Asymptomatic Pisiform-Hamate Coalition: A Case Report

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Carpal coalition is a well-documented skeletal anomaly. Of the various coalitions that have been described, the lunate–triquetrum is the most prevalent. The pisiform–hamate coalition, originally reported in the West African Yoruba tribe, is rare. All 6 cases were incidental findings in asymptomatic patients. Recent reports document patients with tenderness on palpation of the coalition and ulnar neuropathy. Here we report the case of an asymptomatic pisiform–hamate coalition and review the literature.

"Recent reports document patients with tenderness on palpation of the coalition and ulnar neuropathy."

Case Report

A right-hand–dominant man in his early 20s presented with right elbow pain after falling onto his right upper extremity. He had no complaints of pain or decreased range of motion (ROM) in the right wrist. Physical examination of the extremity revealed tenderness to palpation at the radial head without tenderness or decreased ROM at the wrist. He had full ROM in all his digits with no deficit in ulnar function. Radiographic evaluation of the right upper extremity revealed an incidental, asymptomatic pisiform–hamate (PH) coalition (Figures 1, 2), but contralateral films did not reveal any evidence of the coalition on the right side. The patient was treated with a posterior elbow splint for comfort with early elbow ROM. He was still asymptomatic in the right wrist at follow-up.

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Literature Review

Carpal coalition has been linked to rare skeletal syndromes. In 1993, a unique orthopedic syndrome, yet to be named, was reported in native Puerto Rican children.[1] Twenty-three children had a complex of bilaterally dislocated hips, short stature, and dislocated radial heads. Twenty of these children also demonstrated carpal coalition, with capitate–hamate (CH) coalition being the most common. Only 1 child experienced pain or functional limitation at the wrist.

Another recently described syndrome is congenital synspondylism, characterized by dysplastic skeletal anomalies that include vertebral fusion and carpal coalition.[2,3] Five of the 6 reported cases in the literature involve some element of consanguinity. Patients were found to have CH coalition, lunate–triquetrum (LT) coalition, or both. More than 80% of these coalitions were bilateral.

Carpal coalition was also documented in the syndrome arthrogryosis multiplex congenita in 6 patients (5 males, 1 female).[4] Radiographic diagnosis of carpal coalition was made between age 15 and age 31. Five of the 6 cases had complete fusion of all carpal bones.

Although carpal coalition has been linked to various syndromes, there have been isolated cases of carpal coalition, LT coalition being the most common.[5] Since 1959,

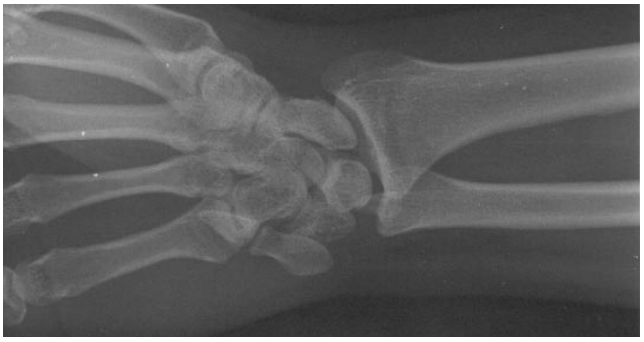

Figure 1. Anteroposterior plain film of the right wrist.

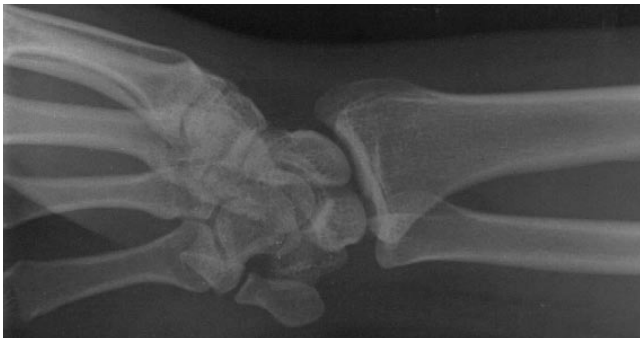

Figure 2. Oblique plain film of the right wrist.

Table. Summary of Documented Cases of Pisiform–Hamate Coalition

Case(s)	Report	Age (y)	Race	Sex	Hand(s)	Dominance	Bilateral	Symptoms	Occupation	Treatment
1-6	Cockshott[6]	?	Black	5M, 1F	Both	?	Yes	Asymptomatic	Yoruba tribe	Untreated
7	Ganos & Imbriglia[8]	23	White	M	R	R	No	Pain	Professional guitarist	Excision
8		33	White	M	R	R	No	Pain	Factory worker	Excision
9	Berkowitz et al[9]	14	White	M	L	R	No†	Pain, stiffness, paresthesia	Student-pianist	Excision
10		14	White	M	L	R	Yes	L (pain, paresthesia), R (asymptomatic)	Student	Excision
11	El-Morshidy et al[7]	30	Indian	M	Both	R	Yes	Asymptomatic	Laborer	Untreated

*M indicates male; F, female; R, right, L, left. †Authors' interpretation of information presented.

there have been only 4 published reports of 11 patients with PH coalition.[6-9] Most cases were asymptomatic and not associated with other skeletal anomalies. Symptomatic patients complained of hypothenar pain, stiffness, and ulnar paresthesia.

There is no classification for PH coalition, but in 1952 Minaar[10] classified LT coalition into 4 variants: type I, a proximal pseudoarthrosis between the lunate and triquetrum; type II, a proximal osseous bridge with a distal notch; type III, a complete fusion of the lunate and the triquetrum; and type IV, complete fusion, in association with other carpal anomalies.

