

Original Article

Müller-Weiss disease: epidemiology, surgical case series, and literature review

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Abstract

Objective: To report a surgical case series, including the epidemiological profile and clinical and functional outcomes of patients with Müller-Weiss disease (MWD), thereby increasing knowledge of the pathology.

Method: Patients diagnosed with MWD who underwent surgical treatment between January 2010 and December 2021 were included in the study. Epidemiological information, comorbidities, reported symptoms, classification according to Maceira, surgical technique, time from symptom onset to surgery, and postoperative complications were recorded. Fisher's exact test was used to analyze the association between categorical variables, and odds ratios (ORs) were calculated with their respective 95% confidence intervals, with a significance level of 5% ($p < 0.05$).

Results: Forty-two patients (44 operated feet) with MWD were included in the sample, with a mean follow-up of 36.3 months. The mean age was 57.9 (± 9.4) years, and the body mass index was 32.2 (± 4.1) kg/m². There was a higher proportion of women (80.9%) and white patients (42.8%). The main comorbidity was systemic arterial hypertension (SAH), observed in 64.3% of cases, followed by obesity and type 2 diabetes (DM2), present in 59.5% and 14.3%, respectively. Pain was the main symptom, reported by 92.8% of patients. Maceira stage IV was the most prevalent (29.5%), and 57.1% of patients had bilateral involvement. The main procedures performed were isolated triple arthrodesis (34%), isolated talonavicular arthrodesis (22.7%), and talonavicular arthrodesis with calcanectomy (18.2%). Most patients (52.3%) underwent surgery within five years of symptom onset. After 12 months of surgery, 59.1% of patients reported residual pain, and 31.8% were reoperated on. Among these patients, patients with DM2 had a higher risk of chronic pain.

Conclusion: In this study, MWD presented a higher prevalence in female patients, white, over 50 years of age, hypertensive, and with obesity. Painful and bilateral presentation and diagnosis in advanced stages were observed in a large portion of cases. Triple arthrodesis was the most frequently performed procedure, followed by isolated talonavicular arthrodesis. One year after surgery, most patients reported residual pain, and this complication was more associated with the presence of DM.

Level of Evidence IV; Case Series.

Keywords: Orthopedics; Flatfoot; Treatment Outcome.

Introduction

Müller-Weiss Disease (MWD) is a complex orthopedic condition characterized by fragmentation and collapse of the navicular bone, with deformity and pain^(1,2). Previous studies have reported greater involvement in women aged 40 to 60 years, with bilateral involvement ranging from 40% to 75% of cases⁽³⁻⁵⁾.

Its etiology is controversial, including combined theories of delayed ossification of the navicular and abnormal distribution of forces in the foot, as well as trauma, congenital dysplasia, and metabolic and autoimmune systemic diseases^(1,2).

Several epidemiological scenarios that influence the development of MWD were identified, including childhood epidemic environmental stress, childhood nutritional stress,

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foot deformities (metatarsal adduct and hindfoot varus), high-performance sports during childhood, and idiopathic involvement, in addition to the late/adult-onset of MWD. Although the first descriptions of MWD suggest that it is osteonecrosis of the navicular bone, histological studies have failed to confirm this initial hypothesis^(1,6,7).

MWD presents as pain in the back of the foot, associated or not with ankle instability, and pain in the fibular tendons⁽⁸⁾. The medial longitudinal arch of the foot may be normal or reduced, associated with hindfoot varus deformity. The paradoxical flatfoot is characteristic of MWD and occurs in advanced stages of the disease, due to lateral collapse of the navicular and consequent lateral and plantar protrusion of the talar head, leading to subtalar joint varization. The inversion calcaneus, associated with plantar flexion of the talus, leads to worsening of hindfoot varus and adaptive external rotation of the tibia, thereby modifying the biomechanics of the lower limb^(2,9).

Maceira and Rochera⁽⁸⁾ reported five stages of the disease based on radiographs obtained in simple lateral incidence in orthostasis. The classification describes progressive changes with navicular fragmentation and associated deformities, such as plantar flexion of the talus and subtalar joint varus^(1,2,10).

