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A probable case of diffuse idiopathic skeletal hyperostosis (DISH) from an early modern crypt in Eastern Germany

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Running title
A probable case of DISH from early modern times in Germany

Keywords
Paleopathology, differential diagnosis, diffuse idiopathic skeletal hyperostosis, DISH, early modern times, crypt burial

Abstract
Diffuse idiopathic skeletal hyperostosis (DISH) is a non-inflammatory joint disease mainly characterised by the ossification of the right anterior longitudinal ligament and the presence of enthesopathies. Studies have shown that the disease typically affects males of advanced age. This is a case report of a female individual, aged between 40-60 years, dating to 1472-1635 AD and found in Eastern Germany. A differential diagnosis was completed following macroscopic examination and radiographic imaging of the affected bones. The results show several pathological features that resemble skeletal characteristics of DISH, besides other diseases. Therefore, we discuss DISH and provide a differential diagnosis of additional pathologies. Our case is particularly important because ancient female DISH cases are

This article has been accepted for publication and undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process which may lead to differences between this version and the Version of Record. Please cite this article as doi: 10.1002/oa.2913

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underrepresented and the burial location indicates a possible noble or monastic context, both linked with a lifestyle known to be related to DISH.

1. Introduction

Diffuse Idiopathic Skeletal Hyperostosis (DISH) is a chronic, age-related condition with characteristic new bone growth, especially at the spine and entheses (Ortner 2003). In the spine, DISH produces an attachment of the fibroligamentous tendon to the periphery of the vertebral bodies, with absence of disc disease, sacroiliitis and changes in the diarthrodial joints of the affected vertebrae (Resnick and Niwayama 1976).

The typical spinal changes were first described by Forestier and Rotes-Querol (1950) as "senile ankylosing hyperostosis". Resnick et al. (1975) introduced the term "DISH", further including extraspinal manifestations. Different definitions for DISH have since evolvingly been proposed (see Kacki et al. 2018). Van der Merwe et al. (2012) show that depending on the diagnostic criteria used, its prevalence in skeletal samples is highly variable. Besides that, DISH has often been misidentified as ankylosing spondylitis (AS) early on or not been recognised as co-existing, as published for Ramesses II (Chhem et al. 2004).

Studies range from various eras and regions. For example, probable DISH cases include that of a Paleolithic Neandertal from Iraq (Crubézy and Trinkaus 1992), Egyptian remains from 3000 BC (Rogers et al. 1985), a probable pre-Columbian individual from USA (Ortner 2003) through to clinical cases (e.g. Macía-Villa et al. 2017). In general, cases of DISH occur more frequently in males than in females (Aufderheide and Rodríguez-Martín 1998). Here, we present an unusual case from either a monastic or noble context in late medieval or early modern Germany.

2. Material

We investigated human skeletal remains from the high medieval monastery of Riesa (Saxony) in Germany. The monastery was founded in the 12th century AD and transformed into a nuns' convent in the 13th century, but after its secularisation in 1542 following the Protestant reformation noble families used its church as burial place (Dannenberg and Westphalen 2019).

At the time of investigation, the commingled human remains were stored in a box containing a minimum number of 19 incomplete individuals (MNI). We hypothesize that they originate from a crypt in the northern annexe of the church (Alterauge and Hoffmann 2019). However, it remains unclear whether they have been originally entombed in this crypt (as a noble) or ended up in there during eviction of other burial places of the church (as a nun).

3. Methods

We cleaned the skeletal remains and registered an inventory after Buikstra and Ubelaker (1994). To distinguish the remains of our individual from the assemblage, biological profile, distinctive bone preservation and measurements helped matching bones, adjacent elements were associated by articulation (White et al. 2012). We believe that the associated skeletal elements belong together; elements with insufficient matching criteria were excluded.

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Sex assessment followed Bass (1995), while the pubic symphysis (Brooks and Suchey 1990) and auricular surface (Lovejoy et al. 1985) were used for age-at-death assessment. Stature was calculated from the average of long bones after Bach (1965).
A radiocarbon AMS dating was conducted by the LARA at the University of Bern (Szidat et al. 2017). The most affected bones were radiographed with a mobile device (Examion® PX-20 BT Plus, X-DR portable detector, Toshiba D-124 X-ray tube). We performed a differential diagnosis by consulting palaeopathological and clinical literature (see discussion).

