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ANATOMIC VARIATIONS

Myositis ossificans traumatica of the piriformis muscle: a rare mature case in an adult African male

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Abstract
Myositis ossificans traumatica (MOT) is a common form of heterotopic ossification associated to trauma. Rare mature manifestations and topographically atypical presentations of MOT are often misdiagnosed as osteosarcoma. This case study discusses a rare, mature case of MOT of the piriformis muscle, potentially clinically associated with piriformis syndrome. The ossification was observed on a dry sacral bone of an adult skeleton belonging to a South African male during routine inventory of the Raymond A. Dart Collection of Human Skeletons, the University of the Witwatersrand, Johannesburg. The MOT was located on the anterior aspect of the sacrum at a site corresponding to the upper portion of the origin of the muscle and extended laterally towards the greater trochanter, beyond the greater sciatic notch. It was cylindrical in shape and measured approximately 52.70 mm in length and 12.10 mm in diameter. Micro-focus CT revealed an extensive and mature bony development of the piriformis muscle with distinct outer cortical and inner trabecular bone. In addition, the skeleton showed widespread healed skeletal trauma, suggesting a history of trauma. The MOT was completely fused to the sacral bone excluding the possibility of congenital anomalies. Information on the MOT of the piriformis muscle is vital to clinicians and radiographers to aid in successful diagnosis and management of the piriformis syndrome and sciatica in the gluteal region. This case also provides a rare example to biological anthropologists, paleoanthropologists and bioarchaeologists of the representation of pathologies like these on a dry bone sample.

Keywords Myositis ossificans traumatica · Piriformis muscle · Heterotopic ossification · Osteosarcoma · Piriformis syndrome

Introduction

Myositis ossificans is a form of heterotopic ossification affecting muscular or deep soft tissues [3]. The most common cause of myositis ossificans is related to trauma [10], but a rare hereditary form has been reported which is referred to as myositis ossificans progressiva [3]. More specifically, myositis ossificans, as a result of a variety of traumatic events (i.e., myositis ossificans traumatica) has been reported following a variety of traumatic insults [1]. This condition is usually localised to the proximal limb extremities and particularly the hip region [7]. Despite its first documented account in the nineteenth century [10], the understanding of the pathophysiology of heterotopic ossification is still lacking [5]. Knowledge of myositis ossificans traumatica (MOT) progression is crucial for diagnosis, as the ossification progression undergoes varying stages and can often be misdiagnosed due to their similarity to certain bone cancers [12]. This misdiagnosis is further exacerbated by the presence of ossification at atypical sites, such as the maxillofacial, hand and deep gluteal regions [1, 2]. Therefore, diagnostically distinguishing MOT from bone cancers can be difficult, requiring multiple tests including various scanning modalities and biopsies [10, 12]. Furthermore, very few cases of mature MOT have been reported [14], possibly due to early identification of this abnormality and consequent treatment.

The current case study reports a novel account of a rare mature case of myositis ossificans traumatica of the piriformis muscle, identified on a dry bone sample from a skeleton belonging to a South African adult male.

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Case report

An unusual bony growth was identified on a dry sacral bone, with features suggestive of heterotopic ossification, during routine inventory of the skeletal remains of cadaveric origin, housed in the Raymond A. Dart Collection of Human Skeletons at the University of the Witwatersrand based in Johannesburg, South Africa. The skeletal remains were documented as belonging to a 38-year-old male South African of Xhosa descent. The reported cause of death was recorded as malnutrition.

The heterotopic ossification was located on the anterior aspect of the sacrum at the corresponding site of origin of the piriformis muscle. It extended from the left anterolateral surface of the sacrum at the level of the second sacral segment (S2) and projected laterally, crossing the greater sciatic notch on the left innominate bone (Fig. 1). The bony projection terminated approximately 12.10 mm lateral to the greater sciatic notch towards the greater trochanter of the femur. In total, the cylindrically shaped bony protrusion measured approximately 52.70 mm in length and 12.10 mm in diameter at its widest point. The heterotopic ossification followed the long axis of the piriformis muscle and corresponded to its superior margin localised at the level of S2. Posteriorly, this ossification demonstrated a groove between the most lateral aspect of the sacrum, which most likely accommodated the anterior sacroiliac ligament (Fig. 1).

