ORIGINAL ARTICLE

Tibiotalocalcaneal arthrodesis in a rare case of tuberculosis of the talus

Chiara Comisi[1,2] , Tommaso Greco[1,2], Michele Inverso[1,2], Antonio Mascio[1,2], Chiara Polichetti[1,2], Marco Barbaliscia[1,2], Vincenzo La vergata[1,2,] , Massimiliano Mosca[3], Silvio Caravelli[3], Nicola Mondanelli[4], Elisa Troiano[4], Giulio Maccauro[1,2], Carlo Perisano[1,2]

[1]Department of Aging, Orthopedic and Rheumatologic Sciences, Orthopedics and Trauma Surgery Unit, Fondazione Policlinico Universitario Agostino Gemelli IRCCS, Rome, [2]Orthopedics and Trauma Surgery, Università Cattolica del Sacro Cuore, Rome, [3]IRCCS Istituto Ortopedico Rizzoli - U.O.C. II Clinic of Orthopaedics and Traumatology, Bologna, [4]Department of Medicine Surgery and Neurosciences, University of Siena, Siena; Italy

ABSTRACT

Aim To assess our personal experience of a case of tuberculosis of the talus, and to provide an overview of the literature about this tuberculosis manifestations, including all its aspects: epidemiology, clinical and imaging presentation, and all the treatments available to the current state of knowledge.

Methods We present our experience in a case of a 34-year-old patient, who came to our attention with difficulty in walking and pain due to a talar tuberculosis, with consequent bone disruption and reabsorption, and foot deformities.

Results A tibiotalocalcaneal arthrodesis with retrograde nail and bone graft was performed after antibiotic therapy. Today, almost two years after treatment, the patient can walk independently with no major limitations in everyday life.

Conclusion Tibiotalocalcaneal arthrodesis with bone graft showed good functional results in this case study, with complete graft fusion and good functional and radiological outcomes.

Key words: chronic ankle disease, foot disease, tarsal bones, tibiotalocalcaneal arthrodesis, foot deformities

Corresponding author:
Chiara Comisi, Department of Aging, Orthopedic and Rheumatologic Sciences, Orthopedics and Trauma Surgery Unit, Fondazione Policlinico Universitario Agostino Gmelli IRCCS
Largo Agostino Gemelli, 8,
00168 Rome, Italy
Phone: +39 3385223830;
E-mail: chiara.comisi22@gmail.com;
ORCID ID https://orcid.org/0000-0001-8555-7411

Original submission:
01 December 2023;
Revised submission:
09 December 2023;
Accepted:
25 December 2023
doi: 10.17392/1699-23

Med Glas (Zenica) 2024; 21(1):222-228

INTRODUCTION

Mycobacterium tuberculosis (MT) was discovered by Robert Koch in 1882 as the etiological agent of tuberculosis (TB), proving the infectious nature of the disease (1). More than two centuries later, it remains one of the major health problems worldwide affecting 10 million people and causing more than 1.5 million deaths each year (among the world's top 10 causes of death) (2). MT is a rod-shaped organism whose main mode of spread is through aerosol droplets exhaled by a patient with active TB, most commonly causing a pulmonary infection (3).

Extrapulmonary TB (EPTB) accounts for 20% of all TB cases (3), and involve most common-ly genito-urinary, cardiovascular, cutaneous, gastrointestinal, and skeletal systems (2). Among the manifestations of bone TB, the spine involvement is the most frequent one, and it ac-counts for approximately 50% of all EPTB forms, causing a condition called Pott's disease (4). TB involving the foot and ankle bones has been reported in 8%-10% of patients with skeletal TB (approximately 1%-3% of all patients with EPTB) (5). In this subgroup talar in-volvement is extremely rare, and only few cases are described in literature.

According to the literature, TB of the foot and ankle can be successfully treated without surgery if antitubercular therapy is started at an early stage (6), but the rarity of the disease and its non-specific clinical presentation, often lead to a delayed or sometimes even missed diagnosis; in these cases an aggressive and invasive surgical treatment is often required (7).

In this study we reported our experience in a case of TB involving the talus but not the adjacent joint or bones, treated with a tibiotalocalcaneal (TTC) fusion with retrograde intramedullary nail and bone graft.

