

Case Report

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Carpal coalition revisited an a propos of 80 cases of LT coalition and a skewed literature view***Corresponding Author: Christian Weinand**

Osnabrueck Klinikum, Section of Plastic, Hand, Reconstructive and Burn Surgery, University of Witten/Herdecke, Germany.
Email: chwscot@yahoo.com

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Abstract

Carpal coalition is a rare congenital entity, the Luno-Triquetral (LT) coalition having the highest incidence. Additionally, LT coalition is described in literature to be most often bilateral and being more common in females. The range of LT coalitions is wide, starting from simple over fibrocartilage coalition to complete osseus fusions. Classification of Minnaar is the one being most often used. Little research is available on the difference of incidence between differences in the type of LT coalition comparing right- and left-hand side.

The unilateral coalition in men is according to literature a rare entity. We describe the case of a unilateral right sided LT coalition in a male Caucasian patient, discovered incidentally after a sustained distal radius fracture. Additionally, we reviewed literature for incidence of the LT coalition, gender distribution, side of incidence, LT coalition type and difference in Minnaar type of coalition from one hand side to the other hand side.

LT coalition in men and women was reported equally often. Unilateral and bilateral LT coalition incidence and incidence between right- and left-hand side were equal. LT coalition type III was most often in both sexes right side more than left in women. Incidence of Minnaar manifestations in LT coalition was for men in right and left side unilateral and bilateral equal, whereas women had unilateral right and left equal incidence but bilateral higher Minnaar classification more often on the left side. Type I and type III (Minnaar) was in man and women most often in unilateral and bilateral incidence. Equal Minnaar classification of LT coalition in bilateral appearance was most often, however Minnaar classification being higher one type and even two types was most often in women and there right more than left-hand side.

In contrast to literature, we found equal incidence of LT coalition in both sexes, left and right-hand side and equal incidence in unilateral and bilateral LT coalition but higher incidence in higher Minnaar classification right over left hand in women. Further research is warranted on difference in manifestation of LT coalition in bilateral incidence between left and right-hand side.

Keywords: Carpal coalition; Os lunotriquetrum; Unilateral vs bilateral.

Introduction

Carpal coalition is an autosomal dominant inherited condition, African population is described to be having the highest number in incidence [1-4]. The side most often affected in literature is the left, often a combination with other congenital differences is found [4]. The most common coalition is of the Luno-Triquetral (LT) coalition (90%) followed by the capito-hamate coalition [1,2].

LT coalition is described to be more often bilateral and to be more common in women in a ratio of 2:1 [1-4]. While osseous coalitions of the lunate and the triquetrum are known to be asymptomatic, fibrocartilage lunate-triquetral coalitions can present as an uncommon cause for ulnar-sided wrist pain [3]. However, the LT coalition is most often discovered by incident [5]. According to this, unilateral LT coalition in a man is a rare incident, however, there is little research on comparing unilateral to bilateral LT coalition incidence. There are several classification systems of the Luno-Triquetral (LT) coalition, such as the classification of De Villiers Minnaar, who developed his classification system according to radiographical appearances, i.e. the classification is more focused on bone focused than non-osseous types of coalition [1,2,5,6]. In our case report and review of literature we adhere to the Minnaar classification, because this is the most known and most widely used system of classification for LT coalition.

Most LT coalitions are described to be bilateral, and, if differing in classification from one side to the other, then their difference is only in one of the Minnaar classification types to the other side [6-8]. However, there is little research on the difference in expression of coalition comparing one hand side to the other.

We present a case of a male patient with unilateral LT coalition, comparing this to incidence described in literature. In addition, we research literature for incident of LT coalition in gender, type of coalition according to Minnaar classification, side of incident, incidence of LT coalition unilaterally and bilateral, as well as difference in LT coalition classification in bilateral cases between sides.

