### Review

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# Avascular necrosis of the navicular (Müller-Weiss). A systematic review

Necrosis avascular del navicular (Müller-Weiss). Una revisión sistemática

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ABSTRACT. Müller-Weiss is a disease characterized by deformation, fragmentation and necrosis of the navicular, which presents with midfoot varus and longstanding pain, mostly in females. It is related to delayed ossification due to physical or nutritional stress, associated with abnormal force distribution. There are still few studies on this condition and there is no consensus in the literature on its classification and treatment. At first, conservative treatment does not show positive results, only acute symptomatic improvement. Isolated talonavicular arthrodesis is the method most commonly adopted by specialists, showing good results and scores in the early stages, while triple and double arthrodesis is the choice in advanced stages. Knowledge of the radiology and findings is important for planning and individualizing the choice of treatment in each case. At the moment, there are several different techniques for approaching Müller-Weiss disease, however, studies evaluating long-term approaches as well as their complications need to be carried out. We present a systematic review on this disease.

**Keywords:** Muller-Weiss disease, ostheonecrosis navicular, foot arthrodesis, avascular necrosis.

**RESUMEN.** Müller-Weiss es una enfermedad que se caracteriza por la deformación, fragmentación y necrosis del navicular, que cursa con varo en el mediopié y dolor de larga duración, mayoritariamente en el sexo femenino. Se relaciona con un retraso en la osificación debido al estrés físico o nutricional, asociado a una distribución anormal de las fuerzas. Aún existen pocos estudios sobre esta patología y no hay un consenso en la literatura sobre su clasificación y tratamiento. Al principio, el tratamiento conservador no muestra resultados positivos, sólo una mejora sintomática aguda. La artrodesis talonavicular aislada es el método más comúnmente adoptado por los especialistas, mostrando buenos resultados y puntuaciones en las etapas tempranas, mientras que la artrodesis triple y doble es la de elección en etapas avanzadas. El conocimiento de la radiología y sus hallazgos es importante para planificar e individualizar la elección del tratamiento en cada caso. Actualmente existen varias técnicas diferentes para abordar la enfermedad de Müller-Weiss, sin embargo, es necesario realizar estudios que evalúen los abordajes a largo plazo, así como sus complicaciones. Presentamos una revisión sistemática en torno a esta enfermedad.

**Palabras clave:** enfermedad de Müller-Weiss, osteonecrosis navicular, artrodesis del pie, necrosis avascular.

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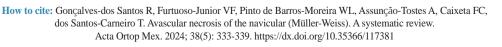
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#### Introduction

Avascular necrosis of the navicular was first reported by Müller and Weiss in the 20th century, who described its radiological appearance of condensation and fragmentation. It is a rare disease that affects adults between the fourth and sixth decade of life, and there are studies that relate its prevalence to women with an increased body mass index, but with limited data. The navicular bone has a large articular area, which is related proximally to the talus, laterally to the cuboid and distally to the cuneiform bones. It is also supplied by branches of the dorsal artery of the foot.

Several theories have been proposed to justify the pathophysiology of the disease, but its etiology is still unknown. The hypothesis that there is a delay in the ossification of the navicular is understood as a prerequisite, in addition to which an abnormal force distribution is necessary to produce the condition.<sup>3</sup> Aseptic or avascular necrosis occurs due to dysplasia of the bone tissue as a result of an alteration in the uneven distribution of compressive stress and delayed consolidation, with onset during childhood due to nutritional-environmental deficits or metabolic alterations.<sup>4</sup>

In addition, Monteagudo M. and Maceira E. demonstrated the association of situations of physical and emotional stress with delayed limitation, which can be seen with an increased incidence of cases in patients who migrated to Barcelona in the 20th century fleeing the war and also in athletes with intensive training in childhood.<sup>5</sup> However, its symptoms only appear in adulthood.<sup>6</sup> The mechanical overload on the navicular along with its composition in the medial arch, resulting in plastic deformation of the chondral structure and the vascular component due to compressive alteration with vascular obstruction are other factors related to the onset of the disease.<sup>3,6</sup> Trauma and stress fractures are important contributors of biomechanical origin, due to the potentiation of intraosseous vascular involvement.<sup>3</sup>

Müller-Weiss disease starts with insidious, vague pain in the midfoot region, patients also report ankle instability, reduced subtalar mobility and peroneal tendonitis.<sup>3,7</sup> Perinavicular arthritis is the main finding in studies and radiographs show osteophytes, sclerosis and dorsolateral fragmentation of the navicular, factors also associated with paradoxical flatfoot.<sup>7,8</sup> Necrosis can be unilateral or bilateral.