The aim of this study is to provide clinical and practical guidance to physicians regarding the management and treatment of this disease.

PATIENT AND METHODS

Patient and study design

A 34-year-old transgender woman was admitted to the Infectious Diseases Department of "Policlinico A. Gemelli" Institution for coughing and dyspnoea, whose chest X-ray was suspected of pulmonary TB. The medical history of the patient revealed HIV positivity, in chronic therapy with Efavirenz 600 mg/day; in addition, laboratory tests showed altered C-reactive protein (CRP) value (76.4 mg/L; normal value: 5-6 mg/L) and white blood cell (WBC) count (11x10^9/L; normal value: 4-10x10^9/L).

Methods

The patient underwent bacterioscopic, culture and molecular sputum examination, and resulted positive for MT complex. She started oral antibiotic therapy initially with 4 drugs (Isoniazid, Rifampicin, Pyrazinamide and Ethambutol) for 2 months, and subsequently with 2 drugs (Isoniazid, Rifampicin) for the next 10 months.

Approximately 2 months after diagnosis, during the oral therapy, lumbar spine pain and a painful swelling in the left foot and ankle developed, with significant impairment of walking.

Investigating the patient's medical history, it was discovered that about 3 years before, she reported a fracture of the talus and tibial plafond, treated with open reduction and internal fixation (ORIF) with plate and screws. Subsequently, the fixation devices were removed due to pain and discomfort. The clinical examination of the left ankle showed a severe varus deformity with instability to varus-valgus stresses, a complete inability to walk, and daily pain with an American Orthopaedic Foot and Ankle Society (AOFAS) score of 22 (8).

RESULTS

The patient underwent a computed tomography (CT) scan and a magnetic resonance imaging (MRI) of the spine, which documented T9-T10 spondylodiscitis, with the presence of intra-abdominal abscesses and within the spinal canal (Figure 1). In addition, X-ray, CT and MRI of the left foot and ankle were performed, reporting destruction of the tibiotalar joint, significant talus resorption, multiple abscess collections, and diffuse spongious bone edema in the hindfoot and midfoot; the diagnostic suspicion of osteomyelitis was high (Figure 2).

Surgical treatment was chosen for the Pott's disease: intra-canal and intra-abdominal abscesses were drained, and a T7-T12 dorsal fusion was

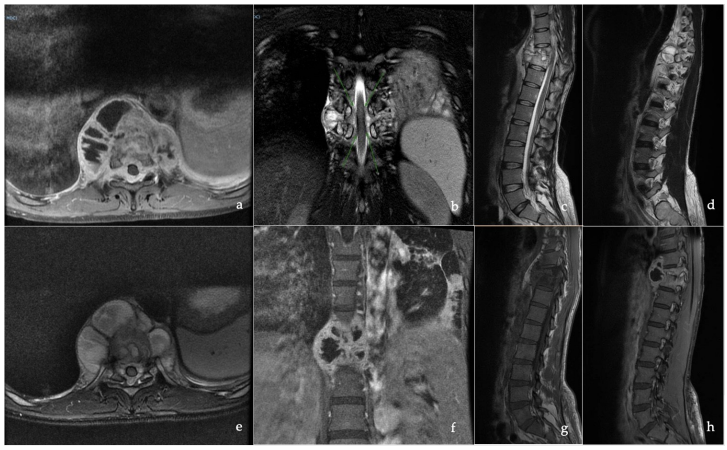

Figure 1. Pre-operative magnetic resonance imaging (MRI) of the spine: A) T2-axial view; B) T2-coronal view; C- D) T2-sagittal view; E) T1-axial view; F) T1-coronal view; G) and (H) T1-sagittal view (Fondazione Policlinico Universitario Agostino Gemelli IRCCS, 2022)

