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Case Report

Osteoid osteoma - case report and literature review

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Abstract

Osteoid osteoma (OO) is a benign osteoblastic bone tumor, most frequently affecting young male patients. Due to its rarity and nonspecific symptomatology, the OO diagnosis in the foot can be postponed and confused with diseases leading to the absence of the correct diagnosis for years. In this report, we present a 22-year-old male patient diagnosed with OO in the talus. His diagnosis was delayed for approximately three years, confused, and treated as a complication after an ankle sprain. We also conducted a literature review on OO in the foot and ankle.

Level of Evidence V; Therapeutic Studies; Expert Opinion.

Keywords: Osteoid osteoma; Bone neoplasms; Talus.

Introduction

Osteoid osteoma (OO) is a challenge for orthopedists in early diagnosis and choosing the best treatment. When it affects the foot bones due to its rarity, much of the literature is based on case reports[1].

Osteoid osteoma is defined as a benign osteoblastic bone tumor, and it is characterized by a nidus surrounded by dense and sclerotic bone, rarely greater than 15 mm[1-3]. Responsible for 10% to 14% of benign tumors, this tumor has a higher frequency in young men (between 5 and 25 years old) in a 2:1 ratio[2,4-6]. The most common locations are the metadiaphyseal and diaphyseal region of long bones, particularly the tibia and femur[2,3,5,7-9]. Foot involvement is rare, ranging from 2% to 10%, and the most affected are the talus, followed by the calcaneus[2,10,11]. Although its pathogenesis is unknown, it seems that a high level of prostaglandins is produced in the nidus center resulting in arteriolar vasodilation and edema, which stimulates the nerve terminals, causing pain[1,6,7].

Eideken classified OO according to its location as cortical, cancellous, and subperiosteal[7,10]. These tumors are usually cancellous or subperiosteal in the foot, where the periosteal reaction is minimal or absent[7].

Its clinical presentation frequently is pain, edema, joint stiffness and/or limitation of activities[7,8,10]. Locating near the joint can lead to synovitis, muscle spasms, mimicking arthritis, or trauma[10]. Classically, the pain worsens at night with improvement after administration of non-steroidal anti-inflammatory drugs (NSAIDs) in about 64% of cases[7,11]. Due to its rarity and nonspecific symptomatology, OO can take years to be diagnosed[4].

From the complementary exams, the simple foot radiograph hardly presents alterations due to the absence of periosteal reaction, but when positive, it points to a radiolucent lesion surrounded by bone sclerosis[7]. Magnetic resonance imaging (MRI) presents nonspecific bone edema, leading to diagnostic errors in 33-35% of cases[7]. Bone scintigraphy can be used to locate the tumor due to its high sensitivity[12]. Computed tomography (CT) is superior to MRI in showing the nidus attenuation and central calcification associated with perilesional sclerosis[7,8,12] and is the main imaging test for diagnosis[12,13]. Among the main differential diagnoses are ankle impact syndrome, stress fractures, tenosynovitis, osteomyelitis, osteonecrosis, chronic ankle instability, and inflammatory arthropathy[8,14].

Study performed at the Department of Orthopedics and Traumatology, Santa Casa de Misericórdia de São Paulo, São Paulo, SP, Brazil.

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Treatment can be surgical and non-surgical[3]. Using NSAIDs and waiting for spontaneous resolution is an alternative. However, surgery is indicated when there is no improvement with conservative treatment or when the medication presents side effects or risks. The success of surgical treatment is achieved with nidus resection or destruction[6,15]. Other surgical treatment options are percutaneous radiofrequency ablations, reaching 60-100% success[3,6,15]. When the lesion is located in the ankle joint, excision through arthroscopy is also a viable option[4].

The aim of this report is to present a 22-year-old patient diagnosed with OO on the neck of the talus who had his diagnosis postponed for three years.

Case description

This study was approved by the Human Research Ethics Committee under the number (CAAE 37100514.7.0000.5479). A 22-year-old male farmer sought medical assistance at our service due to severe pain in the right ankle that started after twisting approximately three years prior. The patient reported previous treatment in another service with a plaster cast for 40 days, followed by removable immobilization for another 20 days, but without improvement. His pain began during work, worsening at night during rest, and relief after self-medication with NSAIDs. He sought medical help a few times, but according to the patient, the specific diagnosis was not concluded.

