, follow-up images were obtained in patients who sustained previous injuries in order to monitor healing; confirm the proper position of pins, screws, plates and other hardware devices used to treat these injuries; or to evaluate localized joint pain or deformities in patients who sustained digital trauma.

Patient-related factors analyzed in this study included: age, sex, race, history of trauma to the wrist, specific type of coalition (s) identified, and whether findings were unilateral or bilateral. Identified coalitions were recorded as unilateral or bilateral only if radiographs of both wrists were available for review. In situations where radiographs of only one wrist were obtained, the status of the patient's laterality was documented as unknown. When unilateral coalitions were identified, the side of the involved wrist was recorded. All radiographs containing identified carpal coalitions were reviewed and classified separately by two hand specialists. Discrepancies in classification were evaluated and clarified by the senior author. All information regarding the

presence or absence of wrist pain and related symptoms, prior to presentation, was elicited from either the medical interview or an in-depth review of patient records.

Results

Review of plain films of the wrist, for patients with identified coalition deformities, was conducted through the Picture Archiving and Registration System (PACS) at Jackson Memorial Hospital in Miami, Florida. Data collected by a hand specialist over the past 4 years, revealed 103 cases of intercarpal fusion among 85 individuals. Of the cases identified, 53 were male and 32 were female between the ages of 6 and 87 years (median 40.6 years). Ethnicity was determined by self-identification information gathered from electronic medical records. Seventy-one of the 85 patients identified themselves as of Afro-Caribbean descent (83.5 %), 13 as non-black Hispanic (15.3 %), and one as Caucasian (1.2 %). None of the patients endorsed a history of previous trauma, infection, or dysfunction of the wrist, and clinical symptoms prior to presentation were subsequently absent in all affected individuals. Additionally, the majority of presenting injuries or complaints in this series were unrelated to the wrist and/or carpals entirely. As a consequence, each case of carpal coalition was discovered as an incidental finding upon review of radiographic examinations of the forearm, wrist, or hand obtained for unrelated reasons (Table 1).

Of the 85 patients with coalition anomalies, 18 individuals (nine male and nine female) were noted to have bilateral coalition upon review of contralateral radiographs. All but one of the bilateral cases demonstrated identical coalition patterns on either side. The remaining case presented with a Minnaar Type I lunate–triquetral (LT) coalition in the left hand and a Minnaar Type II LT coalition in the right. Additionally, ten cases were confirmed to be unilateral, with the remaining 57 cases documented as unknown due to the absence of contralateral plain

Table 1 Descriptive characteristics of the study population

Time of study (months)	48
Total patients	85
Total cases	103
Age (years)	
Mean	40.6
Range	6 to 87
Gender	
Male	53
Female	32
Race (%)	
Black	71 (83.5)
Hispanic	13 (15.3)
Caucasian	1 (1.2)

films available for comparison. Among the confirmed unilateral cases, the frequency of distribution between the right and left wrist was equal. None of the radiographs in this study demonstrated evidence of carpal mal-alignment, arthrosis, or intercarpal diastasis. Table 2 summarizes the clinical characteristics of the identified cases of carpal coalition.

The most common coalition pattern observed in this study was that of the LT variety, which was present in 92 of the 103 identified cases (89.3 %) (Fig. 1). Additionally, there were four cases of capitate–hamate (CH) coalition (3.9 %), three cases of capitate–trapezoid (CT) coalition (2.9 %), and two cases each of scaphoid–trapezium (ST) and scaphoid–lunate (SL) coalition (1.9 %) (Figs. 2, 3, 4 and 5). Further breakdown of the LT coalitions revealed that two cases were Minnaar Classification Type I (Fig. 1a), while 20 cases were Minnaar Type II (Fig. 1b), 69 cases were Minnaar Type III (Fig. 1c), and one case was Minnaar Type IV (Fig. 1d). Hand dominance was noted but was not considered relevant to the patients' condition. Due to the retrospective nature of this investigation, evaluation of uninjured wrists containing a coalition in order to assess for pain or functional limitation was not always possible.

