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International Journal of Paleopathology 19 (2017) 53–63

Contents lists available at ScienceDirect

International Journal of Paleopathology

journal homepage: www.elsevier.com/locate/ijpp

Osteochondritis Dissecans of skeletal elements of the foot in a 19th century rural farming community from The Netherlands

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ARTICLE INFO

Keywords:
Physical activity
Foot bones
Trauma
Footwear
Post-medieval
The Netherlands

ABSTRACT

Osteochondritis Dissecans (OD) is a pathological condition of the subchondral bone and surrounding cartilage of synovial joints, associated with strenuous activity and/or trauma. Reports of OD in archaeological skeletal remains are few and the majority demonstrate low OD prevalence (< 1%). A predominantly 19th century skeletal sample from Middenbeemster, the Netherlands, was assessed for OD. The sample included adult individuals of both sexes. There were no definitive OD lesions in non-pedal elements, yet 12.9% of individuals suffered from pedal OD. Few archaeological and clinical reports specify the prevalence of pedal OD. According to the few that do, the Middenbeemster pedal OD prevalence is distinctly high. Several factors could have contributed to this. First, the rural Beemster community was centered around cattle farming, requiring extensive outside work and animal maintenance; thus, increasing the chances of acute/repetitive trauma in the foot. Second, the footwear worn during that period in the Netherlands was the wooden clog. It is suggested that the hard and inflexible clog, which is poor at absorbing shock and limits the movement of the foot, could have resulted in repetitive microtrauma. These two factors combined may have caused a high frequency of OD.

1. Introduction

1.1. Osteochondritis Dissecans

Osteochondritis Dissecans (OD) is a specific bone lesion correlated by researchers to trauma, either acute or repetitive microtrauma, resulting from vigorous activity (Detterline et al., 2008; Ortner, 2003; Schindler, 2007; Waldron, 2009; Wells, 1974). It affects the subchondral bone and surrounding cartilage of synovial joints. This could lead to the partial or complete detachment of articular cartilage, or both cartilage and subchondral bone, resulting to a loose-body formation within the joint cavity (Schindler, 2007). The fragment detachment area may heal over time, undergoing remodeling with lesion borders becoming sclerotic (Aufderheide and Rodríguez-Martín, 1998). However, when continuous loading is imposed, the healing mechanisms of bone, and thus its remodeling, may decrease leading to bone necrosis (Schindler, 2007). OD frequently occurs in athletes (Aichroth, 1971) and particularly throwing athletes and gymnasts (Baker and Romeo, 2010; Schenk and Goodnight, 1996; Wahegaonkar et al., 2007). In the ideal case of early detection and treatment of the lesion, the repair mechanism, especially in juvenile patients may be entirely successful; yet in most cases it is not (Schindler, 2007). The lesion may eventually be covered by a thin layer of bone, but its surface will remain depressed (Aufderheide and Rodríguez-Martín, 1998; Ortner, 2003).

1.2. Location and classification

OD can develop in any synovial joint. The highest frequency is usually observed in the knee, followed by the elbow, ankle, hip, shoulder, and finally the wrist (Waldron, 2009). Clinical cases document OD lesions on concave bony surfaces such as the posterior surface of the tarsal navicular bone (Bui-Mansfield et al., 2000) and the glenoid cavity (Gogus and Ozturk, 2008). When OD occurs in the foot, the talar dome and the distal end of the first metatarsal are the articular surfaces most frequently affected (Camasta et al., 1993).

In Aichroth's (1971) research of OD in the knee, 69% of the lesions in 105 patients from the Royal National Orthopaedic Hospital occurred on the lateral posterior site of the medial femoral condyle. Thus, this is described as the "classical OD" location. Classification systems for OD lesions located in other joints are scarce, largely because most publications describe single cases (Bauer et al., 1987; Bojanić et al., 2011). Lesions are usually unilateral, but can also occur bilaterally and sometimes in a symmetrical position. Lastly, it is possible for lesions to occur in different joints of the same individual (Detterline et al., 2008; Schindler, 2007).

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http://dx.doi.org/10.1016/j.ijpp.2017.09.005
Received 18 September 2016; Received in revised form 18 September 2017; Accepted 28 September 2017
Available online 26 October 2017

1.3. Etiology

The etiology of OD is not fully understood but trauma, either major or repetitive microtrauma, is certainly among the main causes (Schenk and Goodnight, 1996; Schindler, 2007). Other postulated causes include insufficient blood supply of subchondral bone (ischemia, avascular necrosis) (Hughston et al., 1984; Schindler, 2007), and accessory ossification centers within the epiphyses causing abnormal ossification (Schindler, 2007). Heredity may also contribute to OD occurrence since, according to some studies, members of the same family developed the pathology (Andrew et al., 1981; Stougaard, 1964; Tobin, 1957). However, the cause may be due to shared environment (Ponce, 2010). Despite the frequent appearance of OD in athletes, which supports the repetitive trauma etiology, it is agreed by several researchers that a multifactorial etiology is most appropriate (Schenk and Goodnight, 1996; Schindler, 2007).