On physical exam, the patient presented pain on palpation of the anteromedial region of the right ankle. There was no edema, the ankle was clinically stable, and the pulses were symmetrical and palpable. He did not have a limited range of motion or decreased muscle strength but he presented pain with forced dorsiflexion, both actively and passively, which worsened when standing at his toes or running. At the time, the patient's function and pain were evaluated using the American Orthopaedic Foot and Ankle Society (AOFAS) scores of the hindfoot and ankle, and visual analog pain scale (VAS) presenting a total of 64 in 100 points and 9 in 10 points respectively.

Bone changes were not detected on a simple ankle radiograph (Figure 1). As the patient had longstanding joint pain, an MRI was requested and showed the lesion and indicated circumjacent medullary edema and joint effusion in the anterior recess of the ankle (Figure 2). A CT was also requested and demonstrated a bone lesion on the neck of the talus of approximately 0.9 cm, well delimited, with perilesional bone sclerosis area and central niche (Figure 3). We hypothesized the OO diagnosis on the neck of the talus and indicated en-block resection surgery.

An open procedure was performed through the anterior route of the ankle. The exposed area presented a wine-colored lesion on the dorsal surface of the talar neck. Ostectomy was performed with the lesion en-block resection (Figures 4 and 5), and the material collected was sent for anatomopathological evaluation. Next, an autologous cancellous bone graft was harvested from the metaphyseal region of the ipsilateral distal tibia to fill the cavity (Figure 6). The anatomopathological report indicated the presence of neoplastic tissue characterized by the proliferation of irregular osteoid beams in their shape and size, surrounded by osteoblasts and interspersed by connective tissue with blood capillaries, proving the OO diagnosis.

After surgery, the patient used suropodalic immobilization without weight-bearing for four weeks, followed by assisted weight-bearing for another four weeks. The follow-up was at two weeks and 1, 2, 4, 8, 12, 24, and 36 months postoperatively.

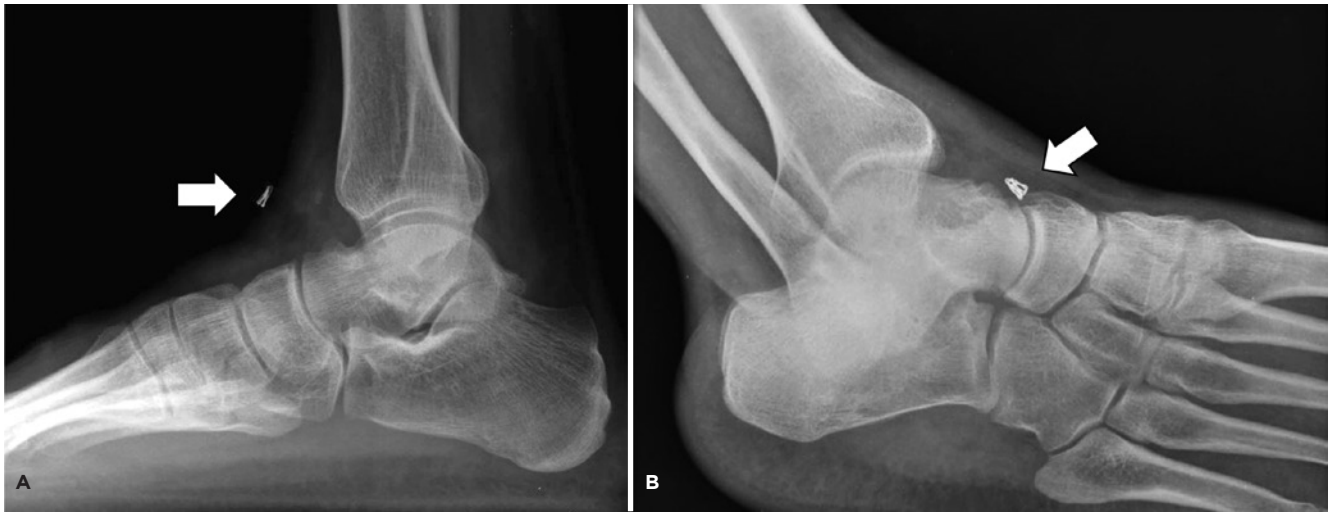

Figure 1. A) Profile radiograph image of the right ankle with metallic marking at the pain site. B) Oblique radiograph image of the right foot with metallic marking at the pain site.