Case

A 58-year-old male patient reported to our outpatient clinic in 2008 after having sustained a bilateral distal radius fracture after a fall at work. He was immediately treated by reposition and plates. On radiological examination a unilateral LT coalition Minnaar type II on the right side was diagnosed. Because of postoperative mal-alignment in the palmar inclination of the right sided distal radius joint line a revision was done, the plate was exchanged and the distal radial joint line was corrected into anatomic position. Healing after that was uneventful, both fractures healed in an acceptable position. The patient received physical therapy for improving wrist motion. In 2009 the plates were removed. The patient reported an increasing pain in his right wrist. A CT scan of his right wrist showed a LT coalition Minnaar type II as well as arthritic changes in the entire wrist and distal radio-ulnar joint. He changed his job due to pain from the arthritic changes for a job with less strenuous activity. On clinical examination the right wrist had a plumper appearance than the left, extension and flexion were on both sides dimin-

ished to 70% of normal range of motion. On radiological examination of the image of the right wrist revealed the known LT coalition Minnaar type II but also showed signs of arthritis and a healed distal radius fracture (Figures 1 and 2). The left sided wrist showed no LT coalition but signs of a healed distal radius fracture.

We researched literature for LT coalition reports and case series. Only literature with at least three aspects of our research clearly stated were included, such as gender, unilateral or bilateral occurrence, side of occurrence, or Minnaar type of LT coalition.

Results literature

86 cases in literature were described in that way so we could include them into our evaluation (Tables 1-3). Equal number of cases of LT coalition were described in male and in female patients, incidence of LT coalition was equal for unilateral and bilateral occurrence (Table 3). In the unilateral cases occurrence in gender was equal, however a higher number of cases was noted to be on the right side. Interestingly, women and men had higher numbers in Minnaar type I and III occurrence (Tables 3 and 4).

When comparing bilateral appearance, men had a higher number in occurrence than women. Most often equal type in classification according to Minnaar was found on both hand sides (50%), type I and III of Minnaar classification were described equally often for the right and left side in men and type I and III in women. Interestingly Minnaar type IV only occurred once in our literature and was described in a man bilaterally but not in female patients (Tables 4 and 5).

The largest number of Minnaar type I and III was on the right side for women, in men, however, type I was noted to be more often on the left side and Minnaar type III equal in number on both sides. Both sexes presented most often with type III, unilateral and bilateral numbers combined (Tables 4 and 5).

In bilateral occurrences on the right side a higher type of Minnaar classification (40%) was noted than on the left side, in 10% of the cases the left side had a higher type of Minnaar classification than the right side. Interestingly, in 25% of the cases the Minnaar classification was two types higher on the right than on the left side. This did not occur vice versa for the left side to be two types higher than the right side. Three types higher or more differences in Minnaar classification between each hand sides were not described in our literature (Table 6).

Discussion

The first case of LT coalition in mankind found was described by Oberlin and Sakka in a neanderthal man [2,4].

LT coalition has the highest incidence of all carpal coalitions possible [1,2,4,8-10]. Some authors state a lack in differentiation during development in the fetal hand as reason for LT coalition [5,10-14], however, the cause is being to be discovered. The development of the joint space happens during the 4th and the 8th intrauterine week. Here four theories are employed: absence of chondrification of the intermediate zone, destruction of the cartilage during ossification, fusion of an additional bone (os epipyramis) or inflammation causing the coalition of lunate and triquetrum [2,5,10,11,15,16].

Table 1: Literature describing cases of bilateral LT coalition.

