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Skeletal evidence of Diffuse Idiopathic Skeletal Hyperostosis (DISH) in a collective burial from Byzantine Greece

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Abstract: The paper reports on a collective burial from a 13th c. AD cist grave in Attica, Greece. The grave was located inside a basilica and held the remains of at least ten adults. Bone representation analysis showed secondary manipulation of previous deceased including long bone selection for reburial in the same grave and/or bones transported from a different burial place. Paleopathological analysis used macroscopy and radiology, and revealed several lesions on the axial and appendicular skeleton expressed mainly by spinal ligament ossification and multiple peripheral enthesopathies. Individuation of the remains pointed to a middle-aged male with DISH, a condition often correlated to high social rank. Byzantine period is marked by increasing development and prosperity in Greece including among others the creation of many local monastic centers. Although the precise social and professional profile of these individuals cannot be revealed, the combined investigation of skeletal and archaeological evidence suggests that the grave gathered the remains of individuals belonging to an upper class social group.

Key words: DISH, Byzantine period, Greece, cist grave, secondary manipulation, high status individuals

Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) (Resnick and Niwayama 1976), also known as Forestier's disease (Forestier and Rotes-Quérol 1950), is a systemic noninflammatory ossifying disorder of unknown cause which affects predominantly older individuals and male sex (Aufderheide and Rodriguez-Martin 1998; Ortner 2003). Clinically this condition is characterized by ossification and/or calcification of ligaments and entheses in the axial and the appendicular skeleton. The lower thoracic and lumbar spines are the most common sites for DISH involvement (El Miedany et al. 2000) but other entheseal regions in the peripheral joints may be affected (Dar et al. 2007; Mader 2003; Mader et al. 2009; Resnick et al. 1975; Sarzi-Puttini and Atzeni 2004). Patients with DISH usually present spine rigidity, decreased mobility, middle and low back pain and dysphagia from esophageal compression (Cammisa et al. 1998; El Miedany et al. 2000; Jansen and Ur Rehman 2013; Resnick et al. 1975).

Although the etiology of DISH remains uncertain, it is associated with

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advanced age, type II diabetes mellitus, obesity (conditioned by calorically adequate diets, over-eating and low level of activity), genetic predisposition, metabolic factors and micro trauma (Cammisa et al. 1998; Denko and Malemud 2006; El Miedany et al. 2000; Kiss et al. 2002; Mader 2003; Pappone et al. 1996; Pillai and Littlejohn 2014; Resnick et al. 1975). The age and dietary factors have been proved of particular interest for paleopathological investigations because they would be indicative of high social status. Bioarchaeological record includes various examples mainly from European monastic cemetery contexts (see Smith et al. 2013; Giuffra et al. 2009; Mays 2000; Reale et al. 1999; Ortner 2003); few cases of DISH are also reported from Greece (Fox 2005; Lagia 1999; Bourbou 2005; Bourbou and Tsilipakou 2009). This paper analyzes a collective burial from the Byzantine church of Taxiarches in Attica, central Greece. The aim is to investigate the mortuary features of the burial and contextually explore the health outcomes of the individuals, with a special focus on DISH, in relation to the historical period and burial location.

Materials and Methods

Material and archaeological context

The rescue excavation at the 13th century AD Byzantine church (basilica) of Taxiarches (Gini-Tsofopoulou 1985) has been conducted in 2003 by the 1st Ephorate of Byzantine Antiquities at Athens, and brought to light a cist grave inside the main part of the church; no archaeological evidence for the presence of a monastic complex is documented. According to the excavation notes (unpublished data), the human skeletal remains have been uncovered in two different layers: the upper layer held the primary burial of an adult skeleton laid on the back with the forearms folded on the chest and abdomen as well as some 'redeposited' adult bones in proximity to the skeleton (no further details are provided). The lower layer included commingled remains of adults. During the excavation no recording of possible bone arrangements and anatomical relationships was operated and the bones have been excavated without the use of microstratigraphic layers. Several bones were not removed with clear distinction between the primary burial and the redeposited or commingled remains.