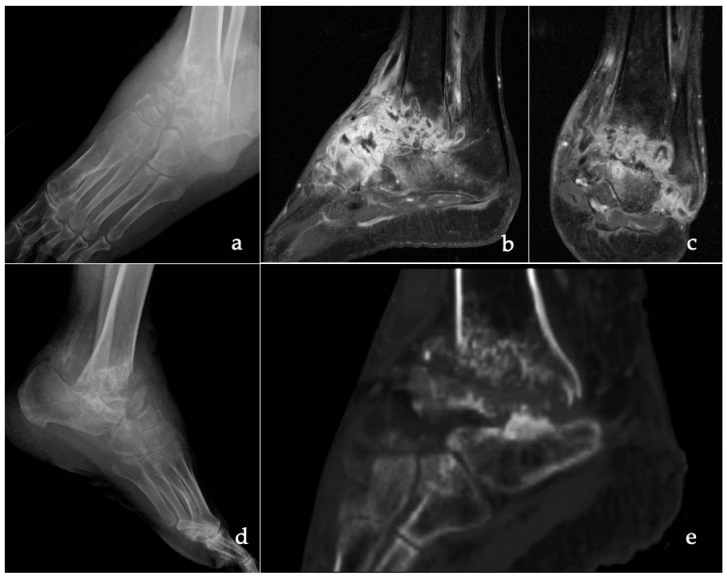

Figure 2. A) pre-operative anteroposterior X-ray of foot and ankle; B) sagittal view; and C) coronal view of pre-operative left foot and ankle MRI; D) pre-operative X-ray lateral view; E) sagittal view of pre-operative CT-scan (Fondazione Policlinico Universitario Agostino Gemelli IRCCS, 2022)

performed to avoid the risk of spinal collapse (Figure 3).

To obtain an etiological diagnosis the foot abscesses were drained, and the purulent material was sent for cultural microbiological examination, which subsequently tested positive for MT complex.

A Walker ankle brace was placed, waiting for the end of the anti-tubercular drug therapy. Subsequently, a TTC arthrodesis with retrograde intramedullary nail was planned, and a bone graft with femoral head was used to fill the talar disruption.

The surgery carried out under general anaesthesia in supine decubitus position. An antero-medial approach to the ankle through the previous surgical scar (Figure 4) was performed. After

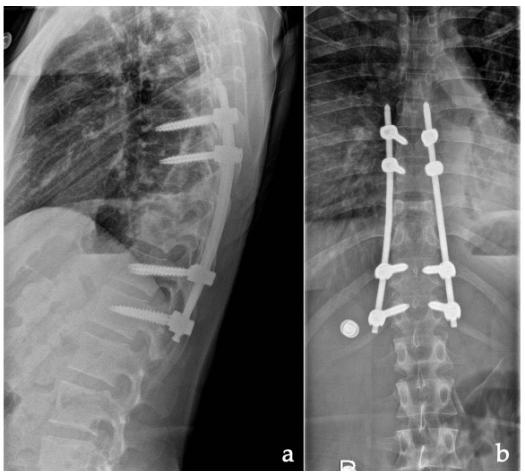

Figure 3. Post dorsal fusion A) lateral and B) antero-posterior view (Fondazione Policlinico Universitario Agostino Gemelli IRCCS, 2022)

Figure 4. Local soft-tissue conditions in A) anterior, B) medial and C) lateral region (Fondazione Policlinico Universitario Agostino Gemelli IRCCS, 2022)

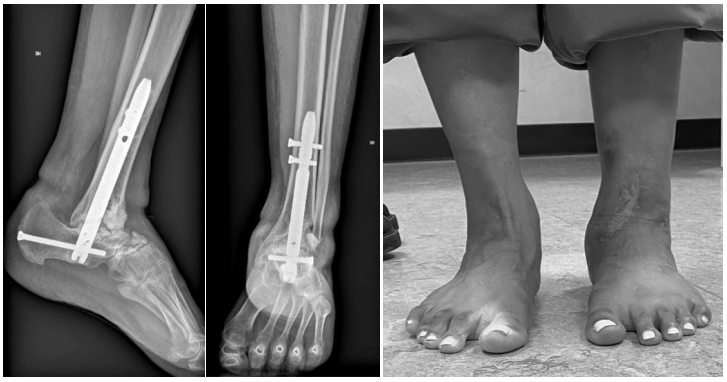

Figure 5. Radiographic and clinical outcomes after 18 months from the surgery (Fondazione Policlinico Universitario Agostino Gemelli IRCCS, 2023)

preparing the articular surfaces and removing talar necrotic residues fragments, the femoral head allograft was implanted. The VALOR™ Ankle Fusion Nail System, designed by Wright Medical Group, was used for the TTC arthrodesis under fluoroscopy, with a time surgery of about 2 hours, and 300cc of blood loss.

