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Myositis Ossificans Traumatica with Associated Pseudarthroses in an Adult from Late Bronze Age Athens, Greece

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SHORT REPORT

Myositis ossificans traumatica with associated pseudarthroses in an adult from Late Bronze Age Athens, Greece

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Abstract

This case study documents an unusual heterotopic ossification with associated pseudarthroses of the lumbar spine. We examined the partial skeletal remains of an adult from a Late Bronze Age (Mycenaean Late Helladic IIB-IIIA1 period, approximately1400–1375 BCE) chamber tomb from the Athenian Agora excavations in Greece. This individual had a large bony mass in the region of the *inter-transversarius* muscle that spanned L3–L5 vertebrae and formed pseudarthroses at the superior and inferior ends. The differential diagnosis of the bony mass included dystrophic and neoplastic calcifications and myositis ossificans traumatica (MOT). MOT is a benign heterotopic bone growth typically found in skeletal muscles. MOT usually results from a trauma and is most commonly found in the thigh, buttocks, or upper arm, although it has been documented clinically in other areas as well, such as in paravertebral muscles. The mature, remodelled cortex of the bony mass and the two well-developed pseudarthroses indicate that this individual lived with this condition for a number of years.

KEYWORDS
Athenian Agora, heterotopic bone, pseudarthrosis, traumatic injury

1 | INTRODUCTION

Myositis ossificans traumatica (MOT) is the most frequent heterotopic bone condition in humans. This condition is the result of bone forming in soft tissue and usually follows from a traumatic event (Karam & McKinley, 2008). MOT is considered to be benign by clinicians (Kim & Choi, 2009) as it normally resolves on its own (Patel, Richards, Trehan, & Railton, 2008). However, patients do present with pain, swelling, and in some cases, limited motion. Symptoms of MOT begin about 4–5 days after the injury (Beiner & Jokl, 2002), and a mass may be visible on radiographs by 3–4 weeks after the injury (Mestan & Bassano, 2001). The maturation of the mass is complete at 6–24 months after the onset of the development of ossification (Saussez, Blaivie, Lemort, & Chantrain, 2006; McCarthy & Sundaram, 2005).

MOT has been well-documented in the paleopathological literature. Although the examples provided by Ortner's encyclopedic work (Buikstra, 2019) show well-developed lesions of myositis ossificans, the majority of skeletal cases presented in the literature are comparatively minor and would have likely caused few limitations on the individuals exhibiting them (Mann, 1993, Lagier & Baud, 1980, Lovell, 1997:146–147, Judd, 2004:43).

The case presented here documents a case of spinal myositis ossificans with a well-developed pseudarthrosis that had not resolved at the time of death. To our knowledge, a paravertebral mass with pseudarthroses has not been documented in the paleopathological

FIGURE 1 Athens, Greece [Colour figure can be viewed at wileyonlinelibrary.com]

literature, although it has been clinically reported (Mourad & Grant, 1983).

2 | MATERIAL AND METHODS

A chamber tomb was excavated during the 1998 and 1999 field seasons of the Agora Excavations in Athens, Greece (Figure 1) by the American School of Classical Studies at Athens under the direction of John McK. Camp. Chamber tomb K2:5 (Figure 2) dates to the Mycenaean Bronze Age, Late Helladic IIB-IIIA1 period, approximately 1400–1375 BCE. The tomb consists of a square cut chamber, probably originally roofed with stone. Access was through a passage or *dromos*, approximately 1 m wide and 2.5 m long. Grave goods included

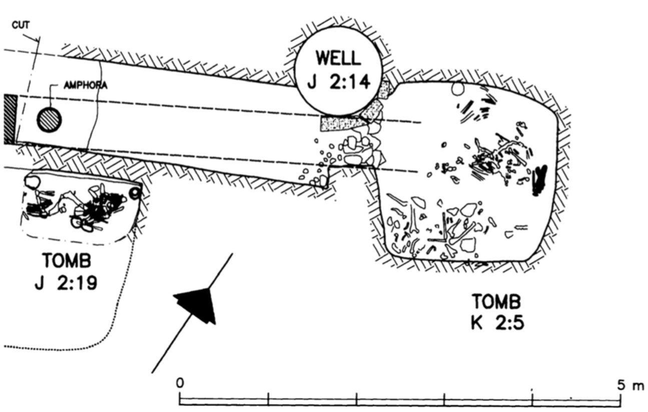

FIGURE 2 Tomb K2:5. The burial chamber is to the right with the entrance passage leading to the left. (American School of Classical Studies at Athens: Agora Excavations)