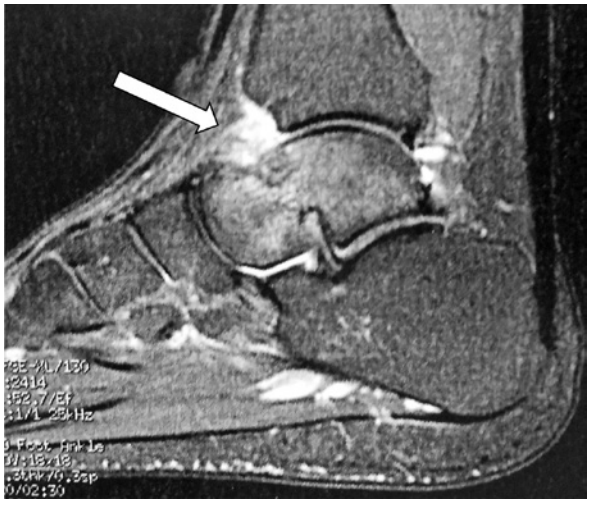

Figure 2. Magnetic resonance imaging of the right ankle, in the sagittal section, showing bone edema of the talus and periosteal reaction in the anterior region of the tibiotalar joint.

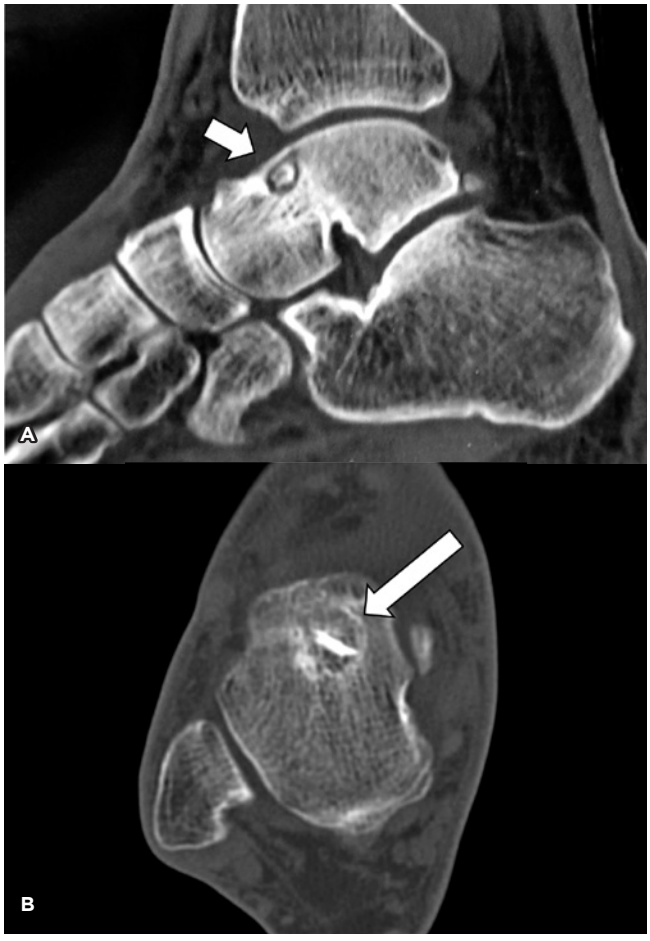

Figure 3. Computed tomography image of the right foot in the sagittal A) and axial B) sections demonstrating the nidus surrounded by sclerotic bone located in the neck of the talus.

On the first return, the patient reported a complete resolution of night pain. At two months postoperatively, the patient started walking with a full load. The ankle and hindfoot range of motion was similar to the contralateral side, with no pain complaints. The AOFAS scale for ankle and hindfoot at the time was 100 points, and the VAS scale evolved to no pain (0 points). The clinical condition, physical examination, and functional clinical evaluation scales remained unchanged until the last evaluation, three years after surgery (Figure 7).