1.4. OD in clinical research

OD is considered a rare pathology with an occurrence of 15–30 out of 100,000 people (Gogus and Ozturk, 2008). Clinical studies demonstrate that OD can occur at any age (Schenk and Goodnight, 1996). The highest peak occurs in adolescence (Schenk and Goodnight, 1996; Schindler, 2007), with the age ranging from 10 to 25 years according to Aufderheide and Rodríguez-Martín (1998) or 13–21 according to Detterline et al. (2008). OD is usually observed in males more often than females (Aufderheide and Rodríguez-Martín, 1998; Ortner, 2003; Ponce, 2010; Schenk and Goodnight, 1996; Solomon et al., 2010; Waldron, 2009), regardless of the joint involved. This could be due to the fact that men generally engage in more strenuous activities, and tend to be more athletic (Ponce, 2010), which consequently would imply a higher probability of trauma, a factor heavily correlated with OD. In clinical studies, there could be an underestimation of the occurrence of OD because the early stages of the condition can be asymptomatic (Resnick and Goergen, 2002). In addition, Bauer et al. (1987) suggest that OD is underdiagnosed because of visualization difficulties with radiology and due to possible healing.

The majority of OD cases reported in clinical literature involve athletes. Examples include Wahegaonkar et al. (2007) and Baker and Romeo (2010) of capitellar OD in baseball players and gymnasts, Koike et al. (2008) of OD in the glenoid cavity of a 22-year-old professional baseball player, and Mahirogullari et al. (2008) of OD in the humeral head of a 15-year-old baseball player. Vanthournout et al. (1991) refer to a 12-year-old tennis player girl, who suffered from OD in the distal end of the humerus. Aichroth's (1971) extensive follow-up research of OD in the knee, demonstrates that more than 60% of the patients' athletic engagement was evaluated from good to excellent and that 46% of them suffered significant trauma on the knee joint. The mean age was 18 years with the full range extending from six to 53 years.

With respect to pedal OD, the clinical literature records that the dome of the talus is most frequently affected, representing 4% of the total general OD cases (Bui-Mansfield et al., 2000; Kadakia and Sarkar, 2007; Santrock et al., 2003; Schuh et al., 2009; Steinhagen et al., 2001). Although trauma is the most likely etiology (Santrock et al., 2003; Steinhagen et al., 2001), Bauer et al. (1987), in their follow-up study on 30 patients, suggest that a circulatory factor could be implicated due to the high prevalence of bilateral lesions (7/30). Publications of OD involving other foot bones are few and they include case reports such as the medial cuneiform (Anderson, 2002), the navicular bone (Beil et al., 2012; Bui-Mansfield et al., 2000; Ozturk et al., 2008), and the 1st metatarsal (MT1) (Bojanić et al., 2011).

1.5. OD as an indicator of past cultural practices

A large post-Medieval (mostly early to mid 19th century) Dutch cemetery in the municipality of Beemster (Fig. 1), is assesed in order to evaluate the relationship between cultural practices and resorptive lesions in the bones of the foot. This may improve our understanding of lesion development that may be tied to physical activity related skeletal trauma. When OD is observed in archaeological skeletal remains, it could be indicative of equivalent types of strenuous activity. For example, Wells (1974) analyzed ancient skeletal material from the British Isles and reports a higher frequency of OD in Romano-British and Anglo-Saxon samples, compared to those of the Bronze Age period. Wells hypothesized that as the areas which the former groups inhabited were rather inhospitable and had to be cleared for agriculture, it lead to trauma-related OD development owing to usage of poor footwear and inefficient equipment. Wells (1974) then postulated that the low frequency of OD in the Bronze Age samples suggests pastoralism as the dominant way of life.

1.6. The Beemster site

The Beemster polder, a piece of land reclaimed from the North Sea and a World Heritage Site announced by UNESCO in 1999, was inhabited by small, rural communities which focused heavily upon dairy farming and less on market and subsistence gardening (De Jong et al., 1998; Falger et al., 2012). The Beemster landscape, is characterized by a central village, the Middenbeemster, and its small conglomerates/hamlets West-, Noord- and Zuidbeemster. Prior to the late 19th century, a lot of walking was required from the farmsteads to the central village where school, markets and other central facilities were located, as well as for the workers living in the village to deliver services back to the farmsteads. In the Netherlands, agricultural industrialization did not occur until the latter half of the nineteenth century, thus mostly after the time-period of the Middenbeemster cemetary sample (Drukker and Tassenaar, 1997; Van Zander, 1996). Historical data and osteoarchaeological research indicate that people engaged in high levels of strenuous manual labour (Gerard et al., 2002; Lindeboom et al., 2012; Palmer, 2012; Saers, 2012).

1.7. Goals of current study

Archaeology is always looking for new and better ways to reconstruct past human behaviour, and the occurrence of pathological lesions in the foot may be a valuable indicator of activity, trauma, and as will be presented, footwear, in this case the wooden clogs, '*klompen*', commonly worn in the Netherlands in the Medieval and post-Medieval periods. The main aim of this study is the examination of a high prevalence of foot lesions in a post-Medieval population from the Beemster polder of the Netherlands, in order to illuminate aspects of past activity and behaviour.

2. Materials and methods

2.1. Analyzed sample

Middenbeemster is one of the villages of the municipality of Beemster, in the province of Noord-Holland. Inhabitants from the whole of Beemster were interred in the Middenbeemster cemetery of the Keyser church from AD 1623–1866. From the last period of cemetery use, between 1829 CE and 1866 (Falger et al., 2012), archival documents including, death records, are available for many of the skeletons. The excavated skeletal sample came mostly from that latter time period. Most graves contained complete, or nearly complete, and well preserved skeletal remains. Males and females are equally represented and individuals of all ages (fetal to > 90 years) were buried in the cemetery (Lemmers et al., 2013).