Although described as a rare disease, Monteagudo and Maceira⁽¹⁰⁾ stated that the pathology is underdiagnosed, and several authors suggest that the prevalence may be higher than that reported^(2,3). In fact, there is a lack of epidemiological records and data in the literature to support indications and potential surgical methods for the treatment of MWD. The objective of this study is to report a surgical case series, including the epidemiological profile and clinical and functional outcomes of patients with MWD, thereby increasing knowledge of the pathology.

Methods

This retrospective longitudinal case series included patients diagnosed with MWD, aged 18 years or older, who underwent surgical treatment between January 2010 and December 2021 and had epidemiological data and complete pre- and postoperative radiographic follow-up in their medical records. Of the 68 patients initially identified, 17 were excluded due to incomplete epidemiological information in the medical record, and nine due to incomplete radiological study. The presence of comorbidities was not considered an exclusion criterion in this study. The final sample consisted of 42 patients, with two cases of bilateral involvement, totaling 44 feet, with a mean time between surgery and evaluation of 36.3 months (ranging from 3 - 99 months). The research was approved by the Institutional Review Board.

The diagnosis of MWD was made through physical and radiological examinations, including simple foot radiographs in anteroposterior and lateral views obtained in orthostasis, showing osteonecrosis of the navicular^(2,8). To define the epidemiological profile and the surgical outcomes presented,

the following were evaluated: demographic information; complaints of joint pain and/or instability; presence of deformities; surgical technique employed; time between symptom onset and surgery; postoperative complications; and the need for reoperations. The severity of MWD was classified according to the classification proposed by Maceira⁽¹⁰⁾.

Consolidation of less than 25% of the joint surface, as evaluated on simple radiographs three months after surgery, was established as a radiographic criterion for pseudarthrosis.

Data description was performed using absolute and relative frequencies, measures of central tendency, and dispersion. Fisher's exact test was used to analyze the association between categorical variables. Odds ratios (OR) were calculated with respective 95% confidence intervals, and a significance level of 5% ($p < 0.05$) was considered. Excel® 2016 version 1804 (16.0.9226.2156) was used to calculate the obtained data.

Results

The medical records of 42 operated patients diagnosed with MWD were reviewed; the cohort comprised 44 feet, 80.9% female, and 42.8% white. The mean age of the patients, based on the date of surgery, was 57.9 (± 9.4) years, and the body mass index (BMI) was 32.2 (± 4.1) kg/m². The main comorbidity reported was SAH, in 64.3% of cases, followed by obesity and type 2 diabetes (DM2), present in 59.5% and 14.3%, respectively. The main symptom reported was pain, in 92.8% of cases, with previous trauma reported in 30.9% of the sample. Of the 42 patients, 57.1% had bilateral involvement. According to the Maceira classification, among the 44 feet evaluated, stage IV was the most prevalent, with 13 cases (29.5%), followed by stages III and V, with nine cases each (Table 1).

Among the surgical procedures performed, the most frequent were triple arthrodesis in 15 cases (34%), calcaneal valgus osteotomy in 12 patients (27.3%), fixed with cannulated screws of 7.0 mm or kirchnner wires, and in eight cases osteotomy was associated with talonavicular arthrodesis, fixed with cannulated screws of 3.5 mm or 4.0 mm, and isolated talonavicular arthrodesis in 10 cases (22.7%) (Table 2) (Figures 1 and 2).

Most patients (52.3%) underwent surgery within five years of symptom onset. As acute postoperative complications, there was surgical wound dehiscence, one patient with skin necrosis, and one patient with pain due to prominence of the synthetic material in the talonavicular arthrodesis. There was no record of pseudarthrosis in our sample. After surgery, 26 patients (59.1%) reported residual pain; among them, patients with DM2 had a higher risk observed for this outcome (OR:10.0, p-value 0.10) (Table 3). And, 14 patients (31.8%) underwent new surgery, seven for synthetic material removal, and seven underwent arthrodesis (primary or new arthrodesis of the adjacent joint) (Table 4) (Figures 3 to 5).