Discussion

Osteoid osteoma is a benign osteoblastic bone tumor usually smaller than 1 cm. When present in the foot, it is preferably located in the talus and less frequently in the calcaneus, phalanges, and metatarsals[7]. The delay in diagnosis occurs mainly due to the vague symptoms that may be present in varied different diagnoses and the absence of radiographic findings[7,8]. Attention to cases of pain at rest that worsens at night, which improves with NSAIDs, and the chronicity of symptoms is a key point for OO suspicion[2]. In the case

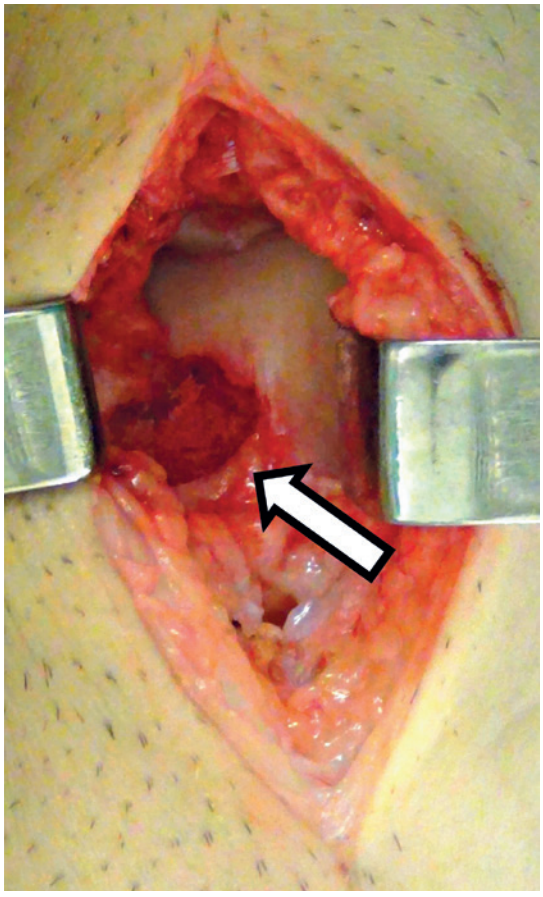

Figure 4. Bone failure on the neck of the talus after resection of the osteoid osteoma.

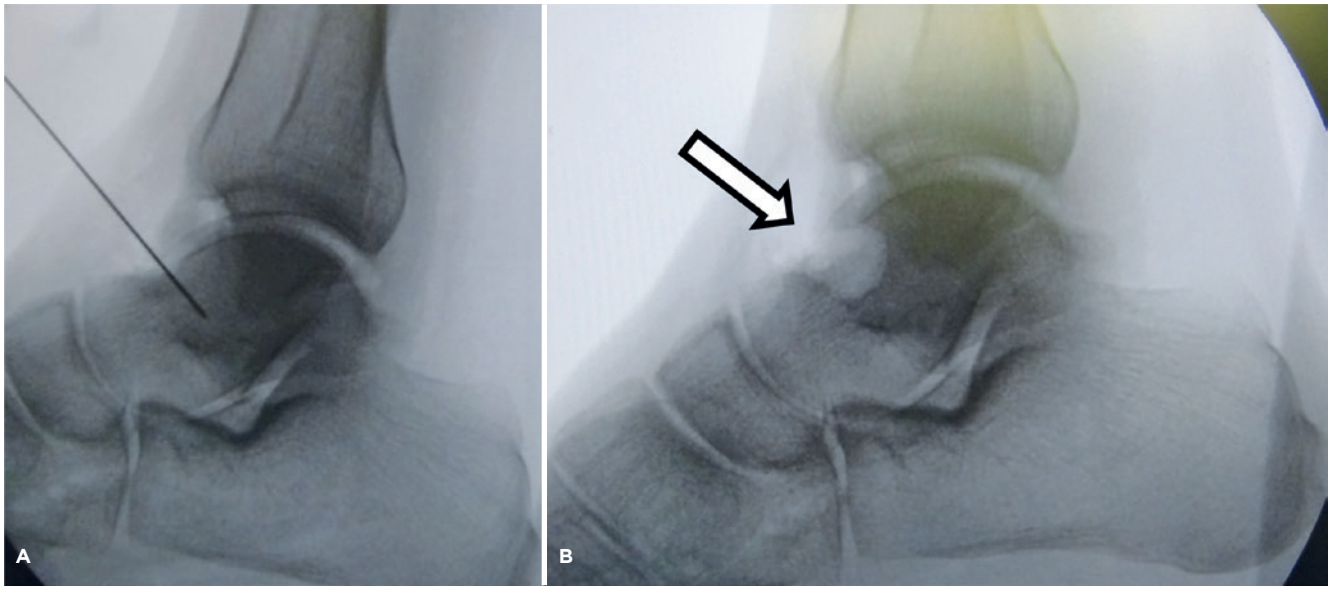

Figure 5. Intraoperative radioscopic images demonstrating. A) the location of the osteoid osteoma marked with Kirschner wire and B) the final result of osteoid osteoma en-block excision.

