Case Report

Late Onset Atypical Fracture Resulting from a Neglected Tibia Malunion: A 45-Year Case Study

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ABSTRACT

Introduction

Neglected or inadequately treated fractures can lead to complications such as malunion, non-union, deformities, and fractures under atypical conditions that mimic pathological fractures due to biomechanical alterations and reduced bone density. Despite numerous case studies, the long-term consequences of neglected tibial malunions remain underexplored. This case report delves into the outcomes of a neglected tibia fracture malunion manifesting after an extended period.

Case

A 65-year-old male presented with severe shin pain and tingling, rendering him unable to walk. The patient had no history of recent falls, trauma, or accidents. However, his medical history included a tibia fracture from a motorcycle accident 45 years earlier, which was treated solely by traditional bone setters. This initial neglect resulted in a malunion, causing a leg length discrepancy and instability. During examination, a deformity on the right shin, tenderness, visible leg length asymmetry, and limited range of motion were noted. The patient's bone mineral density (BMD) was measured at -2.8. X-rays indicated a late-onset atypical fracture associated with malalignment and significantly reduced bone density.

Conclusion

This case highlights the severe complications that can arise from neglected malunion fractures, including atypical fractures resembling pathological conditions, which are primarily due to biomechanical changes rather than disease. It underscores the importance of timely medical intervention and illustrates the risks associated with reliance on traditional bone-setting methods. This case is particularly instructive for trauma care in developing nations, where such practices are more common and often go unaddressed.

Keywords

neglected fracture; malunion; atypical fracture; long-term consequences; case report.

INTRODUCTION

Tibial fractures are prevalent injuries affecting the long bones, making up around 2% of fractures in adults [1]. A comprehensive investigation of tibial shaft fractures using trauma data revealed a frequency of 16.9 per 100,000 individuals, with two notable peaks in occurrence at ages 20 and 50 [2]. The likelihood of experiencing such fractures is slightly elevated in males, with an annual incidence of 21.5 per 100,000, compared to females at 12.3 per 100,000 [3]. Fractures that are neglected or inadequately treated can give rise to a variety of complications, including malunion, non-union, deformities, and even pathological fractures, and these cases in developing nations often arise due to the prevalent practice of traditional bone setting [4,5]. Timely and appropriate medical intervention, involving realignment and stabilization of fractured bones, is imperative to avoid these outcomes. When paired with prompt soft tissue care, intramedullary nailing has been shown to be an efficient early fixing method for open tibial fractures, lowering infection rates, accelerating healing, and guaranteeing biomechanical stability [6].

Neglected fractures can lead to substantial functional impairments that have far-reaching consequences for a patient's quality of life. The cumulative effects of such fractures over

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time can result in severe pain, reduced mobility, and compromised daily activities [5].

The purpose of this case report is to provide an in-depth analysis of this neglected tibia fracture malunion, leading to a pathological fracture. The extended interval between the initial injury and the current pathological fracture, spanning 45 years, emphasizes the rarity and significance of this case. This paper been reported in line with the SCARE criteria [7].

Case Presentation

A 65-year-old male presented at the orthopedic clinic with chief complaints of persistent pain and tingling in the shinbone of his right leg, symptoms that had been worsening over the past 7 days. He reported a numb sensation extending from the shin area to the ankle, accompanied by an inability to move his right toes. This debilitating pain escalated significantly over the last 3 days, leaving the patient unable to walk and confined to sitting or lying down at home. The patient denied any recent falls, trauma, or accidents and had no history of diabetes mellitus (DM) or hypertension (HT).

Figure 1: Visual Representation of the Patient's Right Leg Condition Prior to Hospital Admission.

This image illustrates the significant deformity and leg length discrepancy resulting from the long-standing malunion of the tibia. Noticeable are the abnormal angulation and asymmetry compared to the unaffected leg, highlighting the severity of the neglected condition (Courtesy of Basuki).

Medical History and Initial Findings

The patient's medical history was notable for a motorcycle accident occurring 45 years ago, which resulted in a fracture of the right tibia. Unfortunately, the fracture was only treated by traditional bone setters, resulting in a malunion. This improper healing caused a leg length discrepancy and noticeable deformity, leading to instability and pain during extended walks. Despite these challenges, the patient remained exceptionally active, adapting to the structural limitations imposed by his condition. "This condition was left unaddressed and unmanaged medically for 45 years, during which no formal orthopedic care was sought or provided.