Methods

Human remains were cleaned with water and analyzed macroscopically and radiographically. All remains were sorted and a visual pair matching complemented by differences in age/maturation, size, robustness, comparison of articulating bone portions, and matching of fragmented pieces was applied. In addition, skeletal elements belonging to the same pathological unit were matched and some of them have been individuated (Duday 1987). The Minimum Number of Individuals (MNI) was estimated by the highest number of frequency of non-matching elements. The calculation of the Bone Representation Index (BRI) as employed by Bello and Andrews (2006) was used in order to quantify the skeletal elements uncovered in the grave. The characterization of the burial was performed according to the terminology proposed by Duday (2009).

Age estimation for adults was based on morphological changes of the auricular surface of the *os coxae* (Meindl and Lovejoy 1989). Each adult individual was assigned to one of the age classes proposed in Buikstra and Ubelaker (1994).

Stature calculations were made using the formulae for the maximum femoral lengths (Trotter 1970). Paleopathological analysis used data collecting methods outlined in Buikstra and Ubelaker (1994). Skeletal lesions were inventoried by presence-absence.

Skeletal diagnosis of DISH

In the clinical (Cammisa et al. 1998; Olivieri et al. 2009) and paleopathological (van der Merwe et al., 2012) literature the diagnosis of DISH uses a set of features observed in the spinal and extra-spinal skeleton (Crubézy 1990; Arlet and Maziéres 1985; Resnick and Niwayama 1976; Rogers and Waldron 2001; Utsinger 1985; Waldron 2009). These criteria vary according to the method used; in the present study, the features observed include the ossification of the anterior longitudinal ligament along the anterolateral aspect of at least four continuous vertebral bodies (mainly in the lower thoracic spine), preservation of intervertebral disc spaces, symmetrical extra-spinal osteophytes, and absence of apophyseal joint ankylosis. Extra-spinal enthesal ossifications are included in the diagnostic criteria of DISH when spinal evidence confirms the diagnosis; they affect the skeleton bilaterally and usually include the pelvis, heel, knee, foot, and elbow (Resnick et al. 1975). Finally, DISH is associated with sacroiliac joint bridging (Crubézy 1989; Dar et al. 2007; Haller et al. 1989; Waldron and Rogers 1990) but without intra-articular bony ankylosis (Aufderheide and Rodriguez-Martin 1998; El Miedany et al. 2000). Extra-spinal lesions and sacroiliac joint bridging are also considered for diagnostic purposes in this study.

Results

Skeletal analysis

The MNI of the analyzed sample is ten. The most frequently represented bones with BRI>50% are femora and tibiae (Fig. 1). The BRI for upper limbs and fibulae ranges between 20% and 40 %. Crania and pelves are preserved at 20 %; the rest of the bones are poorly represented (<15%) and most bones of hands, feet and patellae are absent. Dental and

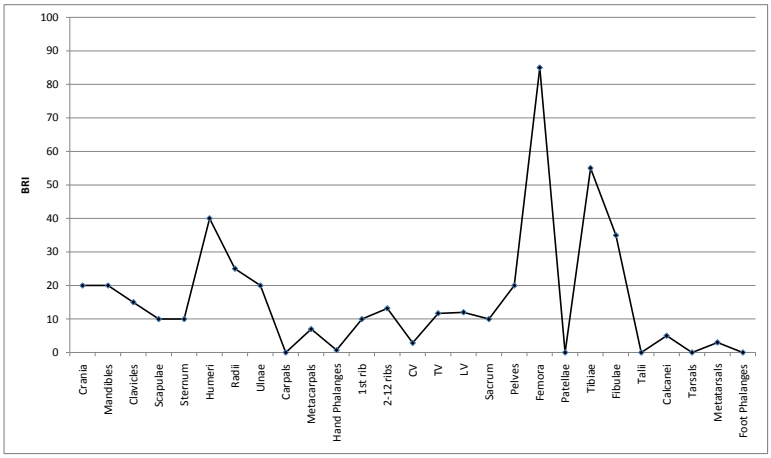

Fig. 1. Bone Representation Index (BRI) for the skeletal remains of the cist grave

alveolar remains from two partially preserved mandibles are observable; in total, 14 teeth and 24 sockets are counted. All individuals are adults. Age-at-death and sex are estimated for two old males (i.e. 50+ years) with the same stature (1.77 m and 1.76 m). The rest of the adults are classified as of indeterminate sex.