Immediately after surgery, the patient started physiotherapy exercises aimed to recover muscle tone-trophism. After 2-days, she was discharged in good clinical conditions with a Walker ankle brace and a 60-days weight-bearing restriction.

The patient returned to the outpatient clinic after 2 weeks for the clinical examination of the surgical wounds. Subsequently, clinical, and radiographic follow-up was performed to one, three, six, twelve and eighteen months after surgery.

At the last follow-up the patient was completely satisfied, and X-rays showed progressive integration of the bone allograft and its fusion, proximally with the tibia and distally with the calcaneus. After almost two years, the patient had no pain while walking (AOFAS score of 56), and she was able to return to daily activities without any restrictions (Figure 5). There were no complications. Only a difference of about 1 cm in the lower limbs' length was observed, but it was considered non clinically significant.

DISCUSSION

After an initial diagnosis of pulmonary TB and Pott's disease, a talar disruption and bone reabsorption has occurred. Imaging and cultural examination confirmed the diagnosis of talar TB, and a TTC arthrodesis was performed in order to reduce pain, and to improve patient's quality of life.

The tubercular disease of the talus is an extremely rare event, only 26 studies are reported with a total of 153 cases with an equal distribution among genders with (77 males and 76 females) (9-50). Furthermore, data from the literature showed that TB of the talus can involve any age, ranging from 6 months to 85 years old (9,10).

The onset of the disease started with common symptoms, such as sudden pain and swelling, history of absence of trauma, and tenderness, which is related to a decrease of range of motion and functional disability (11). For this reason, osteoarticular TB often represents a diagnostic challenge: non-specificity of symptoms and inconclusive diagnostic, and laboratory tests cause diagnostic delay. For this way, Choi et al. (9) refers to talar TB as a "diagnostic dilemma" because they confirmed the biopsy's importance, reporting their study about 15 patients with non-specific symptoms, whose TB of the talus

was misdiagnosed until the final biopsy; diagnostic delay ranges from 3 to 24 months. The early diagnosis prevents disease progression to the adjacent bones and joints, thereby reducing the amount of the tissue destruction. Also, diagnostic delay might cause poor clinical results, and misdiagnosis (12). To obtain a correct and early diagnosis, X-ray in most cases showed osteolytic lesion (13), while MRI and CT were more useful to rule out other confounding pathologies (14–18), to study the spread of the disease into soft tissues and adjacent bones, and to detect the in-volvement of unusual sites (19,20). In our experience, X-ray, CT and MRI of the foot and ankle showed destruction of the tibiotalar joint, talus resorption, multiple abscess collections, and diffuse spongious bone edema in the hindfoot and midfoot.

Concerning MRI, although it is a highly sensitive in detecting soft tissue changes, there is a limited experience (21).

Histological examination usually showed a granuloma with central caseating necrosis, allowing a talar TB certain diagnosis (14); we obtained the same result.

Conservative or surgical treatment can be considered to arrest the progression of the disease and treat its symptoms. In most cases, the conservative approach represents the primary therapeutic choice, aiming to achieve a good clinical response through the administration of a multidrug anti-TB therapy and ankle immobilization with protected weight bearing (7, 13, 21, 41, 46, 48, 49, 50).

Several surgical options are also described in literature. Soft-tissues and talus debridement and curettage were performed in 76 cases, as the only treatment used or associated with more demolitive approaches. For example, Pereirasamy et al.(22) performed a talonavicular joint debridement and subsequent arthrodesis in a patients who presented partial destruction of the talus head, but preservation of the articular cartilage; Halwai et al. (23) used a transmalleolar approach to perform an osteotomy of the medial malleolus and an accurate debridement of lesions. Thereafter, the malleolus was replaced into its normal position and fixated with a screw.