FIGURE 3 Ossified mass ankylosed to portions of L3 and L4. The mass articulates with an expanded left transverse process of L5. Posterior view. (a) Ossified mass; (b) superior articular process of L3; (c) inferior articular process of L3; (d) superior articular process of L4 that has fused completely to the ossified mass; (e) spinous process of L5; (f) expanded left transverse process of L5, forming a joint with the inferior end of the ossified mass [Colour figure can be viewed at wileyonlinelibrary.com]

11 ceramic vessels, two terra cotta figurines, approximately 790 beads, and an unusually rich collection of bronze weapons: two daggers, two blades, and one shaft (Camp, 2003). A minimum of 10 individuals, seven adults (four male adults, three female adults) and three juveniles were buried in the tomb.

The condition of the skeletal remains (catalogue# AA354) reflected burial patterns seen in other Mycenaean chamber tombs. Bodies were normally laid out on the floor or on biers in primary internments, and the bones were later moved aside for subsequent burials (Boyd, 2016). Due to the resulting comingling, and the fragmentary nature of the remains, many of the adult bones in K:25 could not be attributed to an individual nor could sex estimations be performed for most skeletal remains. It was possible, however, to associate distinct units of vertebrae. Skeletal remains were examined macroscopically.[1]

3 | RESULTS

Here, we present the pathologies found on a unit of vertebrae, consisting of the third through fifth lumbar vertebrae (L3–L5) and an ossified mass fused to L3 and L4 (Figures 3 and 4). The vertebrae are

[1]It was not possible to acquire the necessary permits for transport and radiographs of the bone, so the interior structure of the bone cannot be determined.

FIGURE 4 Ossified mass. Lateral view. Superior is to the left, and posterior is up. The superior end (a) has a pseudarthrosis, but its adjoining joint surface was not preserved. The inferior end formed a pseudarthrosis (b) with an expanded transverse process of L5 [Colour figure can be viewed at wileyonlinelibrary.com]

from a mature individual. No specific indicators of sex or age beyond adult could be associated with this individual.

L3 consists of the left half of the neural arch (Figure 3). The left superior articular facet is present and unremarkable. The inferior articular facet is in articulation with the articular facet of L4, and the joint space is preserved. This vertebra is ankylosed to L4 by a large bony mass in the region of the *intertransversarius* muscles. The mass originates on the infero-lateral surface of the left superior articular facet and encompasses the left transverse process of L3.

L4 consists of the left superior articular facet in articulation with the inferior articular facet of L3, the pedicle, and the left transverse process that is enveloped by the bone mass (Figure 3). The bony mass is attached to the lateral surface of the left superior articular process, encompasses the left transverse process, and continues inferiorly. (Figures 3 and 4).

L5 is nearly complete with the exception of the inferior surface of the vertebral body (Figure 3). The left transverse process extends laterally with a large exostosis. The exostosis projects superiorly 17 mm at the lateral end forming a *U* shaped facet that articulates with the inferior end of the bony mass in a pseudarthrosis. The right lamina is thickened on the anterior surface and extends farther into the vertebral canal than the left does. This asymmetry is likely associated with the other remodelling exhibited on the bone.

The paravertebral bony mass is ankylosed with L3 and L4 and articulates with L5 (Figures 3, 4, and 5). The mass is 69 mm in superior–inferior length and 18 mm wide at the midpoint. The location of the mass suggests that it formed within the *intertransversarius* muscle. At the proximal end of the bony mass is the inferior joint surface of a pseudarthrosis (14 × 8 mm). The superior joint surface of the pseudarthrosis is not preserved but was likely on or near the area of the L2 left transverse process. The mass is fused to both L3 and L4 and extends inferiorly to the second pseudarthrosis (18 × 17 mm) at the expanded left transverse process of L5. The mass is covered with smooth cortical bone, which suggests remodelling over time, although no active remodelling was present at the time of death. In areas where the cortical bone was damaged post-mortem, obvious trabecular structures were visible in the underlying bone. The mass clearly was not hollow, as it was quite dense and heavy for its size.