### Material and methods

This systematic review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).

# Search strategy and selection criteria

A systematic literature search was carried out in the following databases: PubMed/MEDLINE, Embase, Cochrane and Scopus. The main health science descriptors

searched in English were: «Muller-Weiss disease» and «Ostheonecrosis of the navicular». The full phrase used for the search was «Muller-Weiss disease» AND «Ostheonecrosis of the navicular» in the databases. Initially, all the studies that discussed the treatment of avascular necrosis of the navicular were approached for screening and selected according to the criteria.

### Search strategy and selection criteria

The inclusion criteria were: **a**) studies that dealt with Muller Weiss disease or avascular necrosis of the navicular; **b**) studies that used validated functional criteria for functional assessment; **c**) followed up cases for at least 12 months; **d**) classified the stage of development of the disease.

The exclusion criteria were: **a**) studies that did not evaluate the morphofunctional changes of the navicular in humans; **b**) studies that carried out tests on cadavers; **c**) complications related to other pre-existing pathologies; **d**) studies that did not fully describe the treatment; **e**) biomechanical tests.

The articles were also selected according to the Population, Intervention, Comparison, Outcome (PICO) strategy.

Population: patients diagnosed with avascular necrosis of the navicular (Müller-Weiss syndrome).

Intervention: functional analysis of conservative or surgical treatment for a period of more than 12 months.

Comparison: by observing the evolution of the disease with the treatment modalities.

Outcome: to indicate the most appropriate approach according to the degree of development of avascular necrosis of the navicular.

### Functional assessment tool

Given that the aim of this study was to assess function based on the degree of development and treatment modality, it was decided to use scales that are widely used, such as the American Orthopaedic Foot and Ankle Society (AOFAS) and the visual analog scale (VAS). However, due to the low incidence of avascular necrosis of the navicular and the lack of studies, other classifications can also be used, so that they can be validated.

### Data extraction

After initial evaluation of the abstracts against the criteria, the relevant studies were selected for full reading and sorted. The data was extracted by a research team made up of five independent reviewers. Disagreements between the reviewers regarding the inclusion or exclusion of a study were resolved by consensus and, when necessary, a sixth reviewer was consulted. The variables collected included mean age, gender, follow-up time, interventions carried out, degree of deformity and the functional results obtained.

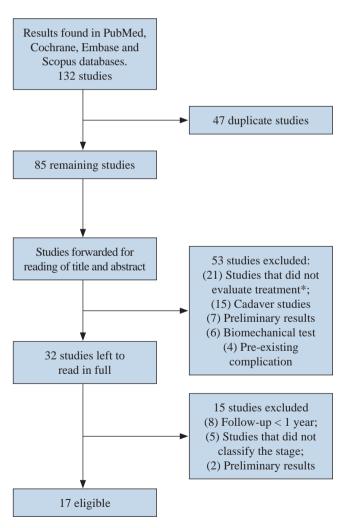
### Quality assessment

To assess the quality of the studies found, the Newcastle-Ottawa scale was used, which evaluates eight items and scores 0-9 points in relation to the selection criteria, comparability and outcomes of the control studies.

#### Results

## Literature search and study characteristics

A content review was carried out, first on titles and abstracts and then on full texts, which found a total of 132 articles. The 47 duplicate articles were excluded. Of these, 53 were excluded because they did not meet the inclusion criteria, so the exclusion was carried out according to the other criteria. After analysis, 17 articles were considered eligible (*Figure 1*).



\* These studies only addressed etiopathological factors of the alterations, as well as alterations in imaging tests such as CT and MRI scans.

Figure 1: Flowchart for screening and selecting the studies analyzed.

#### Characteristics of the references

Among the trials, there were a total of 134 patients (138 feet) diagnosed with avascular necrosis of the navicular (Müller-Weiss) covered in the studies (*Table 1*), whether in the indeterminate, chronic or acute form.

The data collected included possible risk factors for developing the disease. The results analyzed included presenting symptoms, characteristics of navicular changes and forms of treatment.

#### Avascular necrosis of the navicular

From the studies selected, it can be seen that the number of patients found was small, due to the low incidence of this presentation. The average age of the studies varied between 28-69 years, which was related to risk factors such as physical and nutritional stress in childhood and obesity, respectively. Despite this, most of the studies featured individuals over 40 years old. There was also a significant predominance of females in the samples.

Various treatments were used, the most prevalent being arthrodesis, performed alone, twice or three times. Calcaneal osteotomy was also used, as was grafting. Conservative treatment was well evaluated in the early stages (S1-S2), while the surgical approach was better in the remaining stages.