Discussion

Etiology of Carpal Coalition

Carpal coalition represents a congenital or acquired anomaly with the former resulting from failure of differentiation

during embryonic development [22]. The use of the term “fusion” to describe this abnormality should be avoided, as the process that actually occurs is incomplete demarcation of cartilaginous precursors rather than the union of two previously distinct structures [4, 12, 20, 28]. Alternatively, carpal coalition may result from multiple inflammatory arthropathies including rheumatoid arthritis, juvenile arthritis, psoriatic arthritis, and Reiter's syndrome among others [34]. Metaplastic conversion of mesodermal derivatives such as fibrous, cartilaginous, and ligamentous tissue to bone may also lead to intercarpal fusion [34, 38]. In fact, union of the pisiform with either the triquetrum or hamate is accomplished via this mechanism [1, 4, 5]. Trauma-associated fusion may reflect primary injury to the carpus, or alternatively, surgical arthrodesis performed to maintain stability and partial mobility of the joint. Upon review of relevant records, none of our patients reported a history of arthritis, trauma, or previous surgical fusion of the wrist. Thus, the majority of our cases likely reflect congenital malformations in carpal development.

Prevalence of Coalition Anomalies of the Hand

A wide geographic variation in the prevalence of intercarpal fusion has been emphasized in numerous reports [4, 13, 24, 27, 29, 36]. Coalition between the lunate and triquetrum is the most common variant [4, 6, 13, 23, 32]. Among the various populations studied, the highest prevalence of this anomaly is seen in individuals of West African descent, with reports as high as 9.5 % in some groups [4, 13, 32, 33]. In

Table 2 Identified cases of carpal coalition

	LT Minnaar Type I	LT Minnaar Type II	LT Minnaar Type III	LT Minnaar Type IV	CH	CT	ST	SL	Totals
Number of cases	2	20	69	1 ^a	4	3	2	2	103
Laterality/affected Side	–	6	26	–	–	2	–	–	34
Bilateral (identical)				–	–	–			
Unilateral		2 ^b	2				1	1	6
Right	1 ^b		4				1		6
Left							–		
Unknown	1	4	13	1	2			1	22
Right		8	24		2	1			35
Left									
Previous trauma	No	No	No	No	No	No	No	No	–
Prior disease/infection	No	No	No	No	No	No	No	No	–
Prior symptoms	None	None	None	None	None	None	None	None	–

LT lunate–triquetrum, CH capitate–hamate, CT capitate–trapezoid, ST scaphoid–trapezium, SL scaphoid–lunate

^a Minnaar Type IV deformity involving complete osseous coalition between the lunate and triquetrum along with bilateral capitate–hamate coalition

^b Patient with bilateral coalition deformities involving a left-sided Minnaar Type I non-osseous coalition and a right-sided Minnaar Type II osseous coalition with a distal notch

Fig. 1 Lunate–triquetral coalition. **a** Posteroanterior radiograph of the left wrist demonstrating incomplete coalition at the proximal aspect of the LT joint space (*arrow*) (Minnaar Type 1). **b** Posteroanterior radiograph of the right wrist showing proximal coalition between the lunate and triquetrum with a notch (*arrow*) in the distal border of the osseous bridge (Minnaar Type 2). **c** Posteroanterior radiograph of the right wrist revealing complete osseous coalition between the LT bones (Minnaar Type 3). **d** Posteroanterior radiograph of the right wrist in a patient with complete osseous LT coalition accompanied by capitate–hamate coalition (Minnaar Type IV)

the American population, the prevalence ranges between 0.08 % and 0.13 % [37], with an estimated frequency of 0.1 % among white Americans and 1.6 % among Americans of African origin [13].

Second in frequency is coalition between the capitate and hamate, which demonstrates similar variations in prevalence. The reported prevalence in East Africans (0.14 %) and Caucasians (0.25–0.29 %) is known to be lower than in those of West African descent (0.4–0.8 %) [2, 6, 17, 23]. Additionally, pisiform–hamate (PH) coalition is believed to occur most often in African populations, with frequencies ranging from 0.11 % to 0.76 % [1, 5]. Other, less common, forms of carpal coalition have been reported; however, their prevalence has not been the subject of extensive research.