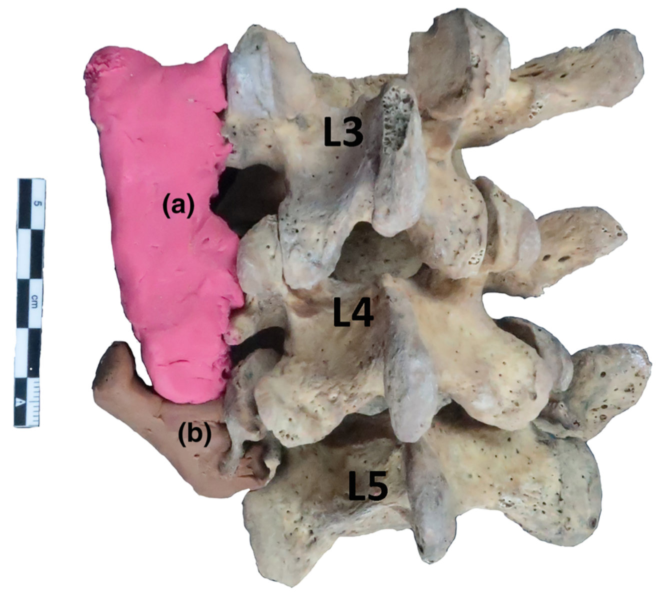

FIGURE 5 Model demonstrating how the ossified mass (a) would have appeared in life adjacent to vertebrae L3–L5. (b) The expanded transverse process of L5 [Colour figure can be viewed at wileyonlinelibrary.com]

4 | DIFFERENTIAL DIAGNOSIS

Ossified tissues fall broadly into three categories: dystrophic calcification, neoplastic calcification, and heterotopic bone formation. The first two are normally characterized by disorganized deposits of hydroxyapatite, while with time, heterotopic bone develops mature cortical and trabecular structures as it matures and remodels (Freire, Moser, & Lepage-Saucier, 2018).

Dystrophic calcifications may occur in calcified cysts and various organ and connective tissues. While these may exhibit vascular structures, they lack distinct cortical and trabecular bone (Komar & Buikstra, 2003). Trauma, with or without infection, can produce dystrophic calcifications, and these have been documented in the paravertebral area. Dystrophic calcifications may efface the intervertebral spaces and produce skeletal changes (Moore et al., 2016; Rothschild, 2014). While the vertebra in the individual from K2:5 are incomplete, the morphology of the lumbar vertebrae has been maintained in the structures adjacent to the mass. The presence of cortical and trabecular bone in the K2:5 mass also conflicts with the usual appearance of dystrophic calcification.

Neoplastic calcifications may form in association with benign and metastatic tumours, including osteosarcoma and ostoblastoma. Osteosarcomas arise primarily at growth plates. In the spine, they frequently present with aggressive periosteal reaction and cortical destruction (Katonis et al., 2013), neither of which is present in the vertebrae from K2:5. Osteoblastoma is a benign bony neoplasm characterized by osteoblastic tumors, sometimes accompanied by lytic lesions (Jans & Verstraetel, 2004). While they are commonly found in the vertebrae, they consist of woven structures that are poorly mineralized (Ortmann & Eady, 2016; Riccomi, Fornaciari, Minozzi, & Giuffra, 2018). Given the smooth dense cortex of the K2:5 mass, a diagnosis of osteoblastoma is not warranted.

Heterotopic ossifications (HOs), unlike dystrophic and neoplastic calcifications, form both cortical and trabecular bone (Freire et al., 2018). The bone mass from K2:5 was covered with smooth cortical bone, whereas post-mortem damage revealed internal trabecular bone structures. We therefore identify this as a heterotopic bone mass, not a dystrophic calcification or other soft tissue calcification. There is a lack of consensus on the terminology for the various expressions of HO, and in clinical literature, the term is used interchangeably or in place of myositis ossificans, although HO may also encompass widespread tendon ossifications of DISH and calcium pyrophosphate deposition disease (Freire et al., 2018; Karam & McKinley, 2008; Moore et al., 2016). Myositis ossificans progressiva or fibrodisplasia ossificans progressiva is a distinct genetic disease with systemic expression and is not relevent here (Vigorita, 1999). Myositis ossificans is a largely benign heterotopic bone growth found in skeletal muscles and occasionally other soft tissue and may present in varying ways. With a single locus in this case, we can reject diagnoses of myositis ossificans progressiva or the manifestations calcium pyrophosphate deposition disease including DISH.