Differently, Jockheck et al. (24) opted for a partial astragalectomy along with tibiocalcaneal fusion using the Charnley fixator. Instead, cases of extended talus destruction were treated with a total astragalectomy (25,26). In the first case, a smooth pin tibiocalcaneal joint fixation was performed. In the second case, the defect was restocked with a cement spacer, and sub-sequently the joint was transfixed with a tibiocalcaneal–metatarsal external fixator which was removed six months later after fusion. Furthermore, Stuart and al.(27) performed a TTC ar-throdesis with screws, Yoshida et al. (28) with intramedullary nail, and Gavaskar and Chow-dary with supracondylar femoral nail (29).

Therefore, data extracted from the literature demonstrate the surgery is required in many cases, especially those diagnosed late and unresponsive to antibiotic therapy alone. In most cases, free pain and complete resolution of the disease were reported. Only few complications were documented: one case of pseudarthrosis, one case of a 4 cm lower limb length discrepancy, and one case of limited range of motion in ankle dorsiflexion.

In our case, we decide to perform a tibiocalcaneal arthrodesis due to the complete destruction of the tibiotalar joint, the significant bone loss of the talus and the severe varus deformity being an effective procedure in case of severe chronic joint disease, severe deformities, chronic degenerative ankle instability and bone defects (30–36).

In conclusion, tibiotalocalcaneal arthrodesis with bone graft showed good functional results in this case report, with complete graft fusion and recovery of ankle and hindfoot deformity.

The study has many important limitations such as the lack of studies in the literature, the small and heterogeneous group of patients, and the absence of a unique type of treatment. Despite these limitations, tibiotalocalcaneal arthrodesis with bone graft has proved to be a safe and efficacy strategy for the treatment of severe talar tuberculosis, showing good clinical, functional and radiological outcomes.

FUNDING

No specific funding was received for this study.

TRANSPARENCY DECLARATION

Competing interests: None to declare.