Nine teeth with slight and moderate calculus deposits (supra- and subgingival), one tooth lost prior to death and four sockets with alveolar resorption (horizontal bones loss) are recorded on one of two dentitions. No carious lesions or enamel defects are observed. The observable teeth do not display nonmetric traits. In addition, the crania do not show abnormalities of size, shape, abnormal new bone or bone loss. The most common pathologies on post-cranial bones are ossifying extra-spinal enthesopathies, osteoarthritis (OA), degenerative joint diseases (DJD) and hypertrophic bone formation; the lesions affect the bones of axial and appendicular skeleton. Bearing in mind the fragmentation of several long bones and the poor representation of the spine, hands and feet, then OA, DJD and extra-spinal enthesopathies seem to affect at least two individuals. On the contrary, hypertrophic bone formation affects one adult. By applying all the criteria for the individuation of the remains, the elements with excessive amounts of bones at joint margins and enthesopathies are attributed to one old male (Fig. 2), most likely the primary inhumation of the burial. The following paragraphs include the description of the spinal and extra-spinal hypertrophic lesions of this individual.

Spinal lesions

The anterior and right side of the spine is affected by ossification of the anterior longitudinal ligament at multiple thoracic and lumbar levels (T4-T5 and T8-L1) resulting in large 'flowing candlewax' bony growths (Fig. 3). Macroscopic observation and radiography show that disc spaces and joint facets are spared, and intervertebral (disc) heights are maintained (Fig. 4). A compression fracture is seen on T12. In addition, the spine displays osteoarthritis on the left superior apophyseal facets of C4 (marginal osteophytes and subchondral bone degeneration), vertebral osteophytes of mild expression (elevated ring) on the anterior/lateral intervertebral margins of L2-L5, and osteophytes on the costal facets of vertebral

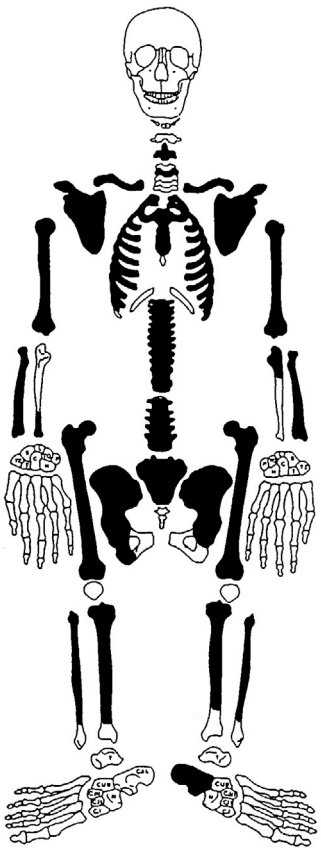

Fig. 2. Skeletal remains attributed to the old male with DISH

Fig. 3. 'Flowing candlewax' ossification on thoracic and lumbar spine (T4-T5 and T8-L1)

Fig. 4. Radiograph of lower thoracic and lumbar vertebrae with DISH. Note the compression fracture on T12

bodies. Fusion occurs at the upper ligamentous portion (bridging osteophytes) of the left sacroiliac joint while the lower two-third part is not involved; no signs of intra-articular ankylosis are observed (Fig. 5). Finally, a new articular facet is formed between the L5 and the sacrum (transverse process) at the area of sacroiliac fusion.