4. Results

In general, the bones are in good state of preservation and the representation is around 50% (Fig. 1). Due to overall gracility and the pelvis, we assessed the sex as female. Assessment of pubic symphysis (phase 4), auricular surface (phase 5) and degenerative joint changes (less than 60 years) resulted in an age assessment of 40 to 60 years. We calculated an average stature of about 159 cm using both humeri, radii, femora and tibiae. The fibula sample dates to 341 +/- 19 uncal. BP, resulting in a calibrated age of 1472-1635 AD (2 sigma).

Palaeopathology

Vertebral column and pelvis

The vertebrae T6-T8 show a continuous, candlewax-like ossification on the right side without affecting intervertebral disc space (Fig. 2a). Ossification of the same ligament is observed on further vertebrae, also ossification of the equivalent posterior ligament in upper thoracic and all lumbar vertebrae. Most vertebrae have laminal spurs, enlarged articular processes and/or facet eburnation and porosity (Fig. 2b), most severe on T3-T5, some exhibit ossification of costovertebral cartilage. Several vertebrae show bone growth at the body margins.
We note the presence of entheseal changes along the pelvic iliac crest in correspondence with ossification of the attachment site of various muscles, most severe in the anterior third (Fig. 3).

Shoulder girdle and upper limbs

The two scapulae show ossifications of the coracoacromial ligament at the coracoid process and the acromioclavicular ligament at the acromion. The glenoid cavity and the acromioclavicular facet show bilaterally strong marginal lipping.
The lateral epicondyles of the humeri show osteophytes, likewise the muscle insertion areas at the lesser and greater tubercle crest and the right radial tubercle. Areas of minor lamellar and smooth, remodelled bone are present at the posterior shafts. Marginal exostoses are present on the left humeral head, the distal articular facet of the right radius and both proximal articular facets of the ulnae.

Lower limbs

The femora present enlarged foveae capitii but no further arthrotic changes on the head. We noted bilateral osteophytes on the greater trochanters, along the intertrochanteric line and linea aspera down to the lower third of the shaft.

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Both tibiae show lamellar bone (fine, longitudinal, pitting striation) with commonly encountered periosteal reactions, concentrated anterior, osteophytes along the popliteal/ soleal line and marginal lipping on the proximal articular facet. The left tibia exhibits extensive periosteal new bone formation on the medial and lower lateral diaphysis (Fig. 4a, b). Both, remodelled and woven new bone are present on large areas, indicating a chronic and long-standing pathological process still active at the time of death. This periosteal reaction is notable on the radiograph of the bone, where cortical remodelling is apparent (Fig. 4a). These images further show radiopaque, non-continuous lines in the distal shaft referred to as Harris lines (Roberts and Manchester 2010).
The talus and calcaneus show marginal lipping at corresponding articulating surfaces. Two inferior calcaneal spurs (1.2x1.7cm anterior and 0.5x1.0cm posterior) are located at the attachment of the plantar fascia (Fig. 5).

5. Discussion

The radiocarbon date of this individual is in accordance with the historical context, when the church belonged first to the convent and later nobles were buried within it. Considering that church burial was a privilege of the social elite, this individual may have been a high-ranked nun, such as an abbess, or a noble.
The most notable pathologies are the hyperostotic entheses and the right-sided fusion of some vertebrae. These features are characteristic for DISH, however, other diseases should be differentiated, for instance ankylosing spondylitis (AS) and osteoarthritis (OA).