One hundred and forty skeletons comprised of 63 males, 69 females, and eight unsexed older subadults, were examined for the presence of foot lesions. The sample was divided into four age-at-death groups; subadults (13–17 years), young adults (18–34 years; which can be

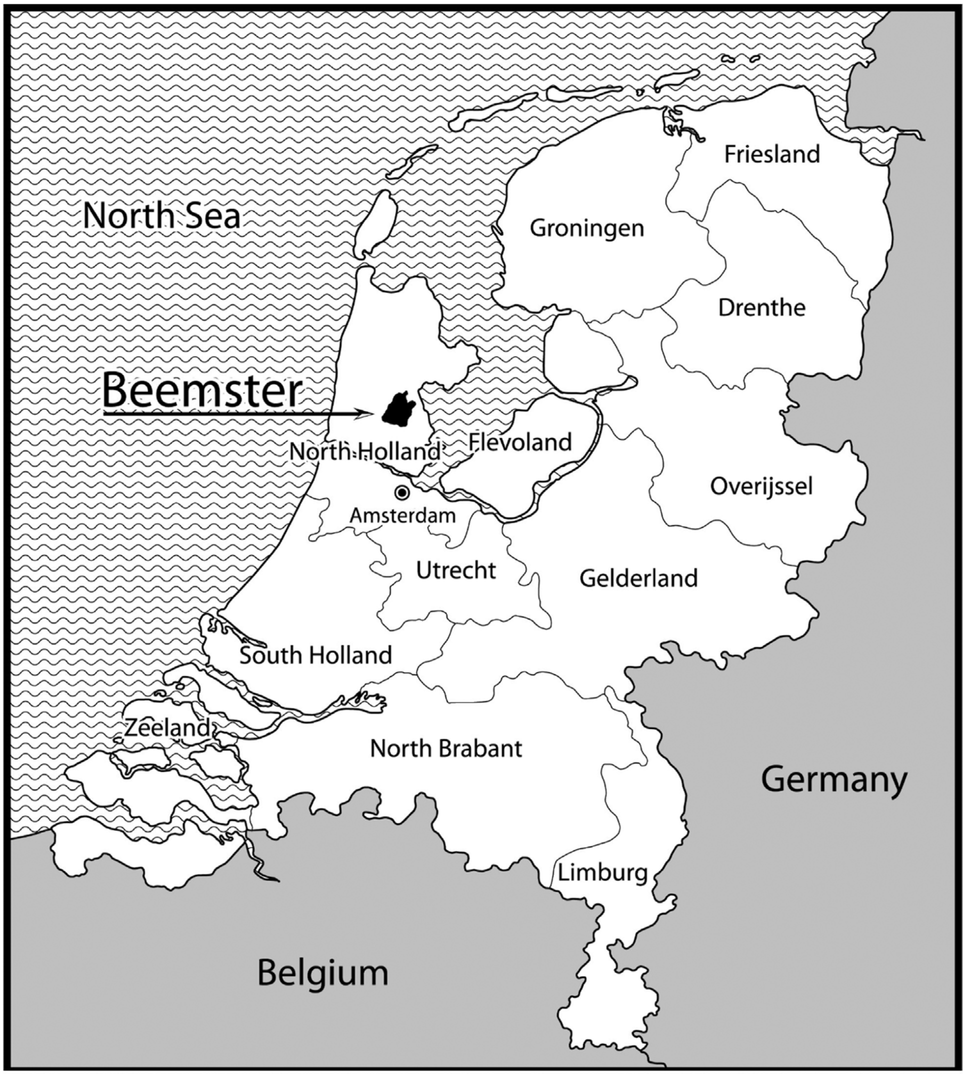

Fig. 1. Map of the Netherlands indicating the location of Beemster.

divided into early young adults aged 18–25 years and late young adults aged 26–34 years depending on the fusion of late-closing epiphyses), middle adults (35–49 years), and old adults (50+ years). Age-at-death and sex estimations were based upon the methods outlined in the Buikstra and Ubelaker (1994) and the WEA (1980) standards. The preservation of most analyzed skeletons was good to excellent, with nearly all skeletal elements present. The subadults had poorer preservation and were less complete. Furthermore, owing to the comparatively small size of the group, subadults were excluded from statistical analyses. Therefore, the statistical analysis was based on 132 adult (18+ years) individuals.

2.2. Differential diagnosis of joint lesions

Osteochondritis Dissecans (OD) results in the formation of a depressed surface of the affected area, resembling the shape of a unilocular "crater" (Solomon et al., 2010). The size of the depression varies depending on the location. Generally the initial defect is 10–20 mm in circumference and up to five mm deep (Aufderheide and Rodríguez-Martín, 1998). The edges of the depression of an OD lesion are smooth and well defined, and the underlying trabecular bone is exposed (Aufderheide and Rodríguez-Martín, 1998). Over time, healing may result in a layer of thin bone forming over the exposed trabeculae but the crater-line indentation will remain (Aufderheide and Rodríguez-Martín, 1998; Ortner, 2003).