Literature	Sex	Number of cases (total)	Bilateral (Number of cases)	Right Side	Number of cases side	Minnaar Classification	Left Side	Number of cases side	Minnaar Classification
1	F	1	1	x	1	II	X	1	I
2	?		1	X	1	II	x	1	I
4	14M 6F	32	5			III			III
6	F	1	1	X	1	I	X	1	I
6	M	1	1	X	1	I	X	1	I
6	M	1	1	X	1	I	X	1	I
6	F	1	1	X	1	III	X	1	I
6	F	1	1	X	1	I	X	1	II
7	M	4	1	x	1	III	X	1	III
7	M		1				X	1	III
7	M		1	X	1	II	?	?	?
8	M	1	1	X	1	III	X	1	I
10	F	1	1	X	1	III	X	1	II
13	M	1	1	X	1	III	X	1	III
14	M	1	1	x	1	IV	x	1	IV
17	2F M	9	3	X	1	III	x	1	I
17	2F		2	x	2	III	x	2	III
18	M	2	1	X	1	II	X	1	III
19	M	1	1	X	1	III	X	1	III
20	M	1	1	x	1	II	x	1	I

Table 2: Literature describing cases of unilateral LT coalition.

Literature	Sex	Number of cases (total)	Unilateral (Number of cases)	Right Side	Number of cases side	Minnaar Classification	Left Side	Number of cases side	Minnaar Classification
2	?		1				X	1	I
3	F	1	1	x	1	II			
4	14M 6F		5			III			
4	14M 6F		2						II
4	14M 6F		1						I
5	F	1	1				x	1	II
6	M	1	1				X	1	I
6	F	1	1	X	1	I			
6	F	1	1	X	1	I			
6	F	1	1	X	1	I			
7	M		1	X	1	III			
9	M	1	1	X	1	I			
11	F	1	1				X	1	I
11	M	1	1				X	1	III
12	F	1	1				X	1	I
15	M	1	1				x	1	I
15	M	1	1				x	1	I
16	M	1	1	X	1	III			
17	2F		2	x	2	III	x	2	III
17	M 2F		3				x	3	I III
17	M 2F		3				x	3	I III
17	2F		2	x	2	III			
17	M		1				X	1	III
17	2F		2	x	2	III			
17	M		1				X	1	III
18	F		1	X	1	III			

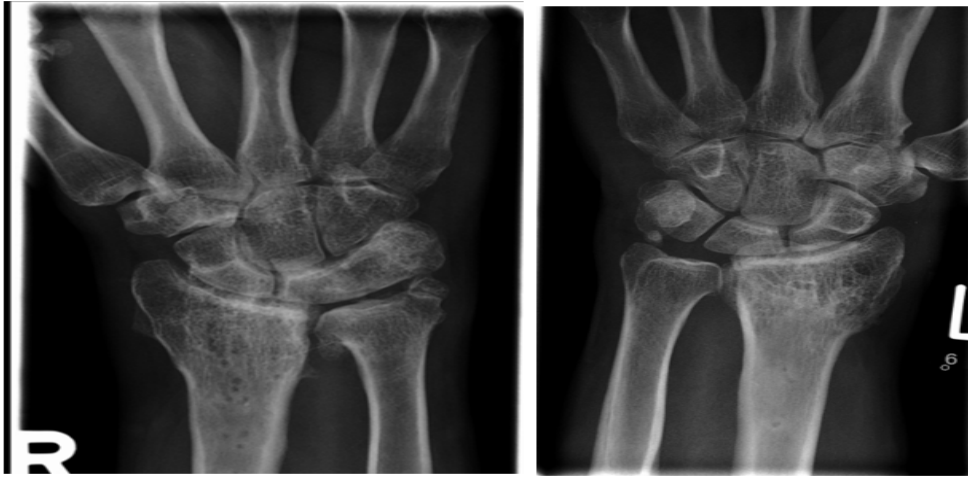


Figure 1: The patient showing healed distal radial fractures and the incidentally found unilateral LT coalition on the right.



Figure 2: CT reveals a type II LT coalition according to Minnaar.

Table 3: Evaluation of total cases evaluable.

n (sex)	23 Male	21 Female
n (side)	43 (right)	43 (left)
	Unilateral	Bilateral
	28	32
Classification Minnaar	Right	Left
n Type I	8	24
n Type II	13	4
n Type III	21	15
n Type IV	1	1

Table 4: Incidence of LT coalition between male and female, classified according to Minnaar.