Discussion

Studies investigating the outcomes of surgical treatment of patients with WMD are still scarce, largely due to misdiagnoses^(1,2,8,10). The main findings of this study were the predominance of female and white patients, a mean age of 57.9 years, and a BMI of 32.2 kg/m². The main comorbidities identified were SAH and obesity. Pain was the main complaint, present in 92.8% of patients, with bilateral involvement in 57.1% of cases. Maceira stage IV was the most prevalent

Table 1. Epidemiological data on patients with Müller-Weiss disease undergoing surgical treatment (n = 44 feet)

		n	%
Maceira classification	Stage I	6	13.6
	Stage II	7	15.9
	Stage III	9	20.4
	Stage IV	13	29.5
	Stage V	9	20.4
Sex	Female	34	80.9
	Male	8	19.1
Race/color	White	18	42.8
	Black	9	21.4
	Mixed race	15	35.7
Main complaint	Deformity	3	7.1
	Pain	35	83.3
	Pain, deformity	4	9.5
Side	Right	16	38.1
	Left	24	57.1
	Both	2	4.8
Time from symptom onset to surgery	Up to 5 years	24	54.5
	6 to 10 years	11	25
	≥ 11 years	9	20.4
Postoperative pain	Yes, less than 1 year	14	31.8
	No	18	40.9
	Yes, after 1 year	12	27.3
Reoperation	No	30	68.2
	Yes	14	31.8

Table 2. Surgical procedures performed in our sample (n = 44)

Procedures	n	%
Triple arthrodesis	15	34
Cacanectomy (Associated with talonavicular arthrodesis)	12 (8)	27.3 (18.2%)
Isolated talonavicular arthrodesis	10	22.7
Talonaviculocuneiform arthrodesis	3	7
Talonavicular and subtalar arthrodesis	3	7
Subtalar and calcaneocuboid arthrodesis	1	2



Figure 1. Preoperative radiographs of a patient with Müller-Weiss disease and symptomatic talonavicular and subtalar osteoarthritis.

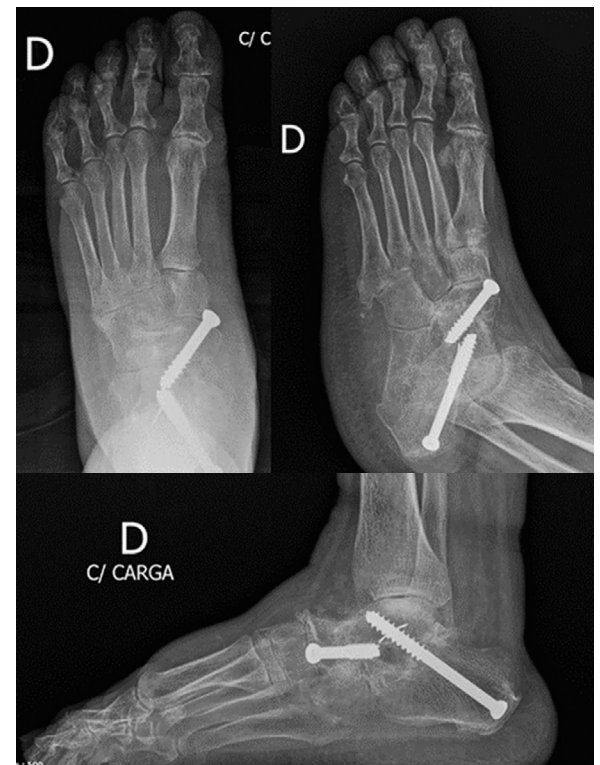


Figure 2. Radiographs of the same patient in Figure 1 showing the procedure used (talonavicular and subtalar arthrodesis with screws).

Table 3. Association between comorbidities and residual pain (n = 42)

Variable	Residual pain (n, %)	No pain (n, %)	OR (95% CI)	p-value
No comorbidities (n = 6)	2 (33.3%)	4 (66.7%)	1.00 (reference)	—
SAH (n = 27)	17 (63.0%)	10 (37.0%)	3.40 (0.54–21.2)	0.19
Obesity (n = 25)	17 (68.0%)	8 (32.0%)	4.25 (0.66–27.3)	0.14
DM2 (n = 6)	5 (83.3%)	1 (16.7%)	10.0 (0.64–15.6)	0.10
Smoking (n = 7)	5 (71.4%)	2 (28.6%)	5.00 (0.56–44.5)	0.15

OR: Odds ratio; CI: Confidence interval; SAH: Systemic arterial hypertension; DM: Diabetes mellitus. Reference group: Patients with no comorbidities. Fisher's exact test was used. P-values $p < 0.05$ are considered statistically significant.