Micro-focus computed tomography (μCT) scans, conducted at the Nuclear Energy Corporation of South Africa, confirmed the bone growth connected to the anterior aspect of the sacrum (Fig. 2). The bone growth (MOT of the piriformis muscle) consisted of a well-defined outer cortical bone layer and an inner core of trabecular bone (Fig. 2c). The μCT scan demonstrated an extensive mature bony mass without positive diagnostic signs of bone cancer or congenital abnormalities.

In conjunction to the above-mentioned MOT of the piriformis muscle, widespread skeletal trauma was observed in other regions of the skeleton. The specific sites of trauma

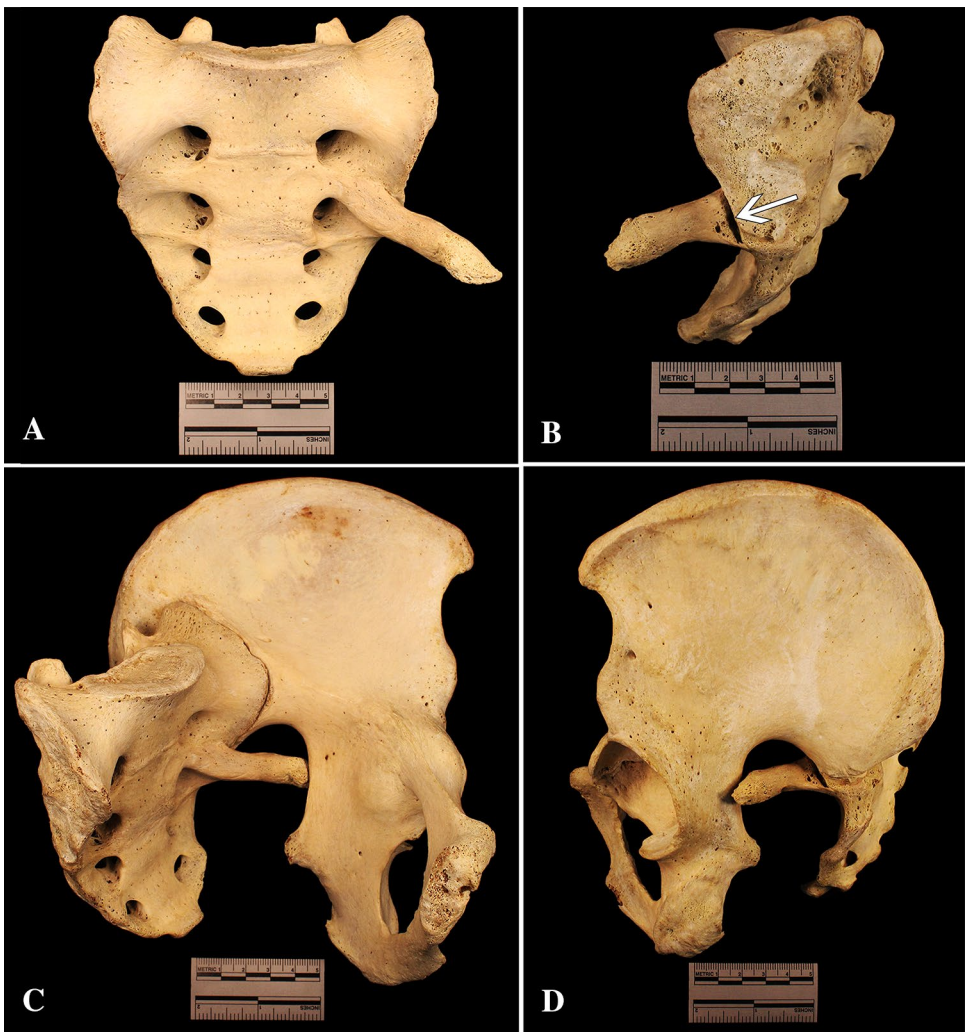

Fig. 1 Photographs of sacrum showing the heterotopic ossification (myositis ossificans of piriformis muscle). Anterior (**a**) and left lateral (**b**) views of the sacrum with attached heterotopic ossification corresponding to the site of origin of the piriformis muscle. A marked groove is seen between the origin of the MOT of the muscle and the sacrum in the lateral view (**b** white arrow), possibly accommodating the anterior sacroiliac ligament in vivo. Mature extent of ossification is visible with the growth extending beyond the sciatic notch in an articulated left hemi-pelvis from an internal view (**c**) and an external view (**d**)