DISH

The “candlewax”-like ossification of the anterior longitudinal ligament is the main characteristics of DISH and typically occurs on the right side of the thoracic vertebrae because of the overlying aorta on the left side (Roberts and Manchester 2010). This anterolateral, flowing ossification does not affect the intervertebral space, and absence of changes in the sacroiliac joint are diagnostic criteria for DISH (Kozanoglu et al. 2002). Enthesophytes are commonly but not exclusively associated with DISH, e.g. at the iliac crest, and should only be an additional criterium (Van der Merwe et al. 2012). According to our vertebrae and additional criteria, we diagnose DISH (Resnick and Niwayama 1976; Arlet and Mazières 1985; Crubézy and Crubézy-Ibáñez 1993; Rogers and Waldron 2001) or possible/early DISH (Utsinger 1985; Waldron 2009) in this individual. The prevalence of DISH is higher in males and individuals older than 40 years (e.g. Aufderheide and Rodríguez-Martín 1998), with cases increasing with advancing age. DISH prevalence in Europe is rather high, for instance 22.7% (males) and 12.1% (females) in the Netherlands (Westerveld et al. 2008), 27.3% (males) and 12.8% (females) in Hungary (Kiss et al. 2002a) or lower in Finland with 3.8% (males) and 2.6% (females), but with 10.1% and 6.7% respectively in people from 70 years onwards (Julkunen et al. 1975). Its actual cause is unknown; however, DISH is more prevalent in obese, diabetic and/or gouty individuals (Mays 2009; Kiss et al. 2002b; Giuffra et al. 2017). The disease is commonly associated with ancient individuals of high status or clergymen, which followed a “monastic way of life” (Rogers and Waldron 2001; Jankauskas 2003; Müldner and Richards 2007; Verlaan et al. 2007; Bondioli et al. 2016; Piombino-Mascali et al. 2017; Kacki et al. 2018).

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The burial location of this individual indicates a possible monastic or noble background, both associated with a diet rich in animal protein (e.g. Polet and Katzenberg 2003; Giuffra et al. 2010). However, we did no stable isotope analysis on the individual's remains to confirm this diet. The reason was the lack of a skull, which would have resulted in doubled testing on the crania from the commingled assemblage.

Ankylosing spondylitis

Individuals with coexisting DISH and AS have been reported (Kuperus et al. 2018). The main characteristic of AS is the involvement of the sacroiliac joint which starts to erode and finally fuse (Roberts and Manchester 2010). Another feature of AS are syndesmophytes and ankylosis of the vertebrae by ossification of the annulus fibrosus, sometimes called "bamboo spine" (Braun and Sieper 2007). None of these conditions were found in our individual. Conclusively, AS is not diagnosed.

Osteoarthritis (OA)

Some changes cannot be assigned to DISH but osteoarthritis (OA), such as lipping of the glenoid, ossifications at the costovertebral joints and alterations of the vertebral articular facets (Fig. 2b). Eburnation, porosity and new bone formation are diagnostic for OA, which follow joint pain starting in the third decade of life with increasing severity over time (Ortner 2003). Primary OA naturally occurs with degradation of cartilage whereas secondary OA is attributed to other causes (Waldron 2009). Because OA and DISH show higher prevalence with increasing age, it is likely to find them simultaneously (Rogers and Waldron 2001). Considering the lesions and age (40-60 years), we might diagnose OA in our case, even though hip and knees are barely affected.

Periosteal reactions

The striations on the tibiae and humeri are consistent with reactions to a periostosis (Roberts and Manchester 2010). Periostosis is a non-specific bone reaction including periostitis, which is only related to infection (Roberts 2017). The size and diffuse nature of the lesions in the left tibia likely indicate a systemic pathological condition (Weston 2008), also, the radiographic images show no trauma. We therefore refer to periostitis, leaving traces in form of pits, striae and new bone formation on the cortical bone (Roberts and Manchester 2010).

6. Conclusions

We examined the skeletal remains of a 40-60-year-old female from Germany, dating to 1472-1635 AD. The reported skeletal lesions are consistent with DISH, along with osteoarthritis and periosteal new bone formation on the tibia. DISH is more prevalent in males than females as shown in clinical (e.g. Westerveld et al. 2018; Kiss et al. 2002a) and paleopathological (e.g. Jankauskas 2003) literature, making this case atypical. However, the burial ground within the cloister church indicates a likely monastic or noble background. Both are associated with a “monastic” diet connected to a higher prevalence of DISH.

Since the exact causes of DISH are still unknown, case studies such as ours can help to understand the aetiology of the condition and make our study a valuable addition to the paleopathological literature. Identifying a possible female skeleton from late medieval or

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early modern times with DISH and associated osteoarthritis, strengthening the relation between the onset of DISH and a high social status or monastic lifestyle.

Acknowledgement
We thank the reviewers for their constructive comments, the parish of Riesa for access to the bones and further information, and Dr. Lucie Biehler-Gomez for assisting with paleopathology concerns.

Conflict of interest
None

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Figure 1: Preserved bones of the examined individual.