Physical Examination

Upon examination, the patient was found conscious and oriented, with vital signs within normal limits: blood pressure 110/70 mmHg, heart rate 86 bpm, temperature 36.8°C, and oxygen saturation 98% on room air. Local examination of the lower extremities showed tenderness at the site of the right tibia fracture, accompanied by visible leg length discrepancy—his right leg being noticeably shorter than his left (Figure 2). Measurements of the true and apparent leg lengths confirmed these discrepancies. Additionally, a deformity was evident on the right shin. There were no signs of swelling, redness, bone exposure, or pus, but the dorsalis pedis artery was palpable. The range of motion (ROM) in the right leg was severely limited due to pain, in contrast to the normal ROM observed in the left leg.

This image shows the deformity in the patient's right leg, which is notably shorter than the left leg, highlighting the extent of the malunion (Courtesy of Basuki).

Diagnostic Findings

Laboratory tests showed normal hemoglobin levels (15.2 g/dL), leukocyte count (7.9 × 10^9/L), hematocrit (44.8%), and platelet count (310,000/μL). The erythrocyte sedimentation rate was low at 9 mm/hour, the neutrophil-to-lymphocyte ratio stood at 2.35, and the absolute lymphocyte count was 2.212 × 10^9/L. Quantitative C-reactive protein was measured at 4.72 mg/L, indicating no underlying

Figure 2: Comparison of Leg Lengths of the Patient at the Orthopedic Polyclinic.

infection or inflammation. X-ray imaging revealed an aged, malaligned fracture at the mid-diaphysis of the right tibia, characterized by callus formation, significant reduction in bone density, and trabeculation, suggesting long-term biomechanical changes and bone deterioration (Figure 3).

Treatment and Outcome

The treatment of the patient was comprehensive and conducted in multiple stages. Initially, the malunion was addressed through reconstruction surgery, which was followed by Open Reduction and Internal Fixation (ORIF) and a bone graft. Post-surgical care included administration of Ketorolac 2x30mg for pain