PH coalition was first reported in 1959, when Cockshott[6] (Table) described 6 asymptomatic cases in the Western Nigerian tribe of Yoruba. The first and second cases were incidental findings of an anthropological survey of more than 1300 adults. Two of the 6 patients were siblings. Likewise, El-Morshidy and colleagues[7] described a right-hand–dominant 30-year-old man with asymptomatic PH coalition. The patient presented after a fall in which he sustained fractures of the right humerus, fractures of the right radial and ulnar shafts, and a palmar dislocation of the right scaphoid. On further evaluation, he was found to have bilateral, asymptomatic PH coalitions.

Ganos and Imbriglia[8] described 2 right-hand–dominant male patients with symptomatic PH coalition. These patients presented with recurrent right hypothenar eminence pain. Both used their hands extensively—one was a professional guitarist, the other a factory worker. Conservative management and steroid/lidocaine injections failed to relieve their pain, and eventually the pisiform was excised to provide relief. Surgical exploration revealed proximal pseudoarthrosis between the pisiform and the hamate (guitarist) and complete fusion of the pisiform and hamate (factory worker).

Berkowitz and colleagues[9] reported on 2 right-hand–dominant male patients with pain and ulnar neuropathy associated with PH coalition. One patient, an avid pianist, complained of left ulnar-sided wrist pain and persistent paresthesias involving the fourth and fifth fingers. On physical examination, he had a positive Tinel sign over the Guyon canal and a positive Froment sign. Surgical exploration of the area revealed an osseous extension between the pisiform and the hamate. Excision of the pisiform decompressed the ulnar nerve and relieved symptoms. The other patient presented with pain at the base of the left hypothenar eminence and paresthesias in the fifth finger. Examination of both wrists demonstrated fullness at the base of the hypothenar eminence with tenderness only in the left hand. Plain films and computed tomography scans showed bilateral PH coalitions, one incomplete (left hand) and one complete (right hand). The ulnar nerve was thoroughly decompressed with excision of the pisiform.

Mutations in 2 different genes have been reported as causing syndromes involving carpal coalitions. Graham and colleagues[11] described a Pro250Arg point mutation in the fibroblast growth factor receptor 3 (FGFR3) gene. FGFR3 is responsible for the syndrome "coronal craniosynostosis with brachydactyly and carpal/tarsal coalition." The carpal coalition most commonly involved in this syndrome is CH coalition.

Dixon and colleagues[12] reported that 3 different mutations in the Noggin (NOG) gene may be responsible for carpal or tarsal coalitions. The normal NOG gene encodes for a protein that negatively regulates the activity of bone morphogenetic protein (BMP). A mutation in the NOG gene is thought to increase BMP activity with excess cartilage formation and osseous synostoses.

Conclusions

We have presented a case of asymptomatic PH coalition and a review of associated syndromes, previously documented PH coalitions, and different theorized causes of carpal coalition. Asymptomatic PH coalition should remain untreated, and symptomatic PH coalition be treated with resection of the pisiform.

Author's Disclosure Statement and Acknowledgement

The authors report no actual or potential conflict of interest in relation to this article.

References

1. Steel HH, Piston RW, Clancy M, Betz RR. A syndrome of dislocated hips and radial heads, carpal coalition, and short stature in Puerto Rican children. *J Bone Joint Surg Am.* 1993;75:259-264.
2. Wiles CR, Taylor TF, Sillence DO. Congenital synspondylism. *Am J Med Genet.* 1992;42:288-295.
3. Lalonde F, Letts M, Davidson D. Congenital synspondylism: diag-

nostic features. *J Pediatr Orthop.* 1999;19:613-616.
4. Orlin H, Alpert M. Carpal coalition in arthrogryposis multiplex congenita. *Br J Radiol.* 1967;40:220-222.
5. Gross SC, Watson HK, Strickland JW, Palmer AK, Brenner LH. Triquetral–lunate arthritis secondary to synostosis. *J Hand Surg Am.* 1989;14:95-102.
6. Cockshott WP. Pisiform hamate fusion. *J Bone Joint Surg Am.* 1969;51:778-780.
7. El-Morshidy AF, Rabia F, Mukaimi A. Bilateral asymptomatic pisiform and hamate coalition—a case report. *Hand Surg.* 2000;5:57-60.
8. Ganos DL, Imbriglia JE. Symptomatic congenital coalition of the pisiform and hamate. *J Hand Surg Am.* 1991;16:646-650.
9. Berkowitz AR, Melone CP Jr, Belsky MR. Pisiform–hamate coalition with ulnar neuropathy. *J Hand Surg Am. 1*992;17:657-662.
10. Minaar AB deV. Congenital fusion of the lunate and triquetral bones in the South African Bantu. *J Bone Joint Surg Br.* 1952;34:45-58.
11. Graham JM Jr, Braddock SR, Mortier GR, Lachman R, Van Dop C, Jabs EW. Syndrome of coronal craniosynostosis with brachydactyly and carpal/tarsal coalition due to Pro250Arg mutation in FGFR3 gene. *Am J Med Genet.* 1998;77:322-329.
12. Dixon ME, Armstrong P, Stevens DB, Bamshad M. Identical mutations in NOG can cause either tarsal/carpal coalition syndrome or proximal symphalangism. *Genet Med.* 2001;3:349-353.

This paper will be judged for the Resident Writer's Award.