



# A Case Report of Symptomatic Gluteal Subcutaneous Heterotopic Ossifications with Persistent Sciatic Pain

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## Abstract

Heterotopic Ossification (HO) is ectopic bone formation within extra-osseous soft tissues. Its two major etiologies include genetic and acquired, the latter of which is more common and often associated with tissue trauma. Although the majority of cases are clinically insignificant and asymptomatic, severe HO may be associated with debilitating clinical consequences, including restricted range of motion, impingement, and pain. It can be a challenging condition to diagnose and manage, especially in patients with multiple co-morbidities and risk factors, and particularly as HO may recur after excision. We report a case of subcutaneous gluteal HO with acute irradiating pain, as well as its successful management. Uncharacteristically, HO had formed within the subcutaneous tissue, away from any surgical field. Our aim is to bring an awareness of atypical HO along with associated risk factors and various management options to help optimize patient care.

**Keywords:** Heterotopic ossification; Subcutaneous; Atypical; Pain

## Introduction

Heterotopic Ossification (HO) is defined as ectopic bone formation, outside of skeletal structures, within soft tissues [1]. HO can be divided into two forms: Acquired and genetic. The acquired form is the most common and is closely related to tissue trauma, be it from surgery or from injury, such as fractures or burns [2]. The genetic form occurs less frequently and is found in hereditary disorders such as fibrodysplasia ossificans, progressive osseous heteroplasia, and Albright's osteodystrophy [3-5]. The most common site for the formation of HO is within large muscle masses, particularly the hip and the thigh, following internal fixation of acetabular fractures or Total Hip Arthroplasty (THA) [6,7]. Several studies have reported the incidence of HO after THA varying anywhere between 5% to 87%, depending on patient risk factors, with only 5% to 12% being of clinical relevance [8-12]. Some cases have reported HO found near the ischial tuberosity to be associated with persistent sciatic pain [13,14]. In severe forms, it can significantly limit mobility and impact quality of life [15].

HO does not only develop in muscular tissue, but can also be found in the skin or the subcutaneous tissue. This condition, known as osteoma cutis, is the presence of bone formed within the dermis and can manifest as an isolated skin condition or secondary to rare genetic syndromes, including Albright's osteodystrophy [4,5]. HO has also been described in the subcutaneous tissue of patients with chronic venous insufficiency, but is often under diagnosed due to chronic ulcerations and scar tissue masking clinical findings [16]. In this article, we report on the findings and management of a case of subcutaneous HO in an elderly patient with no history of local trauma, presenting with atypical pain.

## Case Description

An 86-year-old female patient with a history of advanced lumbar spine degenerative disease affecting segments L4/L5 and L5/S1 and previous repeated revision THA on the right side, was admitted with a 1-week history of new-onset stabbing gluteal and lateral right hip pain, radiating along the lateral thigh down to the knee. Symptoms were mechanical in character and elicited when standing up, walking, or climbing stairs. No recent trauma was reported and there were no additional neurological deficits. However, the pain had become so severe and debilitating that the patient was unable to walk. Clinical findings revealed a tender, palpable mass over the right iliac crest as well as tenderness over the sacroiliac joint. The remainder hip examination was otherwise

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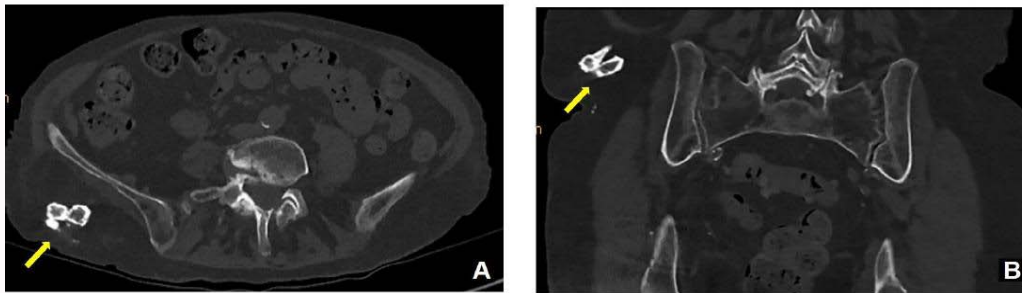
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**Figure 1:** CT scan of the pelvis with (A) axial and (B) coronal views, bone window. Yellow arrows identify well-defined bony formation in the subcutaneous tissue of the right buttock, well away from the muscles.