The results of our investigation demonstrate that carpal coalition presented most frequently, in our sample, among

individuals of Afro-Caribbean (principally Haitian) descent (83.5 %). While the prevalence of this condition has yet to be studied specifically in Hispanic and Afro-Caribbean populations, our findings may reflect the genetic link between these and certain West African populations, thereby providing potential insight into the migration patterns of these ethnic groups. LT coalition comprised nearly 90 % of the identified cases in our sample. CH coalition was the next most common, occurring 23 times less frequently. We suspect that the wide variation in prevalence among studies is attributable to differences in diagnostic ability. Non-osseous coalitions are less likely to be identified by radiography compared to skeletal analysis or physical dissection, bringing to question the validity and relevance of prevalences calculated via this method [1, 7, 36]. By the same reasoning, the nature of this investigation

Fig. 2 Posteroanterior view of the left wrist demonstrating complete osseous coalition between the capitate and hamate bones

Fig. 3 Standard posteroanterior radiograph of the left wrist in a 24-year-old Haitian female revealing osseous coalition between the capitate and trapezoid bones

Fig. 4 Posteroanterior plain film of the wrist demonstrating left-sided osseous scaphoid–trapezial coalition

does not permit accurate determination of prevalence, as the occurrence of non-osseous coalition is likely much higher than that suggested by radiologic studies.

Classification Systems Based on Radiographic Appearance

Currently, there is no standardized classification system that encompasses all potential variants of intercarpal fusion. In 1952, De Villiers Minnaar [8] proposed a four-type classification scheme for LT coalition based on radiographic assessment (Table 3). This system has since served as the most widely used classification device for carpal coalition in general. However, its application to all forms of carpal synostosis is problematic in that it neglects the association between intercarpal fusion and the presence of co-existing anomalies throughout the skeleton [39]. Furthermore, Minnaar's system only addresses variations in osseous coalition (Minnaar Type II and III), compiling the considerably more extensive variation associated with non-osseous coalition into a single discrete category (Minnaar Type I) [1].

To address these shortcomings, Singh et al. [34] proposed a more comprehensive classification system based upon

Fig. 5 Posteroanterior view of the right wrist showing osseous coalition between the scaphoid and lunate bones in a 71-year-old African American male

Table 3 Comparison of the Minnaar classification with Sign's classification and Burnett's carpal coalition dichotomy

Author	Year	Classification scheme
Minnaar	1952	Type I: Proximal pseudarthrosis
		Type II: Proximal osseous bridge with distal notch
		Type III: Complete fusion
		Type IV: Complete fusion with other carpal anomalies
Singh et al.	2003	Type I: Shape of fused bones
		(a) Retaining shape
		(b) Distorted shape
		Type II: Site of fusion
		(a) Palmar fusion
		(b) Dorsal fusion
		(c) Central fusion
		(d) Proximal fusion
		(e) Distal fusion
		(f) Intermediate fusion
Burnett	2011	(g) Complete fusion
		Type III: Apparent fusion
		Type IV: Non-osseous fusion
		Osseous coalition
		Non-osseous coalition

morphology and architecture of the resultant fused bones (Table 3). Like the Minnaar Type I variant, however, this system is flawed in that it does not address variations associated with non-osseous coalition. Burnett [1] classified coalition anomalies as falling into one of two discrete categories: osseous versus non-osseous (Table 3). This simplified terminology captures the two major variations in appearance, which are likely to be associated with differences in clinical significance [1, 3, 35]. Additionally, it avoids the inconsistent and narrow scope of associated anomalies described by Minnaar's scheme.

Syndromic Manifestations of Carpal Coalition

Upon thorough review, none of the cases presented in this report manifested in association with syndromic or developmental abnormalities. However, the association of carpal coalition with a number of malformation syndromes is well documented in the literature (Table 4).

Anthropological Significance

The anthropological significance of this anomaly has been the subject of longstanding debate [35]. Since coalition of the carpus is almost always an incidental finding, it may have anthropologic importance — possibly representing an