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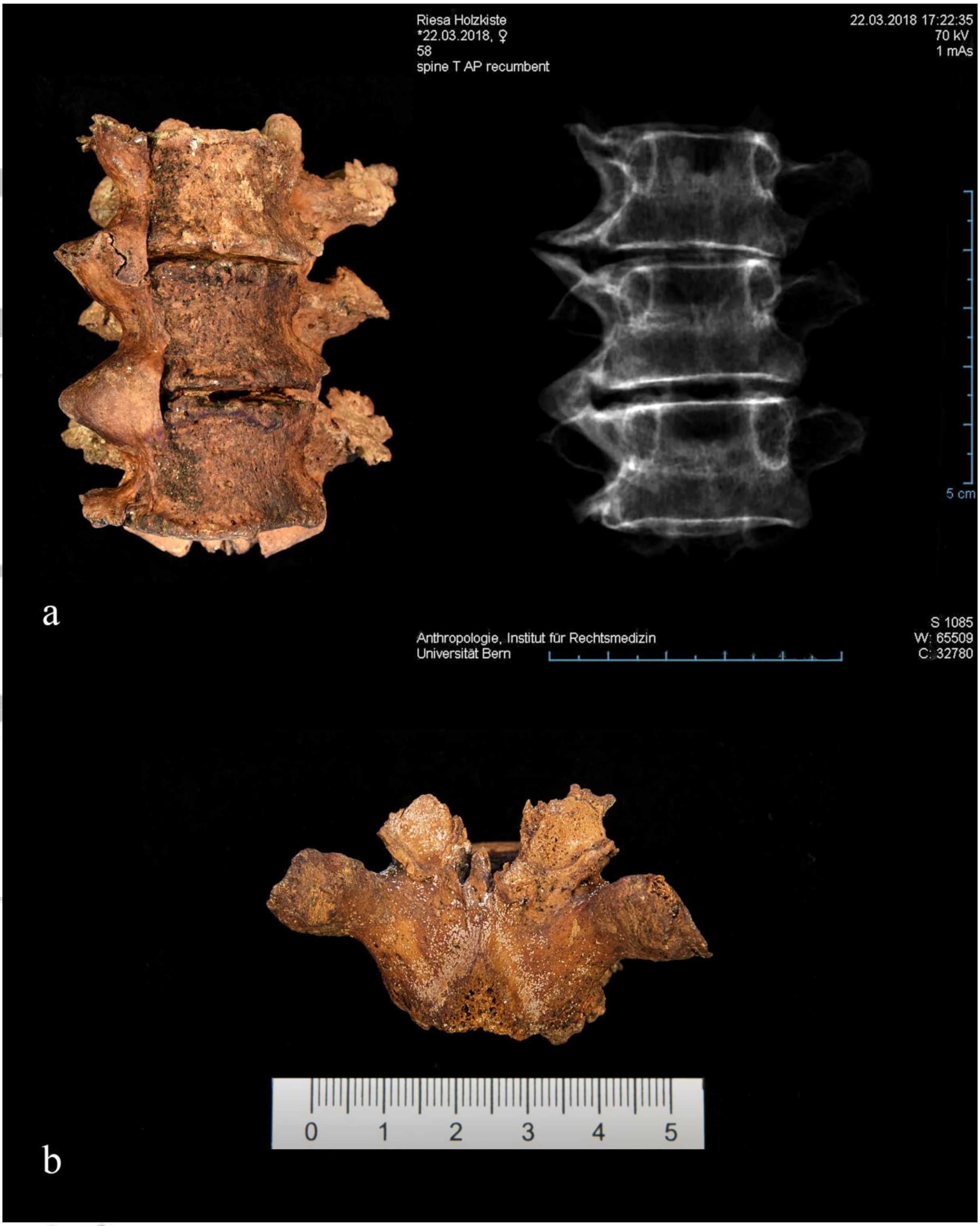

Figure 2: Overview a) Anterior photograph and radiographic image of T6 to T8, showing the candlewax like ossification on the right side of the vertebrae and ossification of costovertebral cartilage. Detail b) the posterior view of a thoracic vertebra with subchondral pitting and osteophytes in the superior articular facets.

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Figure 3: Posterior view on the left innominate with osteophytes along the iliac crest.