REFERENCES

1. Barberis I, Bragazzi NL, Galluzzo L, Martini M. The history of tuberculosis: from the first historical records to the isolation of Koch's bacillus. J Prev Med Hyg 2017; 58:E9–12.
2. Kulchavenya E. Extrapulmonary tuberculosis: are statistical reports accurate? Ther Adv Infect Dis 2014; 2:61–70.
3. Schirmer P, Renault CA, Holodniy M. Is spinal tuberculosis contagious? Int J Infect Dis 2010; 14:e659-66.
4. Samuel S, Boopalan PRJVC, Alexander M, Ismavel R, Varghese VD, Mathai T. Tuberculosis of and around the ankle. J Foot Ankle Surg 2011; 50:466–72.
5. Dhillon MS, Aggarwal S, Prabhakar S, Bachhal V. Tuberculosis of the foot: An osteolytic variety. Indian J Orthop 2012; 46:206–11.
6. Dhillon MS, Nagi ON. Tuberculosis of the foot and ankle. Clin Orthop 2002; 398:107–13.
7. Boussouga M, Tanane M, Bousselmame N, Lazrak K, Taobane H. Tuberculosis of the talus: a rare localization of Koch's bacillus. Rev Chir Orthop 2002; 88:522–5.
8. Leigheb M, Janicka P, Andorno S, Marcuzzi A, Magnani C, Grassi F. Italian translation, cultural adaptation and validation of the «American Orthopaedic Foot and Ankle Society's (AOFAS) ankle-hindfoot scale». Acta Bio-Medica Atenei Parm 2016; 87:38–45.
9. Choi WJ, Han SH, Joo JH, Kim BS, Lee JW. Diagnostic dilemma of tuberculosis in the foot and ankle. Foot Ankle Int 2008; 29:711–5.
10. Gursu S, Yildirim T, Ucpinar H, Sofu H, Camurcu Y, Sahin V, Sahin N. Long-term follow-up results of foot and ankle tuberculosis in Turkey. J Foot Ankle Surg 2014; 53:557–61.
11. Pereirasamy L, Singh NG, Nijhar BK, Rao M, Ali IAH. Isolated tuberculosis of the talus bone. Chest 2012; 142:57A.
12. Hanène F, Nacef L, Maatallah K, Triki W, Kaffel D, Hamdi W. Tuberculosis arthritis of the ankle mimicking a talar osteochondritis. Foot (Edinb) 2021; 49:101816.
13. Karkhur Y, Tiwari V, Lodhi J, Tiwari A. Astragalus Tuberculosis: A case report and review of the literature. Cureus 2017; 9:e1708.
14. Faroug R, Psyllakis P, Gulati A, Makvana S, Pareek M, Mangwani J. Diagnosis and treatment of tuberculosis of the foot and ankle—a literature review. Foot (Edinb) 2018; 37:105–12.
15. Greco T, Cianni L, De Mauro D, Dughiero G, Bocchi MB, Cazzato G, Ragonesi G, Liuzza F, Maccauro G, Perisano C.. Foot metastasis: Current knowledge. Orthop Rev 2020; 12(Suppl 1):8671.
16. Perisano C, Greco T, Vitiello R, Maccauro G, Liuzza F, Tamburelli FC, Forconi F. Mueller-Weiss disease: review of the literature. J Biol Regul Homeost Agents 2018; 32(6 Suppl. 1):157–62.
17. Perisano C, Vitiello R, Greco T, Cianni L, Maccauro G, Sfregola S, Gessi M, Mancino F. A report of a very rare isolated bone metastasis in the midfoot due to cervix cancer. Minerva Ortop E Traumatol 2019; 70(2).
18. Hugosson C, Nyman RS, Brismar J, Larsson SG, Lindahl S, Lundstedt C. Imaging of tuberculosis. V. Peripheral osteoarticular and soft-tissue tuberculosis. Acta Radiol 996; 37:512–6.
19. Muratori F, Pezzillo F, Nizegorodcew T, Fantoni M, Visconti E, Maccauro G. Tubercular osteomyelitis of the second metatarsal: a case report. J Foot Ankle Surg 2011; 50:577–9.
20. Piscitelli L, Bisaccia M, Meccariello L, Falzarano G, Medici A, Maiettini D, Rebonato A, Rinonapoli G, Caraffa A. The emergency and delay management in total talus extrusion: Case report and review of literature after 24 months of follow up. J Acute Dis 2016;5.
21. Prakash M, Gupta P, Sen RK, Sharma A, Khandelwal N. Magnetic resonance imaging evaluation of tubercular arthritis of the ankle and foot. Acta Radiol 2015; 56:1236–41.
22. Halwai MA, Mir BA, Dhar SA, Dar TA, Butt MF. Transmalleolar approach to a tubercular lytic lesion of the talar body: a case report. J Foot Ankle Surg 2011; 50:490–3.
23. Jockheck M, Meeder PJ, Weller S. Tuberculous destruction of the talus--a rare form of extrapulmonary skeletal TBC--a case report. Z Orthop Ihre Grenzgeb 1992; 130:163–5.
24. Ebrahimzadeh MH, Sadri E. Isolated tuberculosis of the talus bone. Arch Iran Med 2006; 9:159–60.
25. Müller F, Zellner M, Füchtmeier B. Treatment and outcome of destructive tuberculosis of the talus. Foot Ankle Surg 2015; 21:e45-47.
26. Stuart D. Local osteo-articular tuberculosis complicating closed fractures: report of two cases. J Bone Joint Surg Br 1976; 58:248–9.
27. Yoshida T, Sakamoto A, Iwamoto Y. Vascularized iliac bone graft in cases of ankle tuberculosis. J Reconstr Microsurg 2009; 25:125–31.
28. Gavaskar AS, Chowdary N. Tibiotalocalcaneal arthrodesis using a supracondylar femoral nail for advanced tuberculous arthritis of the ankle. J Orthop Surg Hong Kong 2009; 17:321–4.
29. Escudero MI, Poggio D, Alvarez F, Barahona M, Vivar D, Fernandez A. Tibiotalocalcaneal arthrodesis with distal tibial allograft for massive bone deficits in the ankle. Foot Ankle Surg 2019; 25:390–7.
30. Greco T, Polichetti C, Cannella A. Ankle hemophilic arthropathy: literature review. 11:206-16.
31. Cianni L, Bocchi MB, Vitiello R, Greco T, De Marco D, Masci G, Maccauro G, Pitocco D, Perisano C. Arthrodesis in the Charcot foot: a systematic review. Orthop Rev 2020; 12(Suppl 1):8670.
32. Mosca M, Caravelli S, Fuiano M, Massimi S, Censoni D, Grassi A, Vocale E, Ceccarelli F, Zaffagnini S. Tibiotalocalcaneal arthrodesis through retrograde nailing for the treatment of juxtaarticular distal tibia aseptic non-unions: A retrospective study at a minimum follow-up of 4 years. Injury 2020; 51:1377–81.
33. Greco T, Fulchignoni C, Cianni L, Maccauro G, Perisano C. Surgical management of tibial metastases: a systematic review. Acta Biomed Atenei Parm 2022; 92:e2021552.
34. Mendicino RW, Catanzariti AR, Saltrick KR, Dombek MF, Tullis BL, Statler TK, Johnson BM. Tibiotalocalcaneal arthrodesis with retrograde intramedullary nailing. J Foot Ankle Surg 2004; 43:82–6.