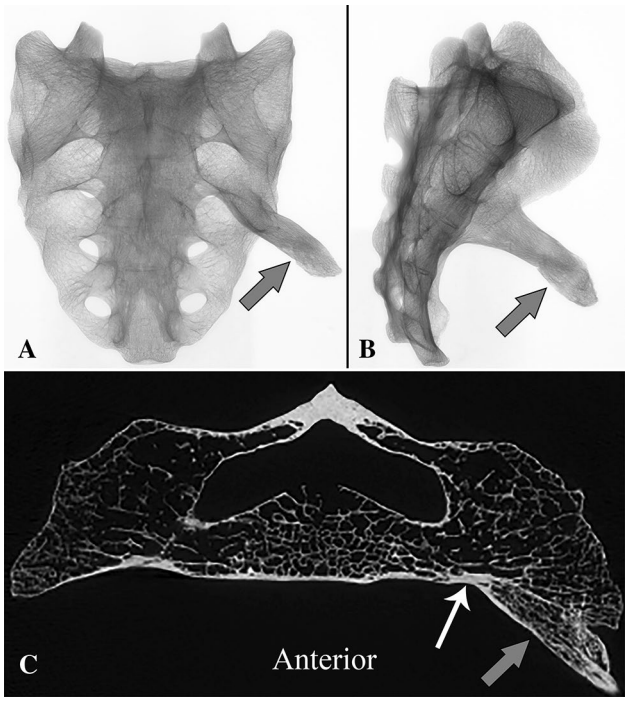

Fig. 2 Micro-focus computed tomography of the sacrum showing the relationship between the sacrum and MOT of the piriformis muscle. Image **a**, **b** demonstrate anteroposterior and lateral views of the sacrum with the attached MOT of the piriformis muscle (grey arrow). Image **c** depicts a transverse µCT orthoslice through the sacrum at the site of heterotopic ossification (grey arrow) attachment, at the level of S2 and lateral to the transverse foramina, respectively. A well-developed and circumscribed bony formation is visible with an outer cortical bone layer with an internal trabecular bone core. The white arrow in image **c** indicates the lack of periosteal reaction on the anterior aspect of the sacrum

included the viscerocranium (multiple bones), the neck (with cervical vertebrae C6 and C7 demonstrating bone exostosis on the anterior aspect as well as C4–C5 and T1 presenting osteophytes on the anterior aspect of their bodies), thorax (the ninth rib on both sides and the tenth and eleventh thoracic vertebrae with associated callus formation) and right femur which demonstrated a complete diaphyseal, healed and misaligned fracture localised in the distal shaft.

Collectively, the combination of the identified heterotopic ossification features observed both macroscopically and through µCT as well as the skeletal trauma profile is highly suggestive of myositis ossificans traumatica of the piriformis muscle.

Discussion

Myositis ossificans traumatica (MOT) is considered the most common type of heterotopic bone formation [3]. It is often identified in limbs of patients with a history of physical trauma and is commonly misdiagnosed as an osteosarcoma [6]. Positive diagnosis of MOT is achieved through computed tomography scans, provided the lesion has developed demonstrable zonal peripheral cortical bone traditionally associated with MOT [12]. In the current study, the MOT of the piriformis muscle showed a well-circumscribed bony mass with a peripheral cortical bone layer and a core of trabecular bone. The characteristic features of the MOT on µCT were typical of a mature progression of the pathology with late stage development of zonal peripheral cortical bone (Fig. 2c) [12]. In addition, the MOT showed features of a non-aggressive bone growth, such as the lack of a periosteal reaction along the bone attachment site (Fig. 2c, white arrow), distinguishing it from bone cancers.

In most cases, the classical appearance of MOT of muscle on plain radiographs is separate from adjacent bones, but in more mature cases, the bone growth may connect to the adjacent bone on the site of the muscle attachment. The presentation of mature MOT connected to the underlying bone can be misdiagnosed as a parosteal osteosarcoma [6, 7, 14]. This diagnosis was discarded based on the non-aggressive appearance of the bone formation and the adjacent sacrum. A form of heterotopic ossification that can be confused with MOT is myositis ossificans progressiva. However, myositis ossificans progressiva is extremely rare, strictly congenital, presents early in life with multifocal periarticular heterotopic bone formation and will progress rapidly to physically debilitate the patient resulting in early death [7]. As such, it is safe to state that the case presented here does not fit the criteria for myositis ossificans progressiva.