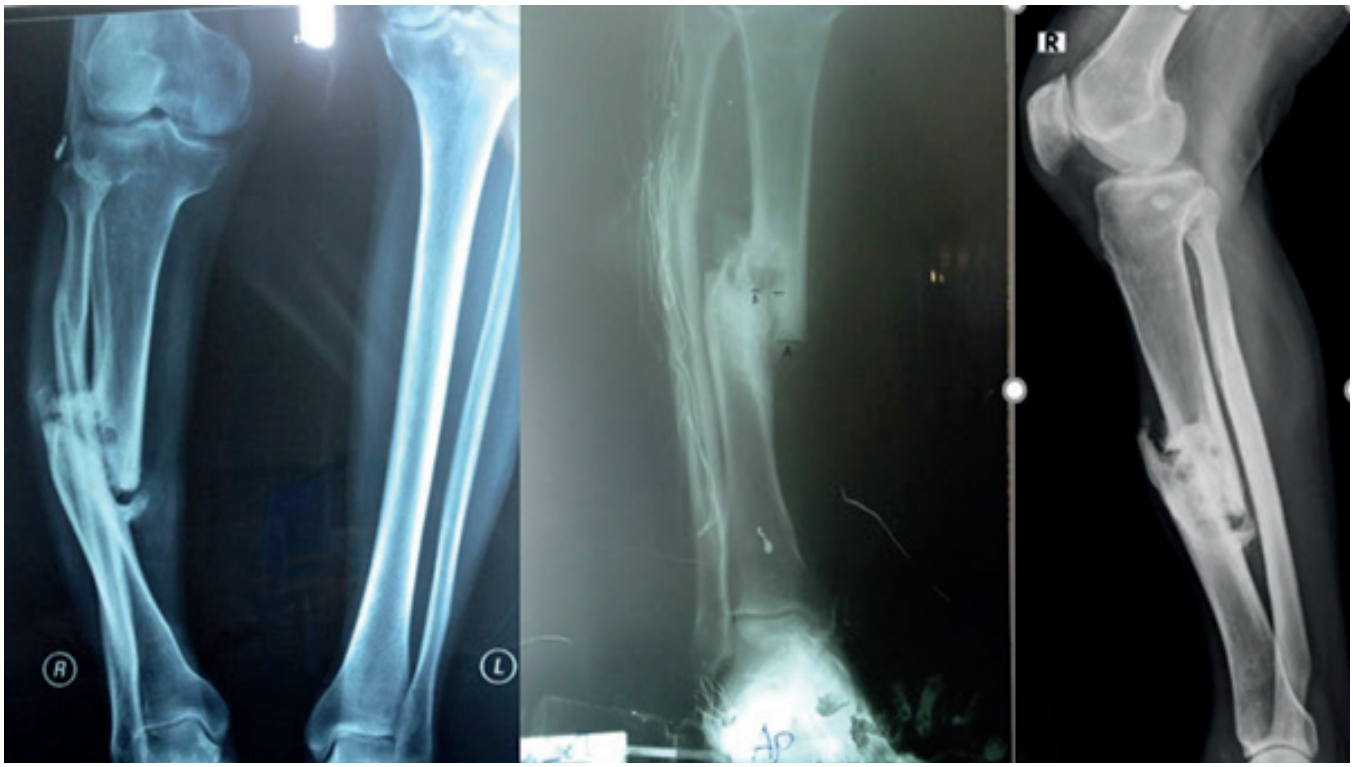

Figure 3: X-ray Imaging of the Patient's Lower Legs.

This image set displays three views: the left panel shows both the right (dextra) and left (sinistra) cruris for comparison; the middle and right panels focus on different angles of the right cruris, highlighting the malalignment and bone density changes associated with the chronic malunion (Courtesy of Basuki)."

Figure 3: X-ray Imaging of the Patient's Lower Legs.

A) shows both the right (dextra) and left (sinistra) cruris for comparison; B-C) focus on different angles of the right cruris, highlighting the malalignment and bone density changes associated with the chronic malunion (Courtesy of Basuki).

relief, Meropenem 1x1 g for antibiotic prophylaxis, a Neurobion injection, and three daily doses of Vitamin C. Notable improvement in the patient's condition was observed three days after the second surgery, as documented in Figure 4. The surgical wound pain decreased significantly, and the sensations of pain and tingling subsided. While the patient regained the ability to walk, he required a cane for full weight-bearing support due to the ongoing recovery process. After discharge, the patient was prescribed a regimen that included cefixime (200 mg twice daily), methylcobalamin (1 mg twice daily), and paracetamol (1 g three times daily) to manage pain and prevent potential infections. A follow-up appointment was scheduled at the orthopedic clinic one week after discharge to assess the patient's progress and ensure proper healing."

Figure 4: The AP (Anteroposterior) and Lateral X-ray Views of the Right Cruris Post-ORIF. This image demonstrates the alignment and stabilization achieved through the Open Reduction and Internal Fixation procedure. (Courtesy of Basuki).

DISCUSSION

Tibial fractures exhibit a bimodal pattern involving both low-energy and high-energy mechanisms. Low-energy injuries are often due to twisting forces, indirect trauma leading to spiral fractures, or a fibular fracture at a different level with minimal damage to soft tissues. High-energy mechanism injuries usually result from direct trauma causing wedge or short oblique fractures, often with significant comminution. These fractures can be accompanied by soft tissue injuries, compartment syndrome, loss of bone substance, and fractures in nearby bones on the same side [8,9].

What distinguishes the case presented in this report is the patient's prolonged experience of living with a malunion from a neglected fracture for an extraordinary duration of 45 years. Despite the considerable biomechanical disadvantages and limitations in daily activities, the patient adapted to live without medical correction until significant complications arose. Demonstrating expertise in trauma management principles has been proven to effectively lower mortality and morbidity rates associated with trauma [10], with some studies reporting improvements of up to 90% [11]. Nevertheless, the application of these principles is inconsistent, particularly in developing nations where barriers to healthcare access and quality often result in neglected traumas. This neglect can lead to severe long-term complications, reduced quality of life, and diminished economic productivity, placing a substantial burden on the healthcare systems and economies of these countries [12].

This case underscores the critical need for comprehensive trauma care frameworks that integrate timely intervention and rehabilitation. It highlights the necessity for ongoing education of healthcare providers in trauma care principles and the importance of public health initiatives to prevent neglect and manage the consequences more effectively.

Neglected fractures pertain to fractures left untreated or improperly managed, resulting in treatment delays, worsened conditions, and even disability [13]. In this case, the patient's initial fracture care was managed solely by traditional bone setters, which is a practice fraught with significant risks. Traditional bone setting lacks the diagnostic precision of X-rays for proper alignment, frequently employs non-sterile methods, and might even involve rebreaking bones that have already healed incorrectly. These practices can lead to severe complications, such as 'traditional bone setter's gangrene,' among other serious conditions. Studies conducted in Indonesia and similar settings have documented these risks, illustrating the grave consequences of relying exclusively on traditional bone

setting methods without integrating standard medical practices [14,15,16].

