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Diffuse idiopathic skeletal hyperostosis (DISH) in a middle Holocene forager from Lake Baikal, Russia: Potential causes and the effect on quality of life

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A B S T R A C T

Diffuse idiopathic skeletal hyperostosis (DISH) is often thought of as a disease of modern culture and lifestyle; however, cases have been reported in antiquity, although relatively sparsely. Using an osteobiographical approach, this study presents the oldest known case of DISH in Asia, a Kitoi individual, 76.1, from the middle Holocene cemetery of Shamanka II (Siberia, Russia). Rather than merely reporting a case of DISH, we explore the question: what would predispose a forager to develop a disease that is frequently associated with obesity and metabolic disorders? Evidence for physique, activity patterns, diet, and status are examined, comparing 76.1 to other individuals of his cultural group, in order to better understand why this individual may have developed DISH, as well as the potential effects of the disease on his quality of life. Results indicate that 76.1 was likely as active as other foragers belonging to his cultural group and, as can be discerned, his diet was similar; however, his physique differed, particularly his body mass estimate. Additionally, mortuary indicators suggest that 76.1 was of lower status than many of the other individuals buried at Shamanka II. Based on these multiple lines of enquiry, several scenarios are proposed as to what contributing factors resulted in 76.1's pathological state, including diet, physiological stress, the influence of status, dietary and genetic variation. Based on modern studies of quality of life in DISH individuals, as well as a lack of evidence for prolonged inactivity or disability, 76.1 did not appear to be significantly affected by the disease.

1. Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is a disease of antiquity (Crubézy and Trinkaus, 1992; Trinkaus et al., 2008), the aetiology of which is unknown; however, metabolic disease, genetics, and/or mechanical factors may all contribute. The prevalence of the disease varies by ancestry (Spagnola et al., 1978; Bloom, 1984; Cassim et al., 1990; Rothschild and Woods, 1991; Weinfeld et al., 1997; Kiss et al., 2002; Kim et al., 2004) and in archaeological collections, studies suggest that those of higher status and/or those consuming more calorically-rich diets were more susceptible to developing DISH (e.g., Waldron, 1985; Janssen and Maat, 1999; Rogers and Waldron, 2001; Jankauskas, 2003; Verlaan et al., 2007; Giuffra et al., 2010). However, the documentation of DISH in the prehistoric record is relatively sparse, which limits a better understanding of the geographic and temporal distribution of the disease. This paper reports on the earliest case of DISH in Asia, unusual in the fact that it is identified in a forager. This individual was a Kitoi, Forest Neolithic male who died approximately 6914 ± 35 years before present (BP) (Weber et al., in this volume). In addition to diagnosis, this paper considers why this prehistoric forager developed DISH, as well as the disease's impact on his quality of life. In order to accomplish this goal, 76.1 is analysed within the context of other Kitoi foragers and using a multifactorial approach.

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2. Background

DISH was first systematically analysed by Forestier and Rotés-Querol (1950) and is commonly associated with aging, found primarily in individuals over the age of 40 years, with males disproportionately affected (Forestier and Lagier, 1971; Julkunen et al., 1971; Utsinger et al., 1976; Bloom, 1984; Rothschild and Woods, 1991; Weinfeld et al., 1997; Cammisa et al., 1998; Westerveld et al., 2009). DISH is frequently described as a condition in which entheses are targeted; however, it may result in the ossification of other soft tissue structures, such as fasciae and joint capsules (Cammisa et al., 1998). In the spine, this process begins toward the middle of the vertebral body, where entheses are located, and spreads inferiorly and/or superiorly, eventually transgressing the intervertebral disk space (Utsinger, 1985). At extraspinal sites, the process results in spurs or other forms of protuberances (Cammisa et al., 1998).

Clinical and palaeopathological diagnostic criteria focus largely on spinal changes (Forestier and Rotés-Querol, 1950; Forestier and Lagier, 1971; Resnick and Niwayama, 1976; Utsinger et al., 1976; Julkunen et al., 1981), specifically ossification of the anterior longitudinal (ALL) and paravertebral ligaments. Progression of the disease results in a melted candlewax appearance to the spine and, if allowed to progress to the more extreme manifestations of the disease, vertebral excrescences articulate and may fuse. In the thoracic column, this is largely limited to the right half of the ALL, due to the prophylactic nature of the descending aorta (Nathan, 1962; Bahrt et al., 1983). However, in the cervical and lumbar columns, the pattern is more symmetric. Studies in modern populations indicate that the symptoms range from asymptomatic, to generalised stiffness and swelling, to more acute problems, such as dysphagia and spinal cord immobilisation, particularly when the cervical spine is involved (Forestier and Lagier, 1971; Utsinger et al., 1976; Bahrt et al., 1983; Utsinger, 1985; Schlapbach et al., 1989; Mata et al., 1997; Cammisa et al., 1998).

DISH may also involve extraspinal sites, such as the ulna, os coxa, patella, and calcaneus (Resnick et al., 1975; Brigode et al., 1982; Littlejohn and Urowitz, 1982; Utsinger, 1985; Fahrer et al., 1989; Haller et al., 1989), where excrescences are produced at the insertion sites for *triceps brachii*, Achilles, and quadriceps tendons, as well as the iliolumbar and sacrotuberous ligaments (Brigode et al., 1982; Littlejohn and Urowitz, 1982; Cammisa et al., 1998; Rogers and Waldron, 2001). Manual changes have also been noted, including hyperostotic bone growth of the metacarpals and phalanges, and tufting of the distal phalanges (Resnick et al., 1975). Notably, the extraspinal pattern is frequently bilateral (Littlejohn and Urowitz, 1982; Utsinger, 1985), which can aid in excluding seronegative spondyloarthropathies (Jankauskas, 2003).