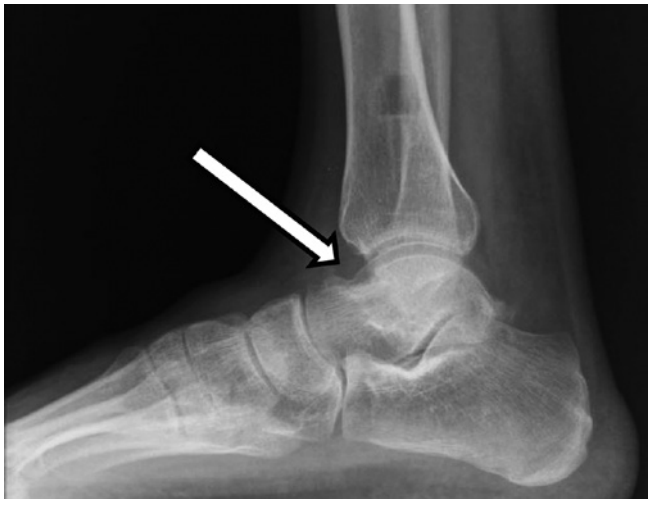

Figure 6. Immediate postoperative radiograph image of open osteoid osteoma resection and grafting with a cancellous bone graft of the right distal tibia.

reported, the patient had ankle pain for three years that worsened at night and improved with NSAIDs, but during this period, the OO diagnosis was not considered, always correlating it to pain due to old torsional trauma or overload at work. Orthopedists must know the disease and correlate the clinical presentation with appropriate complementary exams to suspect OO diagnosis.

From the exams, the radiograph may show a radiolucent lesion surrounded by bone sclerosis[7]. In cases of OO on the neck of the talus, the radiograph has a sensitivity of up to 61.5%[8]. In the case reported, the radiograph and the MRI showed soft tissue edema in the anterior ankle region of nonspecific character. The test considered the gold standard for diagnosis is CT[2,8]. Sharma et al.[16] compared single-photon emission computed tomography (SPECT-CT) with bone scintigraphy and simple CT. They concluded that SPECT-CT had greater sensitivity and specificity (100% and 100%) when compared with CT (77.8% and 92.3%) and bone scintigraphy (100% and 38.4%)[16]. The SPECT-CT is a tool to be used in cases where there is great doubt about the diagnosis, even after exams of less complexity.

Considering the natural course of OO, spontaneous resolution can occur between two and 15 years, but prolonged use of NSAIDs is discouraged due to their side effects and inefficiency in relieving symptoms in one-third of patients[8]. On the other hand, nidus surgical excisions are curative and provide pain relief[12]. The main treatment methods are open or arthroscopic surgical excision and thermal destruction by laser photocoagulation or radiofrequency[6]. The orthopedist should consider the advantages and disadvantages of these approaches and discuss them with the patient, seeking the best method to solve the problem. We present a case of OO on the neck of the talus in which diagnosis was delayed. This differential diagnosis should be remembered in chronic ankle pain, and additional tests should be requested. The lesion en-block resection resulted in complete relief of symptoms.