35. Perisano C, Cannella A, Polichetti C, Mascio A, Comisi C, De Santis V, Caravelli S, Mosca M, Spedicato GA, Maccauro G, Greco T. Tibiotalar and tibiotalocalcaneal arthrodesis with Paragon28 SilverbackTM Plating System in patients with severe ankle and hindfoot deformity. Medicina (Mex) 2023; 59:344.
36. Dahuja A, Dahuja G, Kaur R, Bansal K. Isolated tuberculosis of talus: a case report. Malays Orthop J 2014; 8:61–2.
37. Halwai MA, Mir BA, Dhar SA, Dar TA, Butt MF. Transmalleolar approach to a tubercular lytic lesion of the talar body: a case report. J Foot Ankle Surg Off Publ Am Coll Foot Ankle Surg 2011; 50:490–3.
38. Anand A, Sood LK. Isolated tuberculosis of talus without ankle and subtalar joint involvement. Med J Malaysia 2002; 57:371–3.
39. Anderson JR, Ritchie AW, Scobie WG. Tuberculous osteitis of the talus. Tubercle 1979; 60:115–8.
40. Göksan A, Yazicioglu Ö, Özger H, Temelli Y, Tözün R, Türkmen M. Isolated bone tuberculosis of the talus. Case report. Z Für Orthop Ihre Grenzgeb 1984; 122:657–8.
41. Jockheck M, Meeder PJ, Weller S. Tuberculous destruction of the talus--a rare form of extrapulmonary skeletal TBC--a case report. Z Orthop Ihre Grenzgeb 1992; 130:163–5.
42. Mardanpour K, Rahbar M. Isolated tuberculosis of the talus bone. Casp J Intern Med 2010; 1:75–7.
43. Abbaoui S, Hassan HE, Belahcen M. Tuberculosis of talus: A case report. Int J Surg Case Rep 2022; 95:107208.
44. Haraldsson S. Osteitis tuberculosa fistulosa following vaccination with E.C.G. strain. Acta Orthop Scand 1959; 29:121–33.
45. Baquero-Artigao F, Garcia-Miguel MJ, Merino R, García-Consuegra J, del Castillo F. Multidrug-resistant tuberculosis of the ankle: case report. Foot Ankle Int 2006; 27:642–4.
46. Sahoo SS, Tiwari V, Velagada S. A rare case of tuberculosis as a cause of lytic lesion of talus without adjacent bone involvement in a four-year-old child. Cureus 2021; 13:e16909.
47. Brew CJ, Rao V, Shanker J. Tuberculosis infection of the talonavicular joint. Foot Edinb Scotl 2010; 20:146–8.
48. Korim M, Patel R, Allen P, Mangwani J. Foot and ankle tuberculosis: case series and literature review. Foot Edinb Scotl 2014; 24:176–9.
49. Agarwal A, Qureshi NA, Khan SA, Kumar P, Samaiya S. Tuberculosis of the foot and ankle in children. J Orthop Surg Hong Kong 2011; 19:213–7.