Classification Minnaar	Male	Female
n Type I	10	12
n Type II	7	5
n Type III	16	19
n Type IV	2	0

Table 5: Incidence of cases, classified according to Minnaar, differentiation between sex and unilateral and bilateral.

Unilateral male Minnaar	Right	Left		Bilateral male Minnaar	Right	Left
n Type I	1	3		n Type I	2	5
n Type II	2	0		n Type II	2	0
n Type III	2	4		n Type III	6	5
n Type IV	0	0		n Type IV	1	1
female				female		
n Type I	3	2		n Type I	2	4
n Type II	1	1		n Type II	1	2
n Type III	3	2		n Type III	6	2
n Type IV	0	0		n Type IV	0	0

Table 6: Incidence of equal or higher manifestation of LT coalition according to Minnaar classification.

Bilateral Minnaar	[%]	Male n	Female n
Right = Left	50	7	3
Right > Left	40	3	5
Left > Right	10	1	1
Right 2x > Left	25	2	3
Left 2x > Right	0	0	0

There are different classification systems for LT coalitions, the most common one being used is according to Minnaar. In his system the classification is based on radiological appearance, i.e. coalition between the bones of os lunatum and triquetrum [1,2,6,11,16-19]. However, in literature it is discussed, that this classification lacks to describe the high variance in the non-osseus LT coalition, because there are only 4 types. Minnaar I being the non-osseus coalition, Minnaar II-IV representing incomplete, complete osseus LT coalition and complete coalition combined with other carpal abnormalities. Singh et al. developed a different classification, based on the shape and site of the fused bones, whereas Burnett classified the coalition into osseus and non-osseus [2,10,12,17-20]. In our evaluation we adhered to the classification of Minnaar.

Citing literature, the highest incidence is reported in patients of African descent of 9,7%, in Caucasians the incidence is 0,1%, the incidence in the Asian population is rarely described so far [1,2,4,6-8,12,19]. Additionally, in some literature the highest incidence is described to be in females [1,2,4,6-8,10,12,16,20]. In our case report it is a male patient of Caucasian descent. However, our investigation did not cover incidence in various races.

In our patient the diagnosis of LT coalition was also made by incident, as described in literature [3,5,13,15]. Because the LT coalition was osseous and not inflicted in the injury and did not cause any symptoms, no further therapy of the LT coalition was deemed to be necessary. In literature fractures or ruptures of LT coalitions are discussed controversially. It is described by various authors, that fractured osseous coalitions should be treated operatively first by fusion, however other authors state, that most fractures of LT coalitions can be treated conservatively [2,5,6,9,13,15,19]. In our opinion the symptoms caused by the fracture or rupture of the LT coalition deem the therapy necessary, however further research is needed.

Although LT coalition is described in highly cited literature to be more common in women than in men in a ratio of 2:1 [1,2,4,6-8,10,12,16,20], we did not find a higher incidence in women in our literature research. On the contrary total number of reports of LT coalition were equal in men and in women. Additionally, men had a higher incidence in bilateral LT coalition whereas both sexes had equal incidence in unilateral LT coalition. Sy et al. report on a case series of 32 LT coalitions in 20 patients, the highest incidence was reported to be in 15 men [4]. Spaans et al. reported on developmental differences in the carpus [19]. They hypothesize multifactorial inheritance for LT coalition with a strong sex linkage, which they found in their sample of radiographic analyses. According to Sy et al. Spaans et al and others the incidence is 2:1 female to male. We agree with Spaans and Sy in multifactorial inherence, because in our literature review not only familial traits but disagree with a 2:1 higher incidence of LT coalition in females. The reason for a reported higher incidence in women might be, that most case reports rely on case series of one or two hospitals, thereby the sample is skewed. However, in some literature the incidence is described controversially. In summary the sum of case reports and case series reveals a different picture in incidence between male and female.