The patient described in our report suffered from a significant complication known as malunion, a condition that aligns with the findings of Warman et al. [17], who identified malunion as the most prevalent complication arising from neglected fractures. Additional complications include delayed issues such as avascular necrosis, chronic osteomyelitis, malunion, non-union, joint stiffness, delayed union, secondary osteoarthritis, muscle atrophy, and Volkmann contracture. Tibial malunions involve the healing of tibial fractures in misaligned positions, leading to deformities and potential complications like pain and walking disturbances.

What distinguishes our patient's case is the extraordinary duration—45 years—over which he lived with this malunion, and the eventual development of an atypical fracture at the age of 65. While it is possible for some malunions to remain asymptomatic for years. Delays in medical attention during mass casualty occurrences impact the severity of injuries, highlighting the vital need for structured trauma care systems to meet urgent demands and avoid long-term problems [18]. As seen in this patient, continuous neglect often leads to severe, irreversible complications. This case highlights the critical importance of addressing fractures promptly and adequately to prevent the long-term detrimental effects that can significantly impair quality of life [19].

After enduring the malunion for 45 years, the patient, at the age of 65, experienced a sudden fracture in the absence of any specific trauma. This incident underscores how neglected malunions can alter the biomechanics of the bone, subjecting it to unusual stresses and strains [20]. Over time, this altered biomechanics combined with decreased bone density, especially evident in aged individuals, can lead to increased susceptibility to pathological fracture [21]. Typically, pathological fractures result from weakened bone structures due to various causes such as tumors, osteoporosis, infections, or previously untreated injuries. With advancing age, the physiological decline in organ function contributes to decreasing bone mass, heightening the risk of osteoporosis [22,23]. Patterns of damage may be impacted by significant biomechanical changes. In order to reduce the risk of progressive fractures, this highlights the significance of a multidisciplinary approach in the diagnosis and treatment of malunions [24].

In our patient, the cause was multifactorial. The long-standing neglected malunion predisposed the tibia to altered stresses, and the aging factor further exacerbated this. X-ray findings confirmed a significant reduction in bone density, with a BMD examination indicating a score of -2.8, signifying osteoporosis. When osteoblastic and osteoclastic activity levels are imbalanced, bone resorption occurs, followed by fibrous bone repair. This lowers bone mineral density (BMD) and potentially increases the fracture risk [25].

The patient also reported a tingling and numb sensation extending from the shin area down to the ankle, accompanied by an inability to move his right toes—a condition likely resulting from nerve damage associated with the prolonged neglect of the fracture. These symptoms, which had worsened recently, suggest a progression of the underlying issues leading to severe complications. Following the diagnosis, the patient underwent a straightforward reconstruction procedure to correct the malunion in the right tibia. After this intervention, there was a marked improvement in his condition, illustrating the critical importance of addressing such long-standing orthopedic issues promptly and effectively.

Future research should explore the long-term effects of neglected fractures, particularly focusing on the development of atypical fractures and nerve damage. This case is, to the best of our knowledge, the first documented report of an atypical fracture occurring without associated lesions, such as tumors or infections, following a protracted period of neglect. It underscores the remarkable adaptability some individuals display in managing significant orthopedic issues over extended durations. However, it also starkly highlights the severe long-term consequences of untreated injuries. The reliance on traditional bone-setting practices can lead to dire outcomes. The importance of timely and appropriate medical intervention cannot be overstated, especially given the compounded effects of prolonged trauma and the natural reduction in bone density with age. Early diagnosis and treatment of fractures are crucial to prevent severe future complications and to mitigate the effects of aging on bone health.

CONCLUSION

This case report not only underscores the risks associated with untreated fractures but also illustrates the potential for chronic conditions to mimic acute pathological

processes. This report presents a distinctive case of a patient who lived with a neglected malunion of a tibial fracture for 45 years, culminating in an atypical fracture at age 65 without any recent trauma. This case vividly illustrates the dangers associated with untreated fractures, especially when compounded by age-related osteoporosis. The absence of initial proper medical care and reliance on traditional bone-setting methods significantly contributed to the patient's long-term complications. This case highlights the critical need for timely medical intervention and illustrates the shortcomings of relying solely on traditional practices without the benefits of modern medical oversight.