Alternatively, mature MOT of the piriformis muscle could be confused with congenital abnormalities such as a sacral rib. However, sacral ribs, such as lumbar ribs, generally present with a pseudo-articulation or a proper joint formation between the bony outgrowth and the sacrum and potentially a lateral deviation of the sacrum [8]. Since both features and the exact position of the MOT in this study do not fit the diagnostic criteria, as such it was excluded as a differential diagnosis.

The general limitations of this case study result from this report being based on a fully macerated dry-bone sample and not a living individual. As such, the authors are not privy to the patient history, associated symptoms (e.g., pain and loss of sensation), soft tissue indicators or any additional clinical diagnostic signs other than the reported cause of death. Despite these limitations, the diagnosis of MOT of the piriformis muscle corresponds to the majority of the diagnostic criteria associated to this condition which is further supported by the trauma profile of the individual.

To the authors' knowledge, only a single other case of MOT of the piriformis muscle has been reported [2].

Similarly mature cases of MOT have been found to cause severe topical complications [1, 3], with loss of daily functioning in regions affected (such as the limitation of mouth opening, elbow joint complex range of movement, etc.), disfigurement and impingement of neurovasculature [1–3, 14]. As a result, patient lifestyle and health can be severely compromised dependent on location, extent and associated neurovascular structures to MOT presentation. In the current case, the piriformis muscle was affected. This muscle is clinically considered a crucial landmark, commonly utilised as a guide for many ultrasound-based investigations and surgery, in the gluteal region [4, 11]. Hence, knowledge of the piriformis muscle and its variations is critical for successful management of the disorders of the gluteal region. The arrangement of the muscle demonstrates significant variation, particularly with respect to muscle fibre separation and fusion to other gluteal muscles or even complete muscle aplasia [4, 13]. The piriformis muscle's relations to the sciatic nerve can also vary greatly with the nerve traversing the greater sciatic foramen either superiorly, inferiorly or through the muscle itself [9, 11]. Variations of the piriformis muscle and sciatic nerve pathway, although less common with a prevalence of 2–6% (European populations) [9, 11], may also predispose the sciatic nerve to entrapment from the surrounding structures. Similarly, extensive ossification of the piriformis muscle can present with symptoms typical of piriformis syndrome [2], a compression of the sciatic nerve in close proximity to the piriformis muscle.

This could have likely occurred in the current case due to the extensive mature nature of this ossification. Symptoms of piriformis syndrome include lower limb pain, paraesthesia, pain and weakness as well as buttock and lower back pain [9–11, 13]. Piriformis syndrome, through sciatic nerve entrapment, can also be induced by osteophytes, hematomas, pseudo-aneurysms, external pressure and endometric cysts [2, 9, 11].

Piriformis syndrome treatment commonly relies on resolving the underlying cause of sciatic nerve compression; in this case, it would involve resolving the MOT of the piriformis muscle. Treatment options for early MOT may involve NSAID drug therapy, early mobilisation, physiotherapy and radiation therapy to prevent bone proliferation [3, 13, 14]. In more advanced cases, extracorporeal shockwave therapy and surgical resection of bony masses may be employed [2, 3, 14]. Based on the extent and mature presentation of the MOT in this case, it is highly unlikely for this individual to have received treatment for MOT of the piriformis muscle.

In summary, all the aforementioned features strongly build towards a case of myositis ossificans traumatica of the left piriformis muscle. The degree of muscle ossification shown in this case is quite extensive, which is rarely observed clinically or posthumously, especially on a dry bone sample. As a result, this case represents a rare example of mature MOT to biological anthropologists, paleoanthropologists and bioarchaeologists. The extent of ossification of the piriformis muscle is suspected to be a result of lack of medical attention, as evidenced by the untreated femur fracture, as well as the extensive ossification of piriformis, which was likely to have caused secondary piriformis syndrome. The effects of piriformis syndrome can be debilitating in patients and as such, distinguishing MOT from osteosarcomas and sacral ribs can expedite treatment and prevent additional invasive or harmful procedures. Furthermore, identifying the aetiology of sciatica can lead to successful treatment and management of symptoms.

Author contributions NB, PM, and BKB: project development, manuscript editing, final approval of manuscript. NB and BKB: data collection, analysis and interpretation of data, manuscript writing.

Compliance with ethical standards

Ethical approval The current study was approved by the School of Anatomical Sciences and Human Ethics Research Committee (Medical) (Waiver Number: W-CJ-140604-1).

Conflict of interest The authors declare that they have no conflict of interest.

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