The aetiology of DISH remains somewhat enigmatic, although most theories focus on aspects of metabolism, obesity, and genetics. In the clinical literature, DISH is often co-incident with obesity (e.g., Forestier and Lagier, 1971; Julkunen et al., 1971, 1981; LittleJohn and Smythe, 1981; Mata et al., 1997; Di Franco et al., 2000; Kiss et al., 2002; Sarzi-Puttini and Atzeni, 2004; Denko and Malemud, 2006; Zincarelli et al., 2012), hyperinsulinemia (LittleJohn and Smythe, 1981; Littlejohn and Hall, 1982; Denko et al., 1994; Denko and Malemud, 2006) and type II diabetes (Denko et al., 1994; Kiss et al., 2002; Mader and Lavi, 2009; Mader et al., 2009), as well as hyperuricaemia (Kiss et al., 2002; Sarzi-Puttini and Atzeni, 2004) and dyslipediemia (Vezyroglou et al., 1996). The frequent association of these various diseases with DISH suggests that it is a general manifestation of metabolic dysfunction; however, these conditions are not necessarily requisite for developing DISH, nor do they guarantee development of the disease (e.g., Vezyroglou et al., 1996; Sencan et al., 2005). Genetic studies have yet to identify a locus for DISH, although HLA B27 and COL 11 A2 have been excluded (Brigode and Francois, 1977; Ercilla et al., 1977; Spagnola et al., 1978; Havelka et al., 2001). Support for a genetic predisposition to DISH stems from population differences in its prevalence (Bloom, 1984; Cassim et al., 1990; Weinfeld et al., 1997; Kim et al., 2004), an association of COL6A1 with DISH in a Japanese sample (but not in a Czech sample) (Tsukahara et al., 2005), and the expression of DISH in multiple family members (Havelka et al., 1990; Bruges-Armas et al., 2006).

Several authors have argued that DISH may be caused by mechanical factors (Nathan, 1962; Julkunen et al., 1971; Littlejohn, 1985; Pappone et al., 1996), a result of microtrauma and subsequent entheseal ossification. It is possible that this is particularly relevant to "bone formers" (Resnick and Niwayama, 1976; Rogers et al., 1997), or individuals characterized by genetic differences in enzyme expression that result greater bone formation (e.g., Ohishi et al., 2003). However, it is also possible that bone formers are individuals characterized by metabolic diseases that alter gene expression and the subsequent osseous response (e.g., Littlejohn, 1985), although contradictory results have been found in other sample populations (e.g., Julkunen et al., 1981; Pappone et al., 1996).

In the osteoarchaeological literature, researchers suggest that a calorically-rich diet, leading to obesity and type II diabetes, may facilitate the onset of the disease (Rogers and Waldron, 2001). Classic studies focus on religious clergy (Waldron, 1985; Janssen and Maat, 1999; Rogers and Waldron, 2001; Verlaan et al., 2007), known for their excessive meals and low levels of physical activity. Similarly, DISH is also found in high status Italians (Giuffra et al., 2010) and Lithuanians (Jankauskas, 2003), identified as having richer diets paired with less exertional lifestyles. Stable isotope analyses lend support to a link between diet and DISH in elite Italians (Fornaciari, 2008; Fornaciari et al., 2009), Medieval monks and their benefactors (Müldner and Richard, 2007; Spencer, 2008, 2009), and non-monastic Medieval groups (Spencer, 2008, 2009). However, although a diet rich in meat and calories may be associated with a particular status (e.g., elite, clergy), stable isotope analyses of diet do not always differentiate between those affected and unaffected by DISH (e.g., Müldner and Richard, 2007; Spencer, 2008, 2009), and DISH alone should not be considered an indicator of high status or occupation (Rogers and Waldron, 2001; Jankauskas, 2003; Spencer, 2008, 2009). Additionally, DISH has been identified in active and less socially stratified groups, such as prehistoric maritime foragers in South America (Arriaza, 1993) and Japan (Oxenham et al., 2006).

DISH is rare in the Asian bioarchaeological literature, having been diagnosed in only a few instances in prehistoric remains (e.g., Oxenham et al., 2006; Kim et al., 2012). Individual 76.1, the focus of this paper, represents the earliest case of DISH in Asia. Using multiple lines of evidence, including estimates of physique, a stable isotope analysis of diet, activity induced dental modification and musculoskeletal markers relating to physical activity, and mortuary indicators of status, this paper considers why 76.1 developed DISH, as well as how may it have impacted his quality of life.

3. Materials

3.1. Shamanka II

During the middle Holocene, Forest Neolithic foragers inhabited the Lake Baikal region, with mortuary traditions that resulted in at least some of the dead being buried in relatively large and formalized cemeteries. Shamanka II (Fig. 1), excavated by V.I. Bazaliiskii, is located at the southern tip of Lake Baikal. A total of 165 individuals were disinterred from 107 burial pits, with all life stages represented. Burials at Shamanka II are primarily associated with the Kitoi (n = 96), a Forest Neolithic group, who utilized the

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