Caution must be taken to differentiate pathological from non-pathological lesions, including taphonomic damage and normal anatomical variation. Taphonomic damage is relatively easy to distinguish because it usually causes a break of irregular shape, with sharp, ragged margins, which, if due to recent post-mortem breakage, will have a different color than the surrounding bone. There will be no signs of healing. The generally good to excellent preservation of the analyzed sample enables easy recognition and exclusion of taphonomic damage, and in fact no such damage (in any way similar to an OD lesion) was encountered. In this study, an anatomical variant that was excluded was bony imprints/indentations, which occur in the joint surface due to the morphology of overlying tissues of the synovial joint (vascular and avascular). These will lack a characteristic feature of OD, exposure of trabecular bone on the floor of the defect, and usually have more poorly defined margins, and are thus usually easily excluded (Livingstone, 2003). However, as healed OD also lacks visible trabeculae on the floor of the defect, it may not be possible to definitely distinguish it from an anatomical variant; in such cases the lesion is scored as possible OD and not used within the statistical analysis.

It is also important to exclude pathological lesions resulting from other causes, such as joint or infectious diseases. Osteoarthritis (OA) is a common pathological condition of joint surfaces that results in a range of lesions, including the subchondral cyst, a lytic, periarticular, typically rounded and well defined lesion (Waldron, 2009). OA is usually quite easy to diagnose, identified by diffuse pitting/porosity, osteophytic lipping and outgrowths, joint contour deformation, and/or eburnation on the joint surface (Waldron, 2009). To ensure that subchondral cysts were not included in the count of lesions diagnosed as OD, only joints without OA are considered. Finally, some joint diseases, i.e. gout, rheumatoid arthritis (RA), psoriatic arthritis, and infectious diseases, i.e. osteomyelitis, tuberculosis (TB), can cause erosive lesions that can affect the joint surface. Gout, RA and psoriatic arthritis are characterized by specific affectation locations and appearances, as well as additional bony changes (Aufderheide and Rodríguez-Martín 1998; Waldron, 2009), that permit easy distinction from OD. Infectious diseases are generally easy to distinguish from OD as they rarely begin forming on the joint surface, are often found in multiple non-synovial joint locations, and the lesions have a more destructive appearance with undercut edges and sharp or scalloped ridges (Waldron, 2009).

2.3. Methodology

To our best knowledge, there are no specific guidelines and/or criteria for identifying OD lesions in dry bone from archaeological material. The assessment of both pedal and non-pedal OD in the Middenbeemster sample was based on the criteria given in clinical literature.

Joint surfaces of the shoulder, elbow, hip, knee, ankle, and foot were assessed macroscopically (Table 1). In the foot, specifically the following bones were assessed when present: calcaneus, talus, navicular, the 1st metatarsal (MT1) and the 1st proximal phalanx. Naturally, in some individuals, observation of all bones comprising a particular joint was not feasible, because a bone was absent or poorly preserved. Skeletons that had skeletal deformities that would have affected their range of movement or gait were excluded from analysis. Skeletons with evidence of nutritional deficiencies, i.e. vitamin D deficiency, anemia, were also excluded from analysis. These are the steps followed to detect pathological lesions in the skeleton:

1) Visual examination of well-preserved synovial joints. Joints are evaluated as well-preserved if more than 75% of the joint surface is present and not heavily affected by taphonomic damage. Some bones have more than one possible surface where a lesion could occur. For example, the MT1 can be affected either on the proximal end (base, concave surface) or on the distal end (head, convex surface). In this case, when at least one of the two surfaces are present and observable, it is assessed for the presence of a pathological lesion.
2) OD lesions were classified as definite when all alternative causes (see Section 2.2) could be completely ruled out. Lesions were assessed as possible OD, rather than definitive, when there was a round, crater-like depression in the joint surface but there were no observable exposed trabeculae on the floor of the defect. Possible OD lesions were excluded from statistical comparisons.
3) Each joint surface is scored as having an OD lesion present (P) or absent (A). Missing skeletal elements are recorded with an (M) and when a joint is present but not observable it is indicated as (n.o.).

2.4. Statistical analysis

SPSS 19 was used for statistical analysis. The statistical tests used were the chi-square test or Fisher's exact test if one of the samples is small ($n \leq 5$). Statistical significance was set at $p \leq 0.05$.

3. Results

3.1. Diagnosing OD in the Middenbeemster sample

The good to excellent preservation of the Middenbeemster skeletons helped in identifying OD lesions with the naked eye, making the diagnosis quite straightforward. More specifically, the size of the OD lesions in the foot bones ranged from 0.5 mm (on a base of a 1st proximal phalanx) to 10 mm (on an inferior talar surface) in circumference. OA can be a secondary effect of OD (Aufderheide and Rodríguez-Martín, 1998; Schindler, 2007; Waldron, 2009) yet, in this collection only one individual, a middle adult female, had signs of OA in an affected joint (Fig. 6). The OA modification is slight, below the criteria of definitive OA as per Waldron (2009), with only marginal lipping on the first proximal phalanx. Other signs of OA such as porosity, joint contour deformation, and eburnation were missing.