### **Discussion**

### Clinical aspects

The main characteristic is long-term mechanical pain in the dorsum of the foot, which can be asymmetrical or not.<sup>3,9</sup> The worsening of the condition in 10% of cases occurs after some trauma to the foot or with the arrival of the fifth decade of life.10 The conformational alteration of the navicular can be sustained for years without symptoms, but in some patients it is a limiting condition in the first decades of life. 10 Diagnosis occurs between the fourth and fifth decade of life, with females predominating in 70% of cases.<sup>3</sup> Pain in other joints such as the knees is also observed, which may be related to altered biomechanics in the whole limb, with the secondary manifestations being alterations in the foot. 11 The foot may be normal, with a high arch or low hindfoot, the talar head is on the dorsum of the midfoot with lateral deviation and subtalar movement is reduced.<sup>12</sup> Increased plantar pressure on the midfoot, combined with reduced pressure on the toes, promotes pain, and these considerations can help in the choice of treatment.13

# Radiological findings

The main changes occur in the navicular, but the radiological appearance can mimic osteonecrosis and changes peculiar to the midfoot can distort the diagnosis,

Table 1: Epidemiological characteristics, classification.								
Study	Year	Patients/ Feet	Mean age	Sex	Stage	Treatment	Follow-up	AOFAS Outcome
Volpe A, et al. <sup>41</sup>	2020	4 feet/ 4 patients	59.7	1 male/ 3 females	1 feet/S1 1 feet/S3	A-magnetotherapy (8 h/d) (S1 and S3)	24 months	A-91 B-73
Nelson EW, Rivello GJ <sup>40</sup>	2012	1 feet/ 1 patient	28	1 female	2 feet/S4 1 feet/S3	B-TNA (S4) TNCA	38 months	97
Wang X, et al <sup>42</sup>	2012	7 feet/ 6 patients	54	1 male/ 5 females	4 feet/S4 2 feet/S5	A-TNA (four S4 and one S5)	23.2 months	A-90.8 B-88
Li SY, et al <sup>54</sup>	2017	14 feet/ 13 pa-	56	6 males/	5 feet/S2 4 feet/S3	B-TA (one S5) Calcaneus osteotomy	44.4 months	78.9
		tients		/ Temares	4 feet/S4 1 feet/S5	oscotomy	monus	
Tan A, et al <sup>53</sup>	2011	1 feet/ 1 patient	51	1 female	1 feet/S4	Femoral head allograft	14 months	NA
Adams SB, Danilkowicz	2021	1 feet/ 1 patient	42	1 male	1 feet/S3	Talonavicular Joint-Sparing 3D Printed Navicular	48 months	87.5 (FADI)
Kitaura Y, et al <sup>48</sup>	2019	2 feet/ 2 patients	69	2 females	2 feet/S3	TNCA	60 months	98.5 (JSSF)
Chang SM, et al <sup>51</sup>	2019	1 feet/ 1 patient	38	1 female	1 feet/S2	Light bulb procedure for failed percutaneous decompression	72 months	` 97 <sup>^</sup>
Levinson H, et al <sup>28</sup>	2013	1 feet/1 patient	25	1 male	1 feet/S2	Medial femoral condyle vascularized bone graft	18 months	NA
Janositz G, et al <sup>52</sup>	2011	1 feet/ 1 patient	18	1 female	1 feet/S2	Percutaneous decompression	96 months	NA
Hu D, et al <sup>32</sup>	2017	13 feet/ 13 patients	59	2 males/ 11 females	7 feet/S3 6 feet/S4	TNA	20 months	83.5
Qu F, et al <sup>33</sup>	2019	14 feet/ 14 patients	46.2	3 males/ 11 females	5 feet/S3 9 feet/S4	TNA and calcaneus osteotomy	22.3 months	90.6
Harnroongroj T, Chuckpaiwong B <sup>31</sup>	2018	14 feet/ 14 patients	57.4	2 males/ 14 females	7 feet/S3 9 feet/S4	TNA	36-96 months	87.5
Lu CK, et al <sup>38</sup>	2014	13 feet/ 13 patients	55.6	1 male/ 12 females	2 feet/S3 11 feet/S4	TNA	114 months	87.2
Yuan C, et al <sup>39</sup>	2019	36 feet/ 34 patients	53.7	3 males/ 33 females	9 feet/S2 10 feet/S3 9 feet/S4 8 feet/S5	TNA or TNCA	25-115 months	85.3
Bai W, et al <sup>47</sup>	2023	15 feet/ 15 patients	48.4	2 males/ 13 females	8 feet/S3 7 feet/S4	TNA	74 months	90.2