This case report contributes significantly to the existing literature by documenting a rare instance of a neglected tibial fracture malunion that developed into an atypical fracture after 45 years without preceding trauma. It highlights the critical long-term consequences of inadequate initial treatment and the potential pitfalls of relying solely on traditional bone-setting methods. The case underscores the importance of timely, appropriate orthopedic care and provides a compelling argument for the integration of modern medical techniques even in regions where traditional practices are prevalent. Furthermore, it sheds light on the biomechanical changes that can occur from prolonged neglect, contributing to our understanding of how chronic conditions evolve and the importance of addressing orthopedic issues promptly to prevent severe complications. By detailing this unique case, the report adds valuable insights into the management and potential outcomes of similar cases, emphasizing the need for comprehensive care and follow-up.

This case report has several limitations that should be considered when interpreting the findings. As a single case study, the generalizability of the results is limited, and the conclusions drawn may not be applicable to a broader population without further research. The retrospective reporting over a prolonged period of 45 years may introduce inaccuracies due to potential gaps in historical medical records and patient recall. Additionally, the lack of a control or comparative group restricts our ability to definitively attribute outcomes directly to the care methods employed or compare the effectiveness of different treatment strategies. The reliance on subjective patient reporting for symptoms and outcomes also introduces potential bias. Moreover, the study does not include long-term follow-up data post-treatment, limiting our understanding of the enduring impacts of the interventions. Finally, external factors such as the patient's overall health, lifestyle, and adherence to post-treatment recommendations, which could significantly influence outcomes, were not comprehensively controlled for or detailed in the study.

Future research should include larger studies, longitudinal evaluations, and comparative analyses to deepen our understanding of the long-term effects of neglected fractures. Enhancing our knowledge and refining trauma management protocols can significantly improve patient outcomes, emphasizing the importance of integrating traditional practices with contemporary medical standards.

Authors' contributions

Concept: B.S., H.M.

Design: B.S., H.M.

Data Collection or Processing: B.S., H.M.,

Analysis or Interpretation: B.S., H.M.,

Literature Search: B.S., H.M.,

Writing: B.S., H.M.,

Conflict of interest

The authors have no conflict of interest with the objectives of this study.