Round crater-like depressions with no exposed trabeculae on the floor of the defect were observed in the glenoid cavity of five individuals (5/132) and the tibial plafond of four individuals (4/132). This may be because of healing of the OD or due to a non-OD depression such as that created by anatomical variants from the tissues within and around the joint. According to clinical literature, both the glenoid cavity (Gogus and Ozturk, 2008) and the tibial plafond (Bui-Mansfield

Table 1
Distribution of OD lesions in the joints/bones of Middenbeemster adult individuals.

Joint	Joint surface	# of bones (left and right) observed	# of OD lesions	Total and%
Shoulder	Glenoid cavity	212	0 definitive (6 possible)	0 (2.8)
	Humeral head	242	0	0
Elbow	Radial Head	214	0	0
	Olecranon Process	229	0	0
	Distal Humerus	242	0	0
Hip	Femoral Head	227	0	0
	Acetabulum	199	0	0
Knee	Distal Femur	249	0	0
	Proximal Tibia	143	0	0
Ankle	Distal Tibia	143	0 definitive (6 possible)	0 (4.2)
	Superior Talus	216	0	0
Foot[a]	Inferior Talus	216	3	1.4
	Navicular (posterior and anterior surfaces)	190	3	1.6
	Navicular posterior inferior (concave) surface	190	3	1.6
	Navicular anterior (convex) surface	190	0	0
	1st Metatarsal (MT1) (proximal and distal ends)	218	8	3.7
	1st Metatarsal proximal (concave) end	218	3	1.4
	1st Metatarsal distal (convex) end	218	5	2.3
	1st Proximal Distal phalanx (proximal and distal ends)	172	10	5.8
	1st Proximal Phalanx proximal (concave) end	172	10	5.8
	1st Proximal Phalanx distal (convex) end	172	0	0
	Foot Overall	**796**	**24**	**3.0**

[a] Only foot elements affected by OD are reported.

et al., 2000) are considered rare sites for OD that are difficult to diagnose. These individuals were, therefore, documented as possible cases for future examination but excluded from the analysis to reduce speculation. The purpose of this conservative approach aims to diagnose only true OD lesions and thus produce reliable data upon which various etiological factors can be evaluated. Lastly, there were neither definitive nor possible cases of OD in the knee. Conclusively, the prevalence of non-pedal OD for Middenbeemster is 0%.

3.2. Distribution of OD in the joints

Table 1 lists the bones and joint surfaces of the adult Middenbeemster skeletal sample that were assessed for OD. The foot is the only part of the body in which definitive OD lesions were observed. Statistical analysis is based only on the skeletal elements of the foot since they exhibit definite OD cases.

3.3. Distribution of OD in different bones of the foot

Seven hundred and ninety six foot bones, both right and left sides, were observed, and 24 had OD, resulting in a 3.0% prevalence of OD in the foot. The talus (Fig. 2), the navicular (Fig. 3), the MT1 (Figs. 4 and 5,), and the 1st proximal foot phalanx (Fig. 6) were the skeletal elements of the foot that were affected by OD. The other foot bones were not affected. These results yield a statistically significant difference in OD prevalence among the foot bones (x^2 = 8.221, p = 0.042). In the Middenbeemster population, the 1st proximal phalanx was involved in the majority cases with eight individuals, regardless of sex, affected. Five individuals bore OD lesions in the MT1, three in the inferior talus, and lastly two in the navicular bone. One individual suffered from OD in multiple joints: in the MT1 and the naviculars. The overall prevalence of OD (definite lesions only) in the adults was 17 out of 132, or 12.9%. This means that 12.9% of the individuals from Middenbeemster had OD in their feet whereas no definite occurrence of the lesion was observed in the non-pedal parts of their body (non-pedal OD prevalence is 0%).

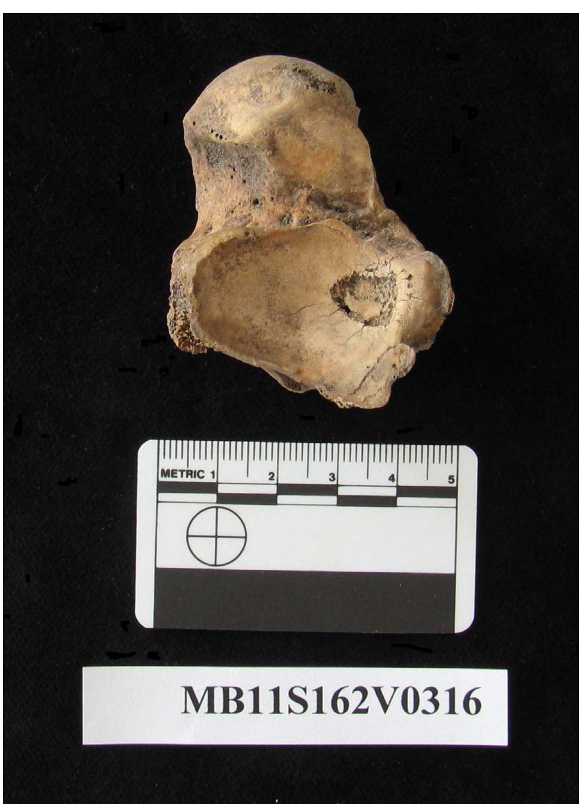

Fig. 2. Individual V0316 exhibiting OD of the posterior subtalar facet of right talus.