Table 4. New arthrodesis procedures performed after one year

First procedure	Reoperation
Valgus osteotomy (2)	Triple arthrodesis
Osteotomy associated with talonavicular arthrodesis	Triple arthrodesis
Talonavicular arthrodesis	Associated with subtalar arthrodesis
Talonavicular arthrodesis (2)	Triple arthrodesis
Talonaviculocuneiform arthrodesis	Associated calcaneocuboid and intercuneiform arthrodesis



Figure 3. Preoperative radiographs of a patient with Müller-Weiss disease and symptomatic talonavicular arthrosis.



Figure 4. Radiographs of the same patient in Figure 3 showing the procedure used (talonavicular arthrodesis with two cannulated screws).



Figure 5. Radiographs of the same patient in Figures 3 and 4 that evolved to symptomatic subtalar arthrodesis. The reoperation was performed with subtalar arthrodesis with a cannulated screw.

(29.5% of cases). The most frequently performed procedures were triple arthrodesis and isolated talonavicular arthrodesis. One year after surgery, 59.1% of patients reported pain, and 31.8% underwent new surgery. Patients without comorbidities had a lower frequency of residual pain (33.3%). Finally, there was a progressive increase in the chance of residual pain in patients with DM2 (OR = 10.0), followed by smoking (OR = 5.0) and obesity (OR = 4.25).

A recent review study⁽¹¹⁾ demonstrated that WMD is more common in women, with greater bilateral involvement, especially between the fourth and sixth decades of life, and in individuals with high BMI. Our findings are consistent with what is most commonly reported in the literature. We observed 80.9% of female patients, bilateral involvement in 57.1%, a mean age of 57.9 years, and a predilection in patients with obesity (mean BMI 32.22 [\pm 4.15] kg/m²). In addition, we noted that 42.8% of the individuals were white, and foot pain represented the main complaint, reported by 92.8% of the sample. The largest study on WMD was conducted by Maceira and Rochera⁽⁸⁾, which described 191 cases. The author reported the association of WMD with external

injuries and social and nutritional stress, related to the Civil War in Spain and the long period of poverty to which the population was exposed⁽⁸⁾. Subsequent studies did not report the same associations but described other factors related to osteonecrosis of the navicular, including diabetes, alcoholism, corticosteroid use, and metabolic disorders⁽¹²⁾.

A previous study involving 16 patients with WMD reported SAH in 25% and diabetes in 18.7%⁽¹³⁾. In our study, we observed partially similar results. The main comorbidities identified were SAH (64.3%), obesity (59.5%), DM (16.7%), and smoking (14.3%). Previous authors⁽⁸⁾ have highlighted the multifactorial theory, citing the rarity of cases observed with isolated predisposing factors. This idea reinforces the need to observe all clinical and epidemiological factors that may contribute to the development and progression of WMD.

Maceira and Rochera⁽⁸⁾ described a staging system comprising five progressive degrees of deformity. Although there is no well-established clinical-radiological-prognostic relationship⁽¹⁾, radiological staging facilitates understanding of the anatomical changes present in the disease⁽¹²⁾. In our study, stage IV was the most prevalent, observed in 29.5% of cases. Of the total sample, 70.4% of patients were in advanced stages of Maceira (III to V), with the majority being symptomatic. Doyle et al.⁽³⁾ published a study including 19 patients and reported that among the 12 symptomatic patients, 11 were between Maceira stages III and V. Other authors evaluated 36 symptomatic patients with WMD who underwent surgical treatment. Of these, 27 had Maceira stages III to V⁽¹⁴⁾. Our sample has a high rate of patients in advanced stages, possibly caused by the late diagnosis of WMD. In addition, our study is subject to selection bias, since only patients undergoing surgical treatment were included, which explains the low number of patients in the early stages of the disease.