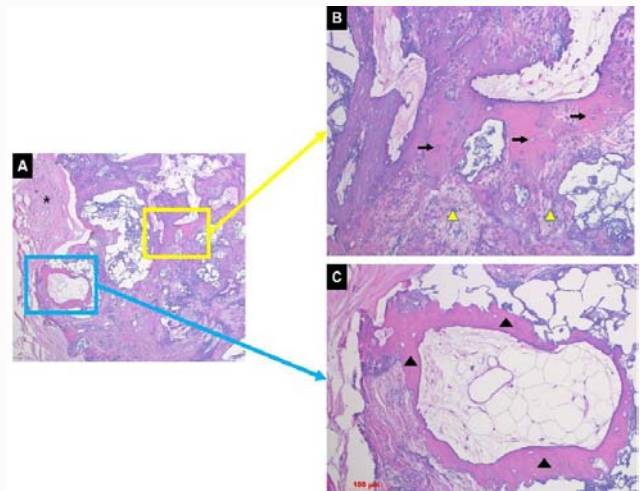


**Figure 2:** Macrophotography of the specimen removed from the subcutaneous tissue of the patient, after formalin fixation: one segment is ossified (left) and one smaller segment is lipomatous (right).

bland. On conventional radiographs as well as on CT scan of the pelvis no signs of loosening of the hip prosthesis were visualized. However, pronounced heterotopic ossification was detected in the subcutaneous tissue over the right iliac crest (Figure 1). MRI of the lumbar spine, performed considering the pattern of pain distribution within the dermatome L2/L3, showed multi-segmental degeneration as well as L2/L3 disc herniation in the right recess with L3 nerve root contact, but without root compression. As no other cause of pain was identifiable, a diagnosis of symptomatic HO in the subcutaneous tissue was considered. Surgical excision was successfully performed (Figure 2) and histology confirmed the presence of heterotopic ossification (Figure 3). The patient's symptoms regressed rapidly and completely, enabling immediate full mobilization and discharge from hospital one day following surgery. At the 7-week follow-up appointment, the patient was asymptomatic and very satisfied with the outcome.

**Discussion**

Heterotopic ossification is a common complication following soft tissue trauma, but rarely as a spontaneous process within subcutaneous fat. Although the underlying mechanisms for HO formation are not yet fully elucidated, several contributing factors have been described. It has been suggested that the requirements necessary for HO formation include having an inducing agent, an osteogenic precursor and a permissive environment for osteogenesis, all having been shown to contribute to proliferation and formation of bone [2]. It has also been proposed that failure to regulate the immune system or inflammatory response can lead to the release of inciting agents that lead to HO formation [17,18]. Further investigations have implicated Bone Morphogenetic Protein Type 2 (BMP-2) as a pro-inflammatory agent by stimulating release of substance p and calcitonin gene-related peptide from sensory nerves [19,20]. The



**Figure 3:** Photomicrograph of part of the resected specimen (hematoxylin-eosin staining).  
 A: Overview of heterotopic ossification displaying various stages of bone maturation and surrounded with a fibrous pseudocapsule (asterisk); x12.5. The tissue morphology has unfortunately suffered some damage due to the decalcification process.  
 B: Detail showing mature lamellar bone displaying few osteocytes (black arrows) and calcification zones of immature bone (yellow triangles); x200  
 C: Fatty marrow island with a central sinusoidal vascular space, absent hematopoiesis and at the periphery lamellar bone trabeculae (black triangles); x50.

clinical presentation depends on the stage of HO development. In the early/inflammatory phase, HO presents with localized pain, tenderness and swelling. In later stages and with gradual maturation of the bone tissue, the swelling regresses, the tissue becomes firm and it may restrict motion [21]. The most common sites of occurrence, in decreasing order, are: the hip, knee, shoulder and elbow [2,22], corresponding to body regions with voluminous muscle masses.