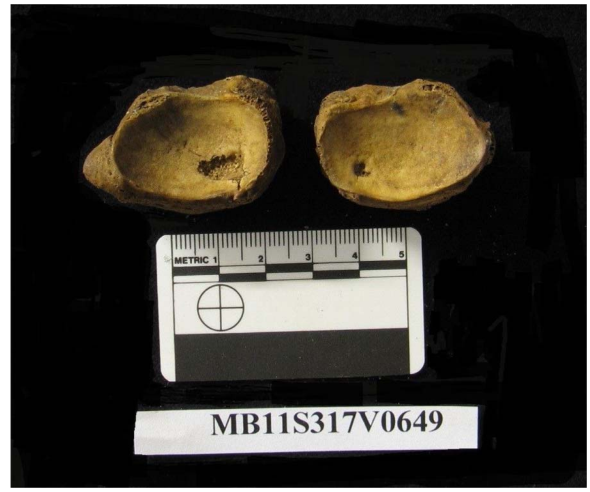

Fig. 3. Individual V0649 exhibiting OD bilaterally on the proximal articular surfaces of naviculars.

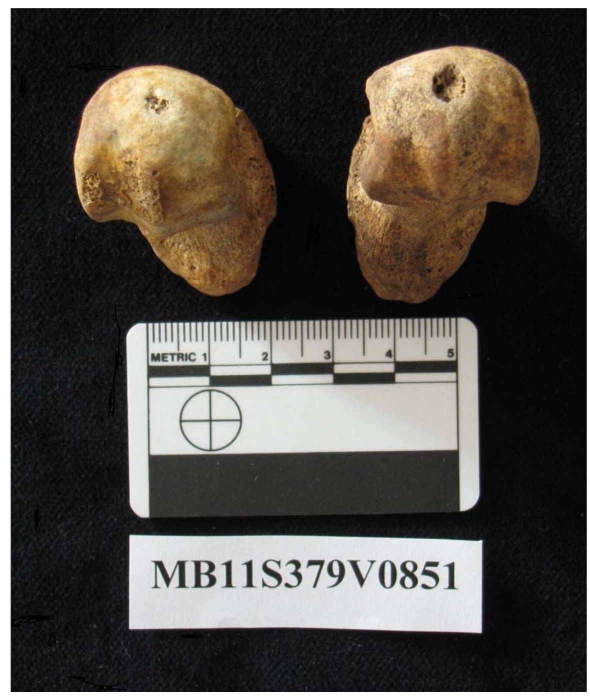

Fig. 4. Individual V0851 exhibiting OD bilaterally on the distal end of the MT1.

3.4. OD distribution in the foot between sexes and age categories

Table 2 illustrates the number of individuals who suffered from OD of the foot according to sex and age categories. The tendency of males to be more affected by OD than females (Aufderheide and Rodríguez-Martín, 1998) is observed in the Middenbeemster sample. From the total number of 132 examined individuals, 11 out of 63 (17.5%) male individuals suffered from OD in one or more skeletal elements of the foot whereas six out of 69 (8.7%) females were affected. However, this difference does not reach statistical significance (x^2 = 2.255; p = 0.133).

From the age groups that exhibit OD of the foot, regardless of sex, the highest frequency, 19.2%, is observed in old adults (50+ years) followed by the middle adults (36–49 years) with 14.3%. The lowest

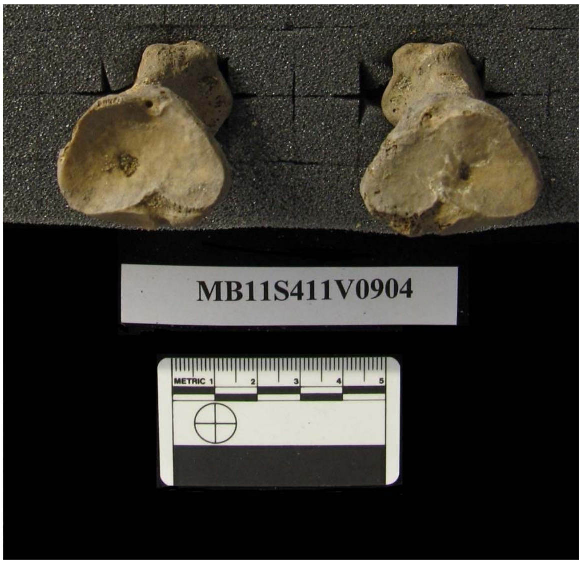

Fig. 5. Individual V0904 exhibiting OD bilaterally on the proximal end of MT1.

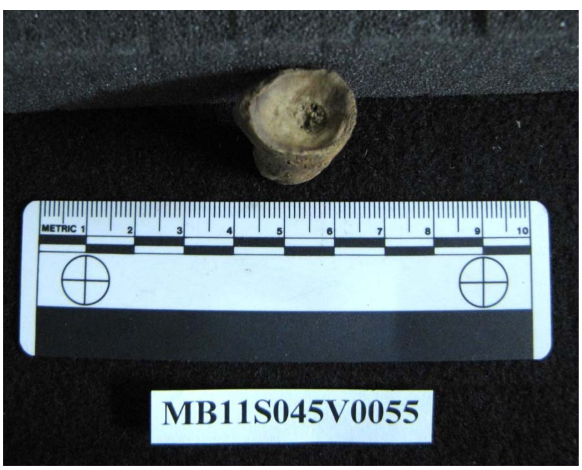

Fig. 6. Individual V0055 exhibiting OD on the base of the left 1st proximal foot phalanx.

frequencies are observed in early young adults (18–25 years) and late young adults (26–35 years) with 9.5% and 6.9% respectively. None of the differences in OD frequency between age groups are statistically significant ($x^2 = 1.164$, $p = 0.762$). Lastly, one subadult (16.5 ± 1 years), had OD on the posterior subtalar articular facet of the left talus.