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REFERENCES

1. Laurila J, Huttunen TT, Kannus P, Kääriäinen M, Mattila VM. Tibial shaft fractures in Finland between 1997 and 2014. *Injury* [Internet]. 2019;**50**(4):973–7. Available from: https://www.sciencedirect.com/science/article/pii/S0020138319301342
2. Anandasivam NS, Russo GS, Swallow MS, Basques BA, Samuel AM, Ondeck NT, et al. Tibial shaft fracture: A large-scale study defining the injured population and associated injuries. *J Clin Orthop Trauma* [Internet]. 2017;**8**(3):225–31. Available from: https://www.sciencedirect.com/science/article/pii/S097656621730245X
3. Larsen P, Elsoe R, Hansen SH, Graven-Nielsen T, Laessoe U, Rasmussen S. Incidence and epidemiology of tibial shaft fractures. *Injury* [Internet]. 2015;**46**(4):746–50. Available from: https://www.sciencedirect.com/science/article/pii/S0020138314006743
4. Kostenuik P, Mirza FM. Fracture healing physiology and the quest for therapies for delayed healing and nonunion. *J Orthop Res.* 2017 Feb;**35**(2):213–23.
5. Musthofa FD, Suhanto S, Marjono T. Neglected fracture tibia dextra with gangrene pedis, post treatment by traditional bone setter. JKKI : *Jurnal Kedokteran Dan Kesehatan Indonesia*. 2018;**9**(3):202–6.
6. Islam MS, Karim MR, Rahim ZZ. Evaluation of the treatment of Open Fracture of Tibia by Primary Nailing. *Bangladesh Journal of Medical Science*. 2024 Jul 29;**23**(3):638–42.
7. Agha RA, Borrelli MR, Farwana R, Koshy K, Fowler AJ, Orgill DP, et al. The SCARE 2018 statement: Updating consensus Surgical CAse REport (SCARE) guidelines. *International Journal of Surgery* [Internet]. 2018;60:132–6. Available from: https://www.sciencedirect.com/science/article/pii/S1743919118316716
8. Ping Song, Lee L Q Pu. The Soleus Muscle Flap: An Overview of Its Clinical Applications for Lower Extremity Reconstruction. *Ann Plast Surg*. 2018;109–16.
9. Bezstarosti H, Van Lieshout EMM, Voskamp LW, Kortram K, Obremskey W, McNally MA, et al. Insights into treatment and outcome of fracture-related infection: a systematic literature review. *Arch Orthop Trauma Surg [*Internet]. 2019;**139**(1):61–72. Available from: https://doi.org/10.1007/s00402-018-3048-0
10. Moore K. Injury Prevention and Trauma Mortality. *J Emerg Nurs* [Internet]. 2016;**42**(5):457–8. Available from: https://www.sciencedirect.com/science/article/pii/S0099176716301234
11. Byun CS, Park IH, Oh JH, Bae KS, Lee KH, Lee E. Epidemiology of Trauma Patients and Analysis of 268 Mortality Cases: Trends of a Single Center in Korea. *Yonsei Med J* [Internet]. 2015 Jan;**56**(1):220–6. Available from: https://doi.org/10.3349/ymj.2015.56.1.220
12. Galo' HF, Rawung R, Prasetyo E. Faktor-faktor yang Berhubungan dengan Kejadian Neglected Fracture pada Pasien di RSUP Prof Dr. R. D. Kandou Manado pada Periode Januari-Desember 2018. E-CliniC. 2020;**8**(1).
13. Solomon L, Warwick D, Nayagam S. Apley's system of orthopaedics and fractures. CRC Press; 2017.
14. Luhur LJ, Dharmawan IPG, Wiguna IGLNAA. A catastrophic presentation of dead limb secondary to traditional bone setter's treatment in Sorong, West Papua: A case report. *Int J Surg Case Rep* [Internet]. 2021;84:106061. Available from: https://www.sciencedirect.com/science/article/pii/S2210261221005630
15. Hantonius H, Rasyid HN, Alpharian GT. Orthopaedic Emergency Cases at Hasan Sadikin Hospital Bandung Caused by Traditional Bone Setter Practice. *Journal Orthopaedi and Traumatology Surabaya.* 2018;**7**(1):77–84.
16. Worku N, Tewelde T, Abdissa B, Merga H. Preference of Traditional Bone Setting and associated factors among trauma patients with fracture at Black Lion Hospital in Addis Ababa, Ethiopia: institution based cross sectional study. *BMC Res Notes* [Internet]. 2019;**12**(1):590. Available from: https://doi.org/10.1186/s13104-019-4643-z
17. Warman PL, Ismiarto YD, Ruhimat U. Complications of Fracture Treatment by Traditional Bonesetters in West Java, Indonesia. *Althea Medical Journal* [Internet]. 2018;5:47–52. Available from: https://api.semanticscholar.org/CorpusID:59179714
18. Nor MA, Sonmez RE, Hassan MY, Ozsoy MS, Alimoglu O. Patterns of injury and death in mass casualty incidents from 2013 to 2018 in Mogadishu, Somalia Mass casualty incidents in Mogadishu, Somalia. *Bangladesh Journal of Medical Science*. 2023 Apr 11;**22**(2):323–8.
19. Patel I, Young J, Washington A, Vaidya R. Malunion of the Tibia: A Systematic Review. Medicina (Kaunas). 2022;**58**(3).
20. Patel B, Gustafson PA, Jastifer J. The effect of clavicle malunion on shoulder biomechanics; A computational study. *Clinical Biomechanics* [Internet]. 2012;**27**(5):436–42. Available from: https://www.sciencedirect.com/science/article/pii/S0268003311003196
21. Nguyen DT, Burg KJ. Bone tissue engineering and regenerative medicine: targeting pathological fractures. *Journal of Biomedical Materials Research Part A* . 2015;**103**(1):420–9.
22. Rizzo SE, Kenan S. Pathologic Fractures. StatPearls [internet]: StatPearls Publishing; 2023.
23. Resnasari SD, Supartono B, EkaPurwani L. The Correlation Between Low Body Mass Index (Underweight) With Bone Strength On Elderly Women. *Jurnal Saintika Medika*. 2020 Jun;**16**(1).
24. Ghaffari S, Azar MS, Kariminasab MH, Ranjbar T, Ghaffari B. The frequency of proximal tibiofibular joint types in patients with malleolar fractures. *Bangladesh Journal of Medical Science.* 2018 Sep 19;**17**(4):625–30.
25. Ali MH, Zulkeflee HA, Mohamed WMIW, Ismail TST. Pathological fractures in teen and middle age women with primary hyperparathyroidism. *Bangladesh Journal of Medical Science.* 2023 Jan 1;**22**(1):234–8.