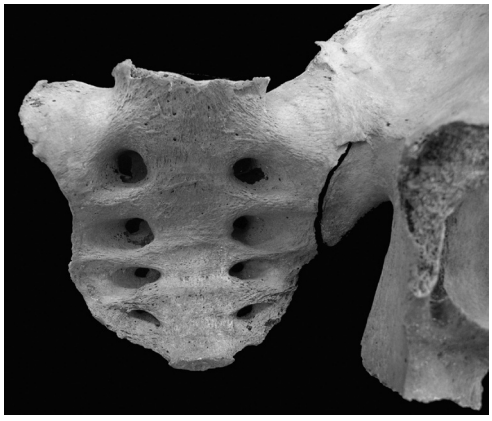

Fig. 5. Bridging osteophytes at the upper ligamentous portion of the left sacroiliac joint. No ankylosis present

Extra-spinal lesions

Extra-axial manifestations include enthesopathies and degenerative changes on joint surfaces. Enthesopathies affect the iliac crests, right ischial tuberosity, pos-

terior (Achilles' tendon insertion) and plantar surface of the left calcaneus, femoral trochanters, linea aspera, and tibial tubercles. The costal cartilage of one left rib (medial shaft) is ossified and the first right cartilaginous costosternal joint is ankylozed. Degenerative lesions affect bilaterally the acromioclavicular and sternoclavicular joints (subchondral bone degeneration and remodeling), shoulder (marginal osteophytes and surface porosity on glenoid cavities and humeral heads), costal facets on vertebral bodies, and elbows (marginal and surface osteophytes on distal humeral epiphyses, and marginal osteophytes of the trochlear notch).

Discussion

Diagnosis

Classic features such as flowing ossification on contiguous vertebral bodies (blocks of two and six vertebrae) and preservation of the inter-vertebral disc spaces with the non-involvement of the facet joints are the spinal criteria met here to propose the diagnosis of DISH (Aufderheide and Rodriguez-Martin 1998; Crubézy 1990; Mader 2003; van der Merwe et al. 2012; Rogers and Waldron 2001); the sacroiliac joint is also affected at the upper (ligamentous) portion (Dar et al. 2007; Olivieri et al. 2009; Rogers et al. 1985; Cammisa et al. 1998; Waldron and Rogers 1990) thus being adopted as pathognomic of the condition. These features are different from those observed in spondyloarthropathy and other joint conditions which are characterized by fusion across the intervertebral spaces, syndesmophytes formation, bilateral sacroilitis, and erosion or fusion of hands and feet (Arriaza 1993; Cawley and Paine 2015; Ortner 2003; Resnick 1976; Rogers et al. 1985; Waldron 2009). The onset of the diseases is also different: DISH is detected after the fifth decade of life while symptoms of ankylosing spondylitis (AS) occur at a young age and rheumatoid arthritis begins between 20 and 50 years of age.

Entheseal ossifications appear in characteristic areas in the examined bones. Extra-spinal enthesopathies often accompany DISH suggesting a systemic skeletal condition (Dar et al. 2007; Mader et al. 2009; Resnick et al. 1975), but they have a multifactorial etiology (e.g. age-correlated) (Villotte 2010) and they cannot be used independently as a diagnostic criterion of DISH (Mader et al. 2009; Van der Merwe et al. 2012; Villotte et al. 2010). In the case of Taxiarches, peripheral enthesopathies appear bilaterally on individuated bones such as limbs and pelvis; the left calcaneus is also attributed to the same individual because of the entheseal changes on the calcaneal tuberosity and plantar surface which, according to the literature, may co-occur in individuals with DISH. Not all extra-spinal manifestations associated with DISH (Resnick et al. 1975) are recorded on the individual from Taxiarches because it has not been possible to reconstruct the whole skeleton.