Table 3 shows the distribution of affected foot bones from the total number of bones examined, left and right sides combined and divided by sex. According to the total number of the four foot bones observed (n = 796), the tendency for skeletal elements in males to be more affected by OD is maintained (OD in skeletal elements of the foot in males = 17/394, 4.3%; OD in skeletal elements of the foot in females = 7/402, 1.7%). This difference is statistically significant ($x^2 = 4.506$; $p = 0.034$). The 1st proximal foot phalanx, the most frequently affected bone, is evenly affected between men and women (5 cases each). The talus exhibited OD in two males and one female. The navicular is affected with OD in one male and in one female. Finally, only males (five cases) had OD in the MT1 bone. The only difference in OD between males and females that is statistically significant is found in the MT1 (Fisher's $x^2 = 8.8775$; $p = 0.003$)

Table 3
Distribution of OD in the different affected bones of the foot (left and right sides combined) between males and females of the Middenbeemster cemetery.

Skeletal elements of the foot	Male	(%)	Female	(%)
1st proximal foot phalanx	5/86	5.8	5/86	5.8
Talus	2/107	1.7	1/109	0.9
MT1	8/106	7.7	0/112	0.0
Navicular	2/95	2.1	1/95	1.0
Σ	**17/394**	**4.3**	**7/402**	**1.7**

Table 4
Distribution of OD in bones of the foot between the left and right side (sexes and ages combined) from the Middenbeemster cemetery.

Skeletal elements of the foot	Left	(%)	Right	(%)
1st proximal foot phalanx	6/88	6.8	4/84	4.8
MT1	3/109	2.8	5/109	4.7
Talus	2/109	1.8	1/107	0.9
Navicular	1/99	1.0	2/91	2.2
Σ	**12/405**	**3.0**	**12/391**	**3.1**

3.5. Distribution of OD between left and right side

Table 4 shows that in the left and right foot, 12 bones from each side had OD. In the 1st proximal phalanx the left side is slightly more affected, while in the MT1 the right side is slightly more affected. However, none of the left-right side differences are statistically significant ($x^2 = 0.009$; $p = 0.926$).

3.6. Bilateral cases and multiple joints involvement

Six individuals were affected bilaterally by OD. One female had OD lesions in the base (proximal end) of both 1st proximal foot phalanges. Five males were affected bilaterally on three different joint surfaces. One bilateral case was found in the 1st proximal foot phalanx. The second case was the bilateral involvement of the navicular. This individual also had OD on the proximal end of the right MT1, therefore the involvement of two joint types. Finally, the third, fourth, and fifth cases of bilateral occurrence involved the MT1 with the heads (distal end) being affected in two cases and the bases (proximal end) in the other.

4. Discussion

4.1. Middenbeemster results in the wider paleopathological context

OD in past populations has been documented in few publications. This could be due to non-retrieval of skeletal elements exhibiting OD

Table 2
Individuals distributing OD in the foot, left and right side combined, according to sex and different age groups (+18) in the Middenbeemster cemetery.

Age group (years)	Males	Percentage (%)	Females	Percentage (%)	Total	Percentage (%)
Early young adults (18–25)	2/10	20.0	0/11	0.0	2/21	9.5
Late young adults (26–35)	0/11	0.0	2/18	11.1	2/29	6.9
Middle adults (36–49)	5/26	19.2	3/30	10.0	8/56	14.3
Old adults (50+)	4/16	25.0	1/10	10.0	5/26	19.2
Σ	**11/63**	**17.5**	**6/69**	**8.7**	**17/132**	**12.9**

lesions and/or bad preservation of the sample caused by taphonomic agents. The latter particularly hinders the diagnosis of OD lesions, and may be a reason why it goes unnoticed and/or unrecorded by osteoarchaeologists. Also, the potential for OD to reveal information about activity practices may not be as well known to osteoarchaeologists as other, more widely published activity-related pathological lesions and shape changes, i.e. OA, fractures, entheseal changes, long bone morphology, contributing to its infrequent reporting.

Research involving large numbers of individuals, notes a low frequency of general OD (i.e. < 1%). For example, Ponce (2010) analyzed a skeletal sample from Chile composed of inland agriculturists and fishermen. The prevalence of OD for that sample was 0.6%, with one individual out of 175 exhibiting bilateral OD of the knee. Likewise, Bourbou's (2010) research on a Byzantine skeletal sample from Crete demonstrated an OD prevalence of 0.3% consisting of only one individual out of 271 with OD in the left glenoid cavity. Furthermore, studies in past populations (During et al., 1994; Loveland et al., 1984; Ortner, 2003; Wells, 1974) usually do not distinguish between pedal and non-pedal OD. Wells' (1974) research in ancient British skeletal material, mentioned that the majority of OD lesions, 95%, occurred in the knee or the foot, without distinguishing the two sites as per OD prevalence.