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Figure 4: Overview a) Anteroposterior and mediolateral photographs and corresponding radiographic images of the left tibia, showing extensive new bone formation on the medial and the lower lateral diaphysis. Detail b) Close-up photograph of the periosteal changes of the tibia, showing woven as well as remodelled bone.

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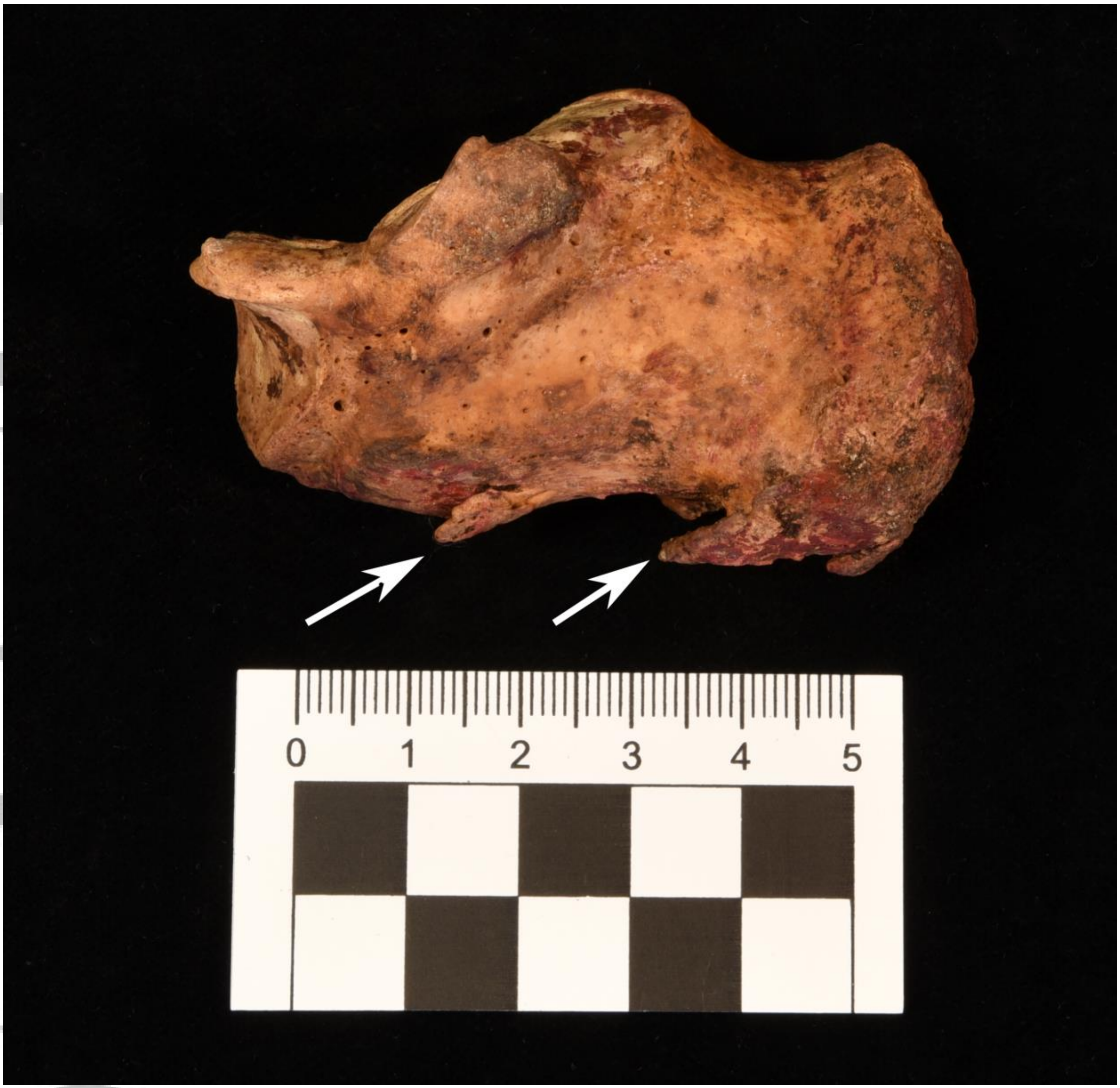

Figure 5: The right calcaneus exhibiting entheseal changes (calcaneal spurs).

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