There are case reports or case series on bilateral LT coalitions and incidence of LT coalition is described to be more often bilateral [2,4]. However, most cases in literature are case reports about unilateral LT coalition, however a radiographic analysis of the contralateral wrist was sometimes not performed [5]. Hence unilateral incidence of LT coalition should be re-evaluated. Our patient presented a one-sided LT coalition Minnaar type II on the right side, in literature the majority of cases are bilateral such as in a large case series of Defazio et al. [2]. Sy et al. report on a large case series of 32 cases in 20 patients, 15 cases were bilateral and 5 of them had the same classification of LT coalition type III according to Minnaar on the left and the right-hand side, 7 had a higher type in classification, however the side of the incidence is not mentioned [4]. When counting case reports of unilateral LT coalition in literature available to us equal incidence of unilateral and bilateral LT coalition was seen. The reason for this might be, that case series most often report on larger numbers and get higher attention, whereas case reports are single report only. In the sum, however, these case

reports seem to be equal to the number of bilateral cases in case series. More research is needed on this topic.

Looking at bilateral LT coalitions and the incidence and difference of LT coalition from side to side according to the classification system of Minnaar very few research has been done so far. Defazio et al. report on a LT coalition case series of 92 cases, 36 cases of them were bilaterally and 1 had a difference in Minnaar classification of LT coalition one type higher on the right-hand side compared to the left-hand side. However, for further evaluation, the table provided did not meet our inclusion criteria [2]. Ritt et al. report on 9 cases, 4 of them were unilateral. 5 cases were bilateral, most cases had equal manifestation in classifications on both sides, however, in one patient the left side had a higher classification than the right side [6]. Förstner reports on 4 cases of LT coalition, 3 were unilateral, one left and one right sided with the same manifestation in classification [7]. An other large case series was provided by Sy et al. They report on 32 cases of LT coalitions [4]. Most cases were in male, and 15 cases were classified as type III according to Minnaar. In 15 cases the LT coalition was bilateral and in 5 cases the manifestation was classified as type III. However, the authors did not provide tables with the side of incidence of the LT coalition combined with the manifestation of the classification, but an overview of all cases and summary of their findings. Therefore, no difference in Minnaar classification could be established. Some case reports [1,2,8,19,20] are on bilateral incidence of LT coalition, showing a difference in manifestation between right-hand and left-hand side. Interestingly, in our investigation of bilateral LT coalitions in literature, incidence of a higher manifestation of LT coalition classified by Minnaar appeared to be on the right-hand side. Additionally, the highest number in incidence in differences in classification was two in type, according to Minnaar, on the right-hand side more than on the left-hand side. Incidence of left-hand being higher in one in classification according to Minnaar than the right-hand side was seen only in 2 cases in literature. However, no range of motion was given for the evaluated wrists.

The reason for the difference in manifestation between the wrist sides might be, that chondrification of the joint to be did not happen, a fusion of an additional bone led to LT coalition, the cartilage was destroyed during the process of ossification or an inflammation led to the LT coalition [2,4,7,10]. However, more research is needed.

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References

1. Lotter O, Stahl S, Luz O, Pfau M, Schaller HE. CASE REPORT Bilateral Paradoxically Symptomatic Luno-triquetral Coalition: A Case Report. *Eplasty*. 2010; 10: e47.
2. Defazio MV, Cousins BJ, Miversuski RA Jr, Cardoso R. Carpal coalition: A review of current knowledge and report of a single institution's experience with asymptomatic intercarpal fusion. *Hand (N Y)*. 2013 Jun;8(2):157-63. Doi: 10.1007/s11552-013-9498-5. Erratum in: *Hand (N Y)*. 2013; 8(2): 245.
3. Ali R, Khan F, Saeed S, Javed N. Lunotriquetral Coalition: An Unusual Cause of Wrist Pain. *Cureus*. 2019; 11(9): e5704. Doi: 10.7759/cureus.5704.