The initial treatment of WMD is conservative and includes the use of anti-inflammatory drugs, orthoses, physiotherapy, and activity restriction⁽¹³⁾; however, unsatisfactory results are commonly reported⁽¹¹⁾. Surgical treatment is the option in refractory cases, with the main objectives of achieving a plantigrade, aligned foot, restoring the height of the medial column, and improving pain^(9,11). Most patients indicated for surgical treatment are in Maceira stages III to V⁽¹¹⁾, similar to those found in our study. There is no established surgical gold standard for WMD⁽¹⁵⁾, and each case should be analyzed individually based on the deformities. Among the numerous techniques described for the surgical treatment of WMD^(9,10), the choice usually involves arthrodesis of the affected joints⁽⁹⁾. In our sample, only four patients did not initially undergo arthrodesis, and only calcaneotomy was performed. However, two of them were later reoperated on with triple arthrodesis due to the persistence of symptoms. Isolated calcaneotomy is indicated when there is poor varus alignment of the hindfoot⁽⁹⁾ and has become the first surgical choice for Maceira and Monteagudo, regardless of the patient's radiographic stage⁽¹⁰⁾. Historically, surgical treatment with arthrodesis was directed at isolated fusion

of the talonavicular or talonaviculocuneiform joint, but was associated with higher rates of pseudoarthrosis compared with triple arthrodesis^(9,10). In cases with more advanced degenerative changes in the subtalar and calcaneocuboid joints, triple arthrodesis presents more satisfactory results^(9,10). In accordance with the literature, triple arthrodesis was the most common procedure in our study, as most patients were already in advanced stages of the disease.


Among the possible complications after surgical treatment are infection, pseudoarthrosis, symptomatic osteoarthritis in adjacent joints, and complications of the synthetic material and residual pain in the foot⁽¹⁶⁾. During postoperative follow-up of our patients, 14 reoperations were required, including skin debridement, removal of synthetic material, and new arthrodesis. Lu et al.⁽¹⁷⁾, performed triple or talonavicular arthrodesis in 12 patients with advanced WMD (stage 3 or higher), with a mean follow-up of 16.8 months. They reported a significant improvement in AOFAS, ranging from 43.4 ± 16.1 to 85.3 ± 6.2 after one year postoperatively, with no cases of skin complications or pseudoarthrosis. Ponz-Lueza et al.⁽¹⁸⁾ reported three complications among the nine patients operated on in the study, including skin necrosis, posterior tibial pain, and sural nerve neuropathy. Postoperative pain occurred in 26 patients, representing 59.1% of the sample. Of these, 14 patients reported pain in the first postoperative year, and 14 patients (38.1%) needed to be reoperated. In our study, we observed a high rate of complications and reoperations, both recent and late, as well as the need for additional procedures. Despite the lack of statistical significance, DM showed the strongest association (OR 10.0), suggesting a possible independent effect on the outcome of residual pain. This finding is consistent with pathophysiological

mechanisms related to microangiopathy and neuropathy resulting from the disease. Collectively, these data reinforce the importance of detailed and individualized preoperative planning that takes into account patients' personal factors, especially comorbidities, to avoid complications and future interventions.

There are several limitations in our study, including a small sample size, a retrospective design based on medical records, and the absence of functional evaluation of pre- and postoperative results, such as the AOFAS score or the visual analog scale of pain. Another limitation is the presence of multiple comorbidities and the use of various surgical techniques in our patients, factors that may have influenced clinical outcomes and reported complications. However, there is a shortage in the literature on the pathology, and further studies are needed to broaden the debate on WMD, especially its surgical treatment, which offers numerous options.

Conclusion

In this case series of patients with WMD undergoing surgical treatment, there was a predominance of females and white patients, aged over 50 years, hypertensive, and with obesity. With high frequency, the initial presentation occurred in advanced stages of the disease (Maceira stage IV), with pain as the main complaint and bilateral involvement in most cases. Surgical treatment with arthrodesis was the main therapeutic choice in the sample, with triple arthrodesis being the most frequently performed procedure. One year after surgery, 59.1% of patients reported pain, and the greatest association with the outcome of residual pain was with DM.

Authors' contributions: Each author contributed individually and significantly to the development of this article: IMCJ *(<https://orcid.org/0000-0002-7815-6086>) Conceived and planned the activities that led to the study; ACLO *(<https://orcid.org/0009-0005-1757-3766>) Conceived and planned the activities that led to the study and wrote the article; BASA *(<https://orcid.org/0000-0001-5269-9106>) and HM *(<https://orcid.org/0000-0001-7527-969X>) Wrote the article; TN *(<https://orcid.org/0000-0001-7988-6307>) and MSG *(<https://orcid.org/0009-0002-1006-633X>) statistical analysis. All authors read and approved the final manuscript. *ORCID (Open Researcher and Contributor ID) 

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