TNA = talonavicular arthrodesis. TNCA = talonaviculocuneiform arthrodesis. TA = triple arthrodesis. NA = not available. FADI = Foot and Ankle Disability Index. JSSF = Japanese Society for Surgery of the Foot.

which is why it is important to know the morphology of the navicular (decreased size, comma-shaped configuration, increased radiodensity, bone fragmentation and protrusion) and the clinical history. 3,14,15,16 All patients initially have a straight midfoot, but may also have biomechanical and morphological complications in other segments. In the ankle and hindfoot, there is a decrease in the talocalcaneal divergence angle, an increase in the retroposition of the fibula in relation to the tibia due to ankle rotation and the orthogonal projection of the tarsal canal becomes visible, as well as the head of the talus being moved laterally. 3,17

In the midfoot, the most affected region, there is anteroposterior shortening of the navicular, which can bring the head of the talus into contact with the cuneiforms.<sup>3,18</sup> The dorsoplantar view best identifies the deformity of the navicular, which can be compressed at its lateral end in the shape of a comma with fragmentation in the sagittal plane, as well as internal rotation of the medial fragment of the navicular producing shortening.<sup>3,19</sup> Some patients have medial subluxation of the cuboid in relation to the calcaneus.<sup>20</sup>

In the forefoot, the metatarsals become parallel to each other with reduced divergence between the first and fifth, and some patients may complain of hallux valgus without joint misalignment.<sup>3,21</sup> Another finding is hypertrophy of the second metatarsal due to force deviation, which makes the first ray insufficient and the second cuneometatarsal ray the main force transmitter.<sup>22</sup>

### Degree of deformity

According to the appearance of the degree of deformity, it is possible to classify navicular involvement into stages. Stage 1 shows radiographs with no changes or minimal changes with slight subtalar varus. Stage 2 occurs with lateral displacement of the talar head and subtalar varus. Stage 3 shows lowering of the longitudinal arch and reduction of the space between the talus and cuneiform. Stage 4 shows hindfoot equinization, and hindfoot varus is accentuated. Stage 5 is a talocuneiform joint with complete extrusion of the navicular.

Maceira's classification is not valid for all cases. In his study, Wong-Chung et al. observed that only 35% of cases could fit into the classification, noting that the heel is not varus in all cases and that the greatest degrees of compression do not necessarily occur in the highest stage, which would not justify the universal proposal of calcaneal osteotomy.<sup>2,3</sup>

### Treatment

#### Conservative treatment

Conservative treatment may be suggested in the early stages of the disease, however there are factors that make treatment unsuccessful, such as midfoot abduction and talonavicular arthritis, which have higher pain and functional disability scores.<sup>26</sup> The initial diagnosis is difficult to make because there are few or no symptoms; tests such as magnetic resonance imaging and bone scintigraphy can help at this stage.<sup>27</sup> An alternative in mild cases is the use of orthopedic insoles with plantar support to immobilize the talonavicular and naviculocuneiform joints, which can lead to symptomatic improvement in the short term.<sup>8,28,29</sup>

### Talonavicular arthrodesis

Treatment should initially be conservative with pain control, changes in activities and supportive or accommodative orthoses, but if this becomes ineffective and leads to functional incapacity, surgical treatment is recommended.<sup>3,7,30</sup> A 3-8 year retrospective study evaluating talonavicular arthrodesis in isolation in 16 patients showed that this procedure provides a stable, non-painful foot for daily activities, although it does impair recreational/sporting activities. He also reported an improvement of 2.5° in the calcaneal inclination angle and 5° in midfoot abduction.<sup>31</sup> If the subtalar and calcaneocuboid joints are healthy, arthrodesis is a reliable option in the study that followed the patient for 14-39 months, with a fusion time

of 12-16 weeks, which was observed with an increase in the American Orthopaedic Foot and Ankle Society (AOFAS) and visual analog scale (VAS).<sup>32</sup>

Osteotomy of the calcaneus using an oblique incision on the lateral border with a 4cm fragment can also be associated with arthrodesis, with satisfactory healing results and clinical outcomes due to the ease of the procedure, and is recommended in patients with midfoot collapse. <sup>15,33</sup> An evaluation of 14 cases followed up for 42 months found that calcaneal osteotomy reduced VAS and increased AOFAS by up to 50 points, as well as being an alternative regardless of the radiological degree of the disease, which has also been observed in other studies. <sup>24,34</sup>