4. Sy MH, Diarra O, Diagne M, et al. La synoste pyramido-lunaire chez le noir African: a propos de trente-deux cas. *Ann Radiol (Paris)*. 1996; 39: 208 - 212
5. Ozyurek S, Guler F, Canbora K, et al. Asymptomatic lunotriquetral coalition: an incidental radiographic finding. *Case Reports*. 2013; 2013: bcr2013009429.
6. Ritt MJ, Maas M, Bos KE. Minnaar type 1 symptomatic lunotriquetral coalition: a report of nine patients. *J Hand Surg Am*. 2001; 26: 261-270. Doi: 10.1053/jhsu.2001.21520
7. Förstner H. Das Os lunato-triquetrum the os lunato-triquetrum. *Z Orthop Ihre Grenzgeb*. 1989; 127(2): 174-82. German. Doi: 10.1055/s-2008-1040106.
8. Ten Berg PT, Foumani M, Strackee S. A Rare Case of Bilateral Lunotriquetral Coalition and Bilateral Scaphoid Nonunion. *J Hand S*. 2015; 40: 1921.
9. Patel RV, Epelman MS, Meyers AB. Radiographically occult symptomatic lunotriquetral coalition in a child. *Cureus*. 2019; 11(8): e5364. DOI 10.7759/cureus5364.
10. Spaans AJ, Beumer A. Carpal Coalitions; Failures of Differentiation of the Carpus: A Description of Cases. *Open Journal of Radiology*. 2013; 3: 1-6. <http://dx.doi.org/10.4236/ojrad.2013.31001>
11. Al Bayati MAF, Al Khishali TJ, Al Bayati MMA, et al. Lunotriquetral Coalition: Report of Two Different Cases. *Clin Case Rep Open Access*. 2020; 3(4): 173.
12. Simmons BP, McKenzie WD. Symptomatic carpal coalition. *J Hand Surg Am*. 1985; 10(2): 190-3. Doi: 10.1016/s0363-5023(85)80103-9.
13. Kennedy R, Ethiraj P, Arun HS. Asymptomatic bilateral lunotriquetral coalition a rare case report. *J Clin Biomed Sci*. 2020; 10(4): 130-132.
14. Kahane S, Isaac SM, Wildin C. A new type of carpal coalition. *J Hand Surg Eur*. 2012; 37(6): 581-2. Doi: 10.1177/1753193412441764.
15. Laurencin CT, Cummings RS, Jones TR, Martin L. Fracture-dislocation of the lunotriquetral coalition. *J Natl Med Assoc* 1988; 90(12): 779-81.
16. Ogut E, Yildirim FB, Urguden M, Oruc F, Oguz N. Abnormal type III fusion between lunate and triquetrum: A case report, *Int J Surg Case Rep*. 2020. <https://doi.org/10.1016/j.ijscr.2020.10.108>
17. Mespreuve M, Vanhoenacker F, Verstraete K. Lunotriquetral coalition, a normal variant that may rarely cause ulnar sided wrist pain. *JBR-BTR*. 2015; 98: 72-78.
18. Mbaba AN, Ogolodom MP, Wekhe C, Maduka BU (2019) Lunotriquetral Coalition: An Infrequent Cause of Wrist Pain-A Case Report. *Health Sci J*. 2019; 13(4): 667.
19. Lotter O, Amr A, Stahl S, Clasen S, Schraml C, et al. Pseudarthrosis after disruption of an incomplete luno-triquetral coalition: a case report. *Ger Med Sci*. 2010; 8: Doc34. Doi: 10.3205/000123.
20. Alves MPT, Netto HBP, Pereira CC. Sinostose luno-triquetrum; relato de dois casos. *Rev Bras Ortop*. 2001; 36: 235-238