Localized heterotopic bone formation associated with skeletal tissues is most frequently termed myositis ossificans. MOT forms after a traumatic injury to soft tissue. Ossification occurs in the area of inflammation, with lesions occurring at the origins or insertions of soft tissue (Redfern & Roberts, 2019) or as a mass completely within the soft tissue. MOT shows a greater frequency in adolescent or young adult males, but is documented at all ages, from infants to individuals in their 80s (McCarthy & Sundaram, 2005). The lesions in MOT typically progress through stages of maturation, resulting ultimately in mature, remodelled, heterotopic bone that may either persist or be resorbed. Cortical and trabecular bone have been documented in MOT cases, both macroscopically and microscopically (Lagier & Baud, 1980; Mann, 1993). MOT with pseudarthrosis has been documented in the clinical literature in the paravertebral area, within the *intertransversarius* muscle, following a blunt force injury to the lumbar region (Mourad & Grant, 1983). The most likely diagnosis of this mature bone structure with associated pseudarthroses in the individual from chamber tomb K2:5 is MOT.

5 | DISCUSSION

The heterotopic new bone affecting the individual from chamber tomb K2:5 is unusual because of its specific location, the *intertransversarius* muscle, and the subsequent development of the pseudarthroses. Most cases of MOT are found in areas of the body more commonly affected by trauma, including the thigh, buttocks, and upper arm (McCarthy and Sundaram, 2005), but there are clinical cases involving the lumbar spine (Abdallah, Gokcedag, Ofluoglu, & Emel, 2014; Karam & McKinley, 2008; Kim & Choi, 2009; Mourad & Grant, 1983). In modern cases, athletes who participate in activities that predispose them to avulsion or blunt force injuries are most at risk (Aufderheide & Rodriguez-Martin, 1998).

The pseudarthroses from the individual in tomb K2:5, between the paravertebral mass and L2 superiorally, and L5 inferiorally, appear to have been caused by continued movement postinjury (McCarthy and Sundaram, 2005). A similar case of MOT with pseudarthroses was documented clinically in a young male adult who was a competitive athlete (Mourad & Grant, 1983). He received blunt force trauma to his lower back in a rowing accident that caused pain and significantly reduced lateral bending. Radiographs revealed heterotopic bone formation of the right side of the lumbar spine between the spinous and transverse processes. Pseudarthoses were documented between the bone mass and the transverse processes. The pseudarthroses developed only after he resumed his extensive physical activity. Our review of the paleopathological literature found no documented cases of MOT in the spine or paravertebral areas in any ancient skeletons. In addition, pseudarthroses in the paleopathological literature are confined to discussions of nonunited fractures and have not been associated with MOT (Brickley & Buckberry, 2015).

Although we cannot know the exact circumstances that caused the MOT and pseudarthroses to develop in the individual from K2:5, we offer the following hypothetical scenario informed by the clinical case presented by Mourad and Grant (1983). The Late Bronze Age adult from K2:5 suffered a traumatic injury to the lower back causing a deep bruise from blunt force. The extensive remodelling of the paravertebral bony mass indicates survival for years after the original injury, and the pseudarthroses indicate physical activity continued following the injury. The ossified muscle mass would have reduced mobility to some degree and pain would have likely been persistent (Mourad & Grant, 1983). Long-term stiffness may have been associated with the bony mass (Vigorita, 1999).

6 | CONCLUSIONS

This case study is important for documenting this rare formation of pseudarthroses on a heterotopic bone mass. MOT is often identified in paleopathology and medical literature provides many case studies, but paravertebral occurrences are rare in both contexts. Likewise, pseudarthroses are unusual co-occurrences but are occasionally reported in medical literature. In a broader sense, it is also important for a glimpse into the life of a Bronze Age individual, who suffered a significant injury, but remained active while healing, resulting in the unusual pseudarthroses.

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