Magnetic resonance imaging has shown that both talonavicular and talonavicular-cuneiform arthrodesis in stage III or IV disease have good clinical results with a solid fusion rate and an obvious improvement in patients' quality of life. 35,36 The use of talonavicular fusion with a tension band has also been studied for stage III or IV and provided clinical improvement and stability. 37

The main associated complications are naviculocuneiform arthritis, pseudoarthrosis and talonavicular arthrosis, which can generate pain and reduce functionality. Fixation with an H-shaped locking plate and an additional retaining screw provides greater stability and compression, with a union rate of 97.2%. However, there is still no gold standard for surgical treatment; several studies using different techniques have shown satisfactory results. <sup>39,40,41</sup>

In this attempt, studies have classified avascular necrosis of the navicular according to surgical indication, with fracture of the dorsal cortex (type I), propagation of the fracture to the navicular body (type II) and propagation of the fracture to another cortex (type III), with surgical recommendation for types II and III after prior cast immobilization for six weeks.

Triple arthrodesis is an alternative for reconstruction of the medial arch and an alternative in cases where there is not only isolated talonavicular involvement. 42,43,44 In addition, its use in stages III and IV of Muller Weiss disease has been shown to improve x-ray length, arch height and reduce the Meary-Tomeno angle. 45 Another study evaluating this procedure also achieved satisfactory postoperative scores and little symptom recurrence. 46,47 Likewise, double arthrodesis (talonavicular and naviculocuneiform) associated with blocked plates and autologous iliac crest bone grafting show satisfactory results.<sup>48</sup> This procedure was evaluated in seven patients followed up for 38 months with complete fusion in 16 weeks and improvement in AOFAS of 33 points.<sup>49</sup> Studies of midfoot and hindfoot arthrodesis with stem allograft followed up for 13 years indicate that this method is also reliable.50

# Grafting

Transillumination is an alternative treatment for the early stages of the disease, with fluoroscopy and transillumination with an arthroscopic light source on the medial aspect of the navicular, associated with percutaneous decompression and grafting demonstrating satisfactory remodeling in patients.<sup>51</sup> Another possibility is the vascularized bone graft from the medial condyle of the femur, which has minimal morbidity in the donor area, has shown a higher functional score and an early return to work, although this is an experimental study of just one case.<sup>28,52,53</sup>

Important findings also suggest that percutaneous decompression in the early stages is an option when prolonged conservative treatment is not efficient, since it was used in young athletes who returned to their sporting activities, however in the long term the results are less favorable with worsening symptoms, with the need for extensive interventions due to the increase in deformity, such as triple arthrodesis.<sup>52</sup>

### Other modalities

Another treatment studied in the literature is the complete replacement of the navicular with a 3D-printed titanium prosthesis with preservation of the talonavicular joint. This procedure is an alternative to the use of grafts, due to the reduced blood supply to the navicular bone and in cases of bone loss. The report followed up and evaluated the replacement for four years, which maintained anatomical alignment, preserved joint function and satisfactory functionality scores.<sup>50</sup>

# **Final considerations**

Avascular necrosis of the navicular or Müller-Weiss disease is a complex foot disorder that presents with mediopepis adductus and long-standing pain, mostly in females. There is no consensus on treatment in the literature, as the studies have several limitations and follow-up needs to be long-term in order to better define the therapeutic choice. It is essential to use radiological evaluation to assess the joints involved preoperatively and then choose the appropriate method to treat the different patients, taking into account the degree of arthritis and type of navicular fragmentation.

In addition, this study has inherent limitations in terms of the level of evidence of the studies, which were not all randomized clinical trials, making it impossible to conclude on treatment modalities.

Various surgical treatments have been proposed to correct the deformity associated with Muller Weiss disease and relieve pain. Isolated talonavicular arthrodesis shows good results in the early stages without compromising adjacent joints (S2-S3). Percutaneous decompression can be used as a palliative treatment in some cases (S5). Double arthrodesis and triple arthrodesis are most commonly used in stages S3 and S4 with greater involvement and deformity. Calcaneal osteotomy associated with arthrodesis can be an alternative in cases of midfoot collapse. Grafts show good results when associated with arthrodesis in the reconstruction of

the midfoot, the most commonly used being from the iliac crest. There is a wide range of techniques available for the treatment of avascular necrosis of the navicular, however the choice of method must be individualized in order to preserve or not preserve the joint.

Thus, in view of the results, new trials are needed using validated protocols as well as assessing the need for resources and the associated costs, as well as adapting to the local reality of each service.

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