Finally, DISH can occur with osteoarthritis (OA). Both conditions develop in older age groups (Mader 2003; Mader et al. 2009; Waldron 2009; Ortner 2003) but they are due to different pathogenic mechanisms: the osteoarthritic process affects the cartilage while DISH is manifested by the thickening, calcification and/or ossification of ligaments and entheses (Mader 2003). Although the skeleton examined here is incomplete and no statistically valid patterns can be estab-

lished, all the major long bones, pelvis as well as thoracic and lumbar vertebrae are present. The joints of the long bones exhibit marginal new bone formation and some of them surface osteophytes; in addition, sternoclavicular and acromioclavicular joints and elbows show subchondral bone degeneration. Taking into consideration the advanced age and the co-existence of distinct lesions typical of each condition, it is possible that DISH contributed to the generation of OA in this old male.

DISH and lifestyle

Bioarchaeological literature often suggests that high animal protein and/or calorific diets (e.g. monks' or dignitaries' diet), obesity, comfortable lifestyles and sedentary occupations (e.g. monastic way of life), usually considered as indicative of higher social status, could be predisposing factors of DISH (Jankauskas 2003; Waldron 1985; Roberts and Manchester 2005; Rogers and Waldron 2001; Mays 2006; Rogers 1982; Rogers et al. 1987; Bruintjes 1987; Janssen and Maat 1999). Stable isotope analyses on human skeletons from monastic and non-monastic funerary assemblages in Medieval Europe showed elevated isotope values for the DISH than the non-DISH samples (Spenser 2008; Quintelier et al. 2014) suggesting differences between the two groups although not always detected statistically. These differences should reflect wealthier lifestyles and nutritional status in both monastic and non-monastic, higher status individuals (Mays 2006; Quintelier et al. 2014). Similarly, isotope results obtained by Müldner and Richards (2007) showed that individuals diagnosed with DISH are consistent with a diet rich in animal protein.

Byzantine textual sources and biochemical analyses show that at that time diet was regulated by rigorous eating restrictions mainly because of fasting rules. An average Christian had to fast for at least 170-180 days a year and abstain from several foodstuffs, especially meat. Clergy formed a special segment of the population, and their fasting periods were extended including more days throughout the year (Koder 2005; Koukoules 1955). Furthermore, meat was not included in the everyday menu of the average Byzantines either because it was expensive or because it was difficult to preserve it during high temperature seasons (Koder 2005). Biochemical analysis on Greek Byzantine skeletal series revealed that Byzantine diet was primarily based on C_3 staples (wheat and barley) and domesticated animals that fed on C_3 plants. On the other hand, a number of obtained values suggested substantial consumption of meat or, more likely, dairy products. In some cases, Greek Byzantine populations consumed significant amounts of marine protein that could be due to dietary restrictions and fasting regulations imposed by the Orthodox Church (Bourbou et al. 2011; Koukoules 1952). Similar results are obtained from Medieval European sites where seafood consumption and low caries rates are recorded (see Mays 2006).

Despite the absence of isotopic data for Taxiarches, several indicators infer the health status of these individuals. None of the recovered teeth was affected by linear enamel hypoplasia (usually considered as an indicator of infectious disease, malnutrition/starvation or periods of stress and growth arrests in childhood) or caries (infectious disease process characterized by the focal demineralization of dental hard tissues by organic acids pro-

duced by bacterial fermentation of dietary carbohydrates, especially sugars) (Lukacs 1989). These observations show that at least two individuals (including probably the one with DISH) were not exposed to infections or risks capable to disturb their development during childhood. In addition, high stature is a sensitive indicator of stress and overall health status during growth (Maat 2005; Steckel 1995); it is considered that protein consumption is favorable for stature. The stature values obtained for the old males of Taxiarches are among the highest obtained from Byzantine populations in Greece and, along with age-at-death, suggest the overall wellbeing of these individuals.

DISH and burial location

Historical sources and archaeological evidence attest an intense economic development at the region of Attica during the Byzantine period. The construction, among others, of numerous churches and monasteries suggests the presence of rich landowners who funded the construction of ecclesiastical edifices and foundations (Gini-Tsofopoulou 2001; Kazanaki-Lappa 2002; Kazhdan 1991: 229). This activity laid to the creation of small, local ecclesiastic centers and groups of people with respective duties.