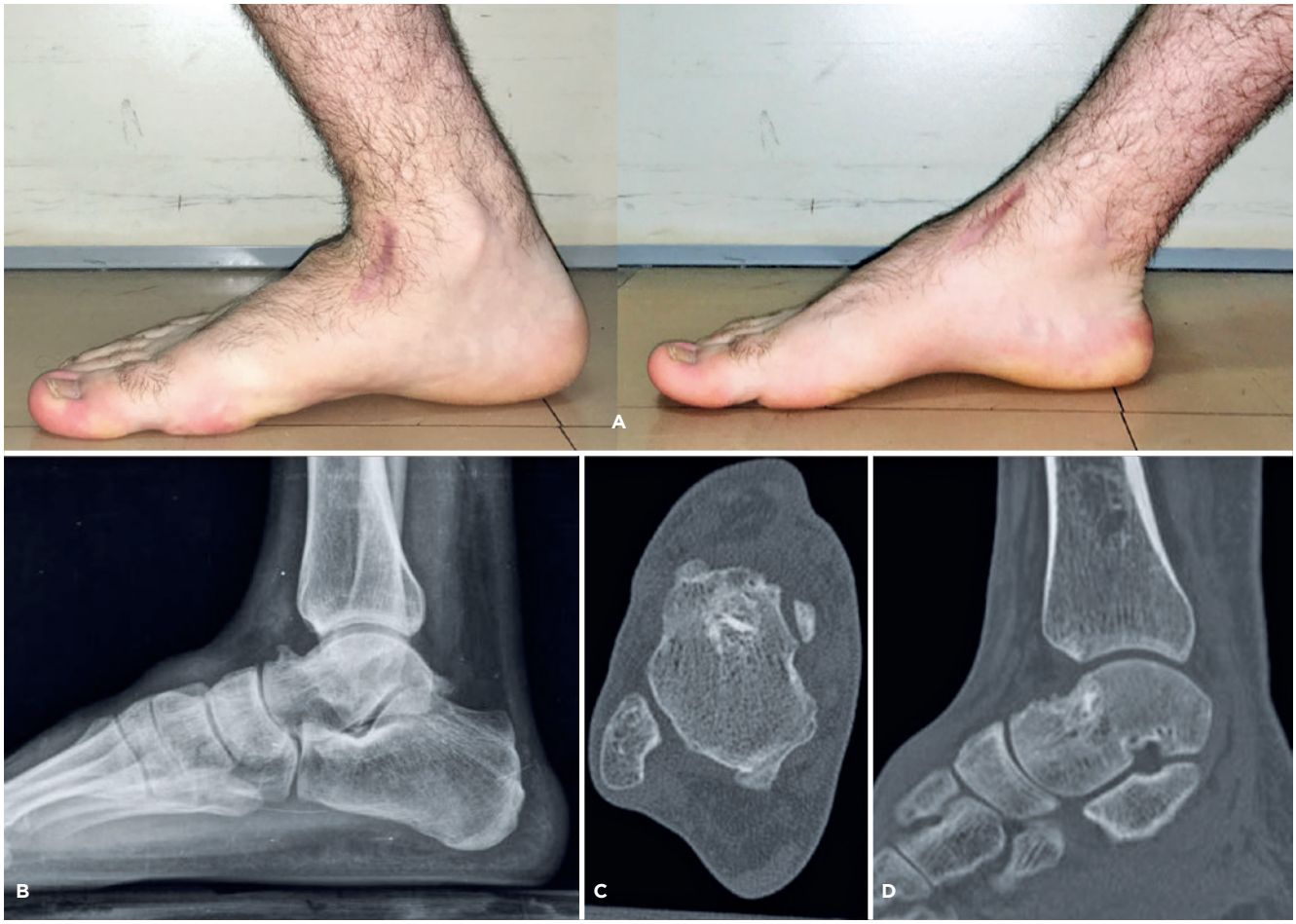

Figure 7. Images three years after osteoid osteoma treatment. A) Clinical image showing the leg-foot range of motion; B) Radiograph in profile; C and D) Computed tomography scans showing the eradication of the tumor lesion.

Authors' contributions: Each author contributed individually and significantly to the development of this article: MMC *(https://orcid.org/0000-0001-8133-7892) Conceived and planned the activity that led to the study, wrote the article, participated in the review process; IFL *(https://orcid.org/0000-0002-4973-8271), and LM *(https://orcid.org/0000-0001-5967-0541) Datta colletion, conceived and planned the activity that led to the study; JMPB *(https://orcid.org/0000-0002-5280-1673), and NMN *(https://orcid.org/0000-0001-7696-2220) Conceived and planned the activity that led to the study, participated in the review process; MTC *(https://orcid.org/0000-0001-9411-9376) Wrote the article, participated in the review process. All authors read and approved the final manuscript.*ORCID (Open Researcher and Contributor ID).

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