

CASE REPORT

A Case Report on Carpal Synostosis in an Eight-Year-Old Male

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Abstract

Introduction: Carpal synostosis is a medical condition in which adjacent carpal bones become fused with or without other associated limb deformities. According to literature, carpal synostosis has a relatively low incidence rate of 0.1%.

Case report: We report a case of an isolated complete lunotriquetral synostosis of an eight-year-old male revealed incidentally upon x-ray examination to rule out fracture of the distal forearm post trauma.

Discussion: There is but little literature of an isolated carpal coalition of a pediatric at the

time of this report. Also, there is no standardized classification system for carpal coalition due to insufficient imaging data by virtue of rarity of the condition and incomprehensive description of the morphology of coalitions, among others. Though patient's lunotriquetral synostosis was not seen to impair wrist function, the radiologist report revealed negative ulnar variance, thus the need for further investigation into its possible association.

Conclusion: This report adds up to existing literature to contribute a better classification system of carpal synostosis. Association between carpal synostosis and ulnar variance should be further explored.

Key Words: *Carpal synostosis; Pediatric; Asymptomatic; Lunotriquetrum; Anatomical variant*

Introduction

Carpal synostosis is a medical condition in which adjacent carpal bones become unionized and ossified with or without other associated limb deformities [1]. It can either be a congenital or acquired anatomical variant [1]. Carpal synostosis begins as an intrauterine fusion of carpal bones due to inability of precursor cartilages to separate followed by subsequent chondrification and ossification during 4th to

8th week of intrauterine life [1-3]. Other literature, however, suggest that carpal coalition might arise from a failure of differentiation since cartilaginous anlagen of more than eight carpal bones are present in early embryonic life [4]. Carpal synostosis, which is one of the two main forms of carpal coalition, distinguishes as an osseous coalition of two or more carpal bones. Non-osseous coalition, on the other hand, involves the union of carpal bones either

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by cartilage (synchondrosis) or fibrous tissue (syndesmosis) [5].

This condition is mostly asymptomatic and is generally detected as an incidental finding during imaging [6]. This explains why Carpal synostosis has a relatively low incidence rate of 0.1 [1,2,7]. It also has more preponderance in females and people of African descent [3]. The most common site of carpal synostosis anomaly is at the lunate-triquetral region, followed by capitate-hamate region; certain types of synostoses have a higher incidence among Africans [2]. Carpal synostosis may present as a solitary condition or be associated with a syndrome. When the coalition is seen as an isolated finding and not part of a syndrome, the proximal carpal row is mostly affected [4]. When the synostosis involves the distal carpal row, the finding may be an isolated one, but there is a greater propensity of a syndromic association [4]. Several syndromes identified include but are not limited to arthrogryposis, diastrophic dwarfism, dyschondrosteosis, Ellis-van Creveld syndrome, symphalangism, turner syndrome, among others [6].

Case Report

An eight-year-old male schoolboy presented with complaints of mild pain and swelling at the right wrist, having been smashed by a door. Conventional x-ray showed normal radiocarpal, intercarpal and carpometacarpal alignment with no narrowing. There were no fractures seen or abnormal bone densities noted. The soft tissues were normal. While most radiographic features were normal, there was a complete lunotriquetral coalition. X-ray image is demonstrated below in Figure 1.

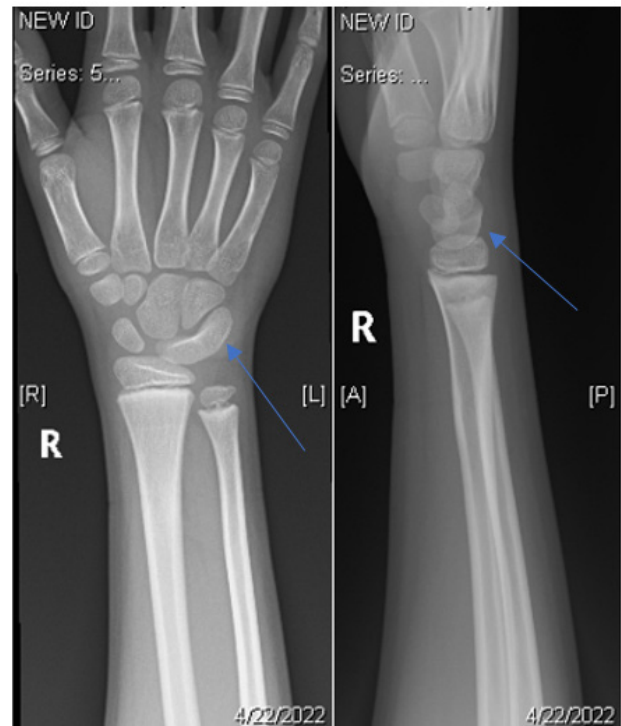


Figure 1) Arrows showing a complete lunotriquetral coalition.

Discussion

There was little literature of an isolated carpal coalition among pediatrics at the time of this report.

Previous cases have been incidental. Spaans & Beumer reported six different retrospective cases of isolated carpal coalitions of patients between the ages of 18-45 years [2]. Kumar *et al.* also reported on multiple carpal coalitions of a 43-year-old [1]. Our pediatric finding reveals how early complete fusions could occur and this makes room for further investigation in subsequent studies on the exact timeline of carpal synostosis.

Although there have been numerous carpal coalition classification systems across literature, none is standardized because each system has its characteristic pitfalls. These pitfalls are due to paucity of imaging records, incomprehensive consideration of the morphologic nature of synostosis, among others [5]. Minnar de Villiers described four categories of carpal coalition (Table 1) [2,8].

TABLE 1
Classification of lunotriquetral coalitions according to Minnaar de Villiers (1952).

Type 1	Incomplete fusion resembling a pseudo-arthritis (fibro-cartilage coalition)
Type 2	Incomplete osseous fusion
Type 3	Complete osseous fusion
Type 4	Complete osseous fusion associated with other carpal anomalies

In effect, our case of lunotriquetral coalition falls within Type 3 according to Minnaar de Villiers classification of lunotriquetral coalitions.

Carpal synostosis is largely asymptomatic but can be complicated following trauma as indicated by De Fazio et al. and Noureldin et al. [3,5]. In line with this, our patient had not expressed any concerns at the area of interest until he was hit by the door. Upon physical assessment, our case revealed only mild post traumatic swelling and pain but no major complications. Though asymptomatic, it is important that there is continuous care provided to look out for any possible futuristic ramifications when this anatomical variant is subjected to trauma. Carpal synostosis poses some critical implication in the approach of care

should there be a fracture or subluxation post trauma [1,3].

Although complete lunotriquetral synostosis is rare, it is the most common form of intercarpal fusion followed by capitate-hamate synostosis [2]. Our finding also reflects that of Rayan & Upton who indicated a much greater chance of proximal intercarpal fusion when there is no associated syndrome or deformity [4].

Finally, there was an impression of negative ulnar variance. While this observation opens further investigation on the relationship between carpal synostosis and ulnar variance, the latter could be due to changes in incident angle of x-ray beam or positioning of the forearm during imaging [9].

Conclusion

Carpal synostosis is rare and mostly asymptomatic. Isolated carpal synostosis, as discussed above, does not impair wrist function. However, its possible association with other anomalies such as negative ulnar variance should be explored. This report could corroborate with existing literature for a standardized classification system carpal synostosis.

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