Burials inside churches were common in Byzantium; it is generally argued that the area inside a religious edifice was prestigious and held the inhumation of particular social/professional groups, such as members of the clergy (e.g. monks, priests), high-status citizens and members of powerful families (e.g. the founders of the church, lay benefactors, dignitaries) (Laskaris 2000; Marinis 2009; Teteriatnikov 1984); in the latter case, the right to be buried inside a church was hereditary and transmissible (Emmanouilidis 1989). On the contrary, average people were not allowed to be buried inside churches. At Taxiarches, there is no archaeological evidence for the presence of a monastic community; as such, the possibility that people buried in the church were clergymen is weakened.

Cist graves usually included collective burials and they were considered as of elaborate construction; for this reason they served the needs of individuals from distinct social and/or professional classes (Laskaris 2000; Tritsaroli 2006). The multiple use of a grave was a common Christian custom; the living often exhumed an individual in order to empty the grave for the next deceased and then they redeposited back in the grave the bones of the previous inhumation on the top of the latest deposit or on the edges of the grave (Laskaris 2000). Byzantine cemeteries landscape analysis has shown that the location of elaborate graves inside churches or aligned in front of a church's entrance was indicative of the distinct social status of the deceased (Tritsaroli 2006; Tritsaroli and Valentin 2008). The grave of Taxiarches meets all the above criteria of location and construction for it to be the burial place of an upper class group.

Bone representation pattern shows a complex secondary treatment for the individuals held in the cist grave of Taxiarches. On the one hand, the over-representation of femora, tibiae, fibulae and humeri suggests a practice of bone selection from a different burial place of the densest and most robust bones. In contrast, small-sized bones are quasi-absent; bearing in mind that several factors of bias (e.g. taphonomy, excavation techniques and post-excavation damage) can influence the preservation and representation of skeletal elements from archaeo-

logical contexts, however small elements, such as bones of hands and feet, are not expected to be largely included in a secondary deposit away from the original grave (Andrews and Bello, 2006). On the other hand, it would be unwise to conclude that all the bones found bellow or next to the primary burial were selected and brought from a different place of burial. As far as it can be suggested from the limited archaeological information, the grave was re-used several times; it is possible that at each re-use, the skeleton of the previous inhumation was removed from the grave and then selected bones were redeposited back inside. In sum, the individuation of several skeletal elements showed at least two adults whose skeletons were fairly well represented in the grave that was probably their original place of burial; the remaining bones of at least eight individuals were the outcome of secondary manipulation resulting from retention of certain bones for reburial or even the transfer of some of them from their original place of burial.

Conclusions

The Byzantine church of Taxiarches in Attica comprises the collective burial of at least ten adults, one of which shows skeletal evidence of DISH. Burial location attributes to these individuals a distinct social profile. The practice to inhume inside churches was not uncommon in Byzantine society and was considered as of high status burial location for members of the clergy, local dignitaries, and powerful families. On the other hand, the relationship between DISH and high rank or monastic lifestyles is not straightforward; yet, the overall archaeological context of this burial and the lack of evidence for the presence of a monastic community suggest that these individuals should be identified as of laymen rather than religious. Although juveniles are absent from this context, it cannot be excluded that the grave held (some) members of the same family. The absence of an age-at-death and sex profile for most of individuals hinders further interpretation. It cannot be assumed that because DISH may be related to the monastic way of life or to a high status, the opposite is necessarily true (Rogers and Waldron 2001). Nevertheless, the longevity and stature of this individual are suggestive of good living standards and nutritional status. Finally, this paper shows the potential of human osteology for shedding light on Byzantine burial customs in Greece; the contextual analysis of human skeletal remains and cemetery landscape, even from periods for which textual sources are abundant, can provide valuable information on the social status of the deceased and the behavior of living society towards the dead. It is obvious that Byzantine funerary assemblages have still many things to reveal towards this direction.

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Conflict of interest

The Author confirms that there are no conflicts of interest regarding this publication.

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