The extraspinal causes of sciatic pain are rare and can easily be missed. Two cases of persistent sciatica due to HO located near the sciatic nerve have been reported recently [13,14]. Pelvic tumors may also trigger sciatica and can cause a delay in diagnosis with consequently a worse outcome [23]. Following hip arthroplasty, such as with our patient, the ossifications usually form around the femoral neck component and lateral to the greater trochanter [24]. However, in our patient, the ossification had developed in the subcutaneous tissue at the level of the iliac crest, outside of the surgical field of the THA. Despite this location, pain irradiated into the thigh mimicking a radicular pathology L2 or L3.

Assessment of the extent and classification of HO severity is

predominantly made based on radiographic evaluation. In the early stages, it is typically a soft tissue mass without overt calcification and can often be missed, whereas in mature HO, lamellar bone is formed and becomes visible on X-ray images [25]. Radiography and CT scan remain the gold standard for diagnosis due to their ability to detect early bone formation and the relatively cheap cost [26,27]. The degree of HO is most commonly graded according to Brooker's classification, which was initially developed using anteroposterior radiographs of the hip [28]. However, there is no classification system for HO present in the skin such as with autoimmune disorders or rare genetic disease [21]. In our patient, histopathological examination of the excised tissue documented the presence of mature bone tissue within subcutaneous fat (Figure 3). The zonal architecture with a predominant peripheral ossification is a hallmark of HO, including thickened lamellar bone trabeculae with central fatty marrow and vascular spaces resembling bone marrow sinusoids [21].

Current management options for HO include prophylactic strategies to prevent or mitigate the severity of HO and excision to improve symptoms and function once the condition has occurred. Due to the large variability in the etiology for HO and specific patient risk factors, there is no consensus as to which prophylactic protocol is the most effective. Non-Steroidal Anti-Inflammatory Drugs (NSAIDs) and radiotherapy have both been shown to play a role in minimizing HO formation. Prophylactic NSAIDs have shown to reduce the occurrence of ectopic bone formation when given perioperatively compared to placebo, but at the expense of medication side effects such as gastrointestinal ulcers, bleeding, and delayed bone healing [29,30]. Radiation therapy has also proven to effectively reduce HO formation; however, its utility is limited by high costs and potentially reduced patient acceptance, especially considering it has not been shown to be more effective than NSAID therapy [2]. Surgical management currently remains the only effective treatment for established HO [26]. Indications for surgery include symptomatic disabilities and radiographic evidence showing the cessation of bone growth. It has previously been recommended not to perform surgery before 12 to 18 months of evolution, to allow maturation of the lesion in order to decrease intraoperative complications and HO recurrence [31,32]. However, it is the authors' opinion that waiting for this length of time is unnecessary and should be considered as soon as the patient becomes symptomatic. In this case, the HO was promptly excised and has proven to be an effective treatment for the patient.

## Conclusion

Although most cases are clinically insignificant and asymptomatic, severe HO may be associated with considerable clinical consequences including pain and reduced function. In our patient the HO caused irradiating pain, which resolved following its excision. Sciatic pain without clear clinical and radiological correlation should be investigated in the context of other extraspinal causes such as in sciatic nerve pathology or bone tumors, as previously reported. It is important to consider space-occupying lesions such as HO because they can develop outside of periarticular regions such as within the subcutaneous tissue. Part of the assessment includes histological analysis to rule out conditions such as osteoma cutis. Awareness of HO and an understanding of the associated risk factors along with the various management options will enable health care practitioners to optimize their patient care.

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