Table 4 Carpal coalitions in congenital malformation syndromes

Condition	Reported coalition deformities
Ellis van Creveld	Capitate–hamate (most common); massive fusions involving multiple carpals [9]
Osteochondritis dissecans	Capitate–trapezium [15]
Holt Oram syndrome	Abnormally shaped scaphoid fused to other carpals; narrowing of joint space between scaphoid and trapezium; presence of accessory carpal bones noted [27]
Oto palatal digital syndrome	Fusion of scaphoid with other carpal bones; various forms of intercarpal union; comma shaped trapezoid and transverse position of capitate [27]
Fetal alcohol syndrome	Capitate–hamate [18]
Brachydactyly	Capitate–hamate; unspecified or miscellaneous coalitions [2]
Turner’s syndrome	Lunate–triquetrum; abnormal shaped proximal row carpals [10]
Liebenberg syndrome	Triquetrum–pisiform; abnormal shape and size of various carpals [21]
Banki syndrome	Lunate–triquetrum [2]
Ulnar and Fibular Dimelia	Capitate–hamate [31]
Diastrophic dwarfism	Deformed carpal bones; narrowing of joint space with unspecified carpal coalition is common [27]
Dyschondrosteosis	Lunate–triquetrum (described in Madelung’s deformity) [27]
Arthrogryposis multiplex congenita	Variable carpal and tarsal coalition deformities; more often acquired evidenced by presence of remnant joint space in dissected specimens [25]
Symphalangism	Triquetrum–hamate (most common); diverse patterns of carpal and tarsal coalitions have been reported [16]
Hand–foot–uterus syndrome	Scaphoid–trapezium; deformity of the scaphoid is often evident [27]
Nievergelt’s syndrome	Massive bilateral carpal and tarsal coalition [25]
Mitral insufficiency, conductive deafness, fusion of cervical vertebra	Capitate–hamate, Lunate–Triquetrum, Scaphoid–Trapezium; tarsal fusions also noted [11]

advance toward specialization of the hand or an attempt to stabilize the postaxial border of the hand — as suggested by De Villiers Minnaar in 1952 [8]. This concept of adaptability has been perpetuated by the theory that fusion of the scaphoid with the os centrale in humans and African apes, as compared to monkeys, imparts stability for pinch grip or knuckle walking [19]. However, the theory that carpal fusion represents an atavistic trait is unsupported by the hominid fossil record and is not consistent with comparative anatomic studies of past and present primate wrist anatomy [19]. Furthermore, the increased susceptibility of the coalesced wrist to fracture upon subjection to physical stress negates the likelihood that coalition serves to strengthen the carpus [2]. Thus, knowledge of the functional advantage offered by carpal coalition demands an enhanced understanding of carpal kinematics which is required to thoroughly grasp the evolutionary implications, if any, of this condition [30].

Clinical Significance and Management of Symptomatic of Intercarpal Fusion

As demonstrated by the cases presented, carpal coalition is largely an asymptomatic condition discovered incidentally on radiographs taken for unrelated reasons. Neither osseous nor non-osseous coalition appears to have a discernible

effect on wrist function [8, 37]. However, symptoms may manifest by virtue of the biomechanical alterations that result from structural fusion at the wrist [34]. Loss of movement between the fused bones and a compensatory increase in motion at surrounding joints, theoretically predisposes individuals to recurrent sprains and pain under conditions of excessive physical stress [20]. This may be particularly true for non-osseous coalitions, in which deficient intra-articular cartilage formation may lead to a clinical and anatomical state resembling degenerative arthritis [14]. Furthermore, persistence of grooves, notches, and cavities at the fusion site appears to be a contributing factor toward the predisposition to fractures in patients with osseous coalition [3, 34, 35].

Asymptomatic or mildly symptomatic cases of coalition rarely require anything more than conservative management. However, in cases where the severity and duration of pain warrants surgical intervention, limited wrist arthrodesis, achieved through compression of the involved bones, has demonstrated good reliability with acceptable effects on postoperative wrist motion [14, 29, 33]. Postoperative immobilization, until radiographic confirmation of fusion, is necessary for successful recovery and may take between 2 and 5 months to achieve [26]. Furthermore, extensive postoperative rehabilitation is essential to ensure optimal recovery of wrist motion and functional grip strength.

In our review, all cases of carpal synostosis presented asymptotically with the highest proportion of cases noted among individuals of Afro-Caribbean (most commonly Haitian) descent. The most frequently isolated variant in our study population was that of lunate–triquetral coalition, a finding consistent with the majority of previously published accounts. While a thorough understanding of carpal kinematics has yet to be fully developed, further investigation of patients with carpal coalition, utilizing advanced dynamic imaging, could prove valuable in elucidating our understanding of functional wrist anatomy and carpal biomechanics. Although beyond the scope of this investigation, in general, information derived from these studies may generate improvements in the management of patients who present with congenital or acquired carpal dysfunction.

Conflict of interest The authors declare that they have no conflicts of interest, commercial associations, or intent of financial gain regarding this research.

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