In the Netherlands, two large skeletal collections from the urban sites of Dordrecht (1275-1572 CE) (Maat et al., 1998) and Alkmaar (1448-1572 CE) (Schats, 2012), have been assessed for OD. The frequency of affected individuals was 3.5% (11/316) and 4.2% (7/165), respectively. Unfortunately, the report from Dordrecht only mentions the number of the affected individuals and not the bones in which OD occurred. Schats' (2012) research on adult skeletons from Alkmaar yielded a general OD prevalence of 4.2% (7/165). Four individuals exhibited pedal OD resulting in a prevalence of 2.4% (4/165). The affected bones were the talus and the MT1 with two individuals involved in each case. All individuals exhibiting pedal OD were males. Remarkably, one of the two individuals with talar OD was affected bilaterally and in both the talar dome and subtalar facets. The second individual suffered unilaterally from OD only in the subtalar facet. The MT1s, of the other two individuals, demonstrated OD lesions in the proximal ends (base) in both cases. It should be mentioned that only 63 out of 165 individuals had both feet present, without however implying that all of the skeletal elements of each foot were retrieved in every case. The three individuals exhibiting non-pedal OD were females and had the acetabulum (hip joint) affected in two cases and the 3rd metacarpal in one case. The large difference in pedal OD frequency between urban Alkmaar (2.4%) and rural Middenbeemster (12.9%), suggests there may be factors associated with the lifeways and activity patterns of Dutch rural populations, prior to industrialization, that contributed to OD development.

4.2. Diet and malnutrition in the Beemster

The diet of an average Dutch person in the 1800s consisted mainly of bread and potatoes (Wintle, 2000). Sometimes a meal would be enriched by vegetables, eggs, along with meat or fish (Wintle, 2000). Adult osteological stature reconstruction and archival stature data from males in the 1800s revealed measurements that were either at or above the mean of other Dutch populations from post-Medieval times (Lemmers et al., 2013). Consequently, this suggests that an adequate diet was the norm. The Beemster inhabitants focused mainly on dairy farming, and as a result their diet may have been further enriched in dairy products, such as milk, cheese and butter. Vitamin D deficiency was present in some young Middenbeemster subadults (i.e. < 5 years of age) (Veselka et al., 2013), but was absent in older individuals. The analyzed Middenbeemster sample consisted of adult individuals who did not exhibit signs of Vitamin D or any other possible dietary deficiency. Consequently, malnutrition was removed as a possible factor that could be associated with OD formation in the Middenbeemster sample.

4.3. OD and age

According to clinical literature, the most common onset age of OD is adolescence (Schenk and Goodnight, 1996; Schindler, 2007). In this case, OD is classified as juvenile OD and its highest peak occurs between 10–13 years of age (Schindler, 2007). These lesions frequently tend to heal (Schenk and Goodnight, 1996; Schindler, 2007). The lesions that do not heal are due to continued activity and subsequently often lead to the seeking of medical attention (Schenk and Goodnight, 1996). It is speculated, in both juvenile and adult OD, that the subchondral layer of bone fails to support the articular cartilage, which leads to the softening of the latter (Schenk and Goodnight, 1996). In Middenbeemster, OD occurred in an adolescent (one case on the posterior subtalar articular facet of the left talus). While there is no statistically significant difference among the age groups, the highest number of OD lesions occurs in the old adults, which is consistent with OD continuing to form throughout life. The fact that the highest frequency is observed in older individuals should not be confused with the onset age of OD (Schenk and Goodnight, 1996). In the current sample, there is no way of knowing when the affected individuals first developed OD. Therefore, it can be assumed that individuals were affected at a young age and lived long enough, resulting in an accumulation of the observed lesions in the older age categories and particularly in the old adults (50+ years).

As noted, OD is often asymptomatic, meaning individuals would not lessen their activity in order to promote healing. When symptoms do arise from OD in the ankle and foot particularly, they are likened to a sprain with variable levels of pain, discomfort, swelling, and movement limitation (Beil et al., 2012; Bui-Mansfield et al., 2000; Ozturk et al., 2008; Schenk and Goodnight, 1996). However, considering the often demanding conditions of life during the nineteenth century in rural communities, it can be suggested that people did not take much time to rest after seemingly minor injuries, a factor that could contribute to a lesion not healing.

4.4. Biomechanical factors contributing to pedal OD development

Some biomechanical studies (Al-Ali et al., 2002; Athanasiou et al., 1998; Athanasiou et al., 1995; Bruns and Rosenbach, 1990) describe the mechanical properties of foot joint cartilage. These studies explain why certain bones or regions are more prone to developing OD. These results suggest cartilage thickness and degeneration are important factors in OD occurrence in the foot (Al-Ali et al., 2002; Athanasiou et al., 1998; Athanasiou et al., 1995; Bruns and Rosenbach, 1990). While this idea cannot be tested with the current sample, we can see if the pattern of OD in Middenbeemster can be better understood in light of information about cartilage thickness.

The pedal OD observed in Middenbeemster, as discussed in 4.3, exhibits an interesting pattern. The talus is the third most frequently affected bone. All three OD lesions occur in the subtalar facets, not the talar dome. Part of the explanation for the occurrence of subtalar OD could be that the talocalcaneal average cartilage thickness (0.72 ± 0.11 mm mean thickness) is smaller than the talar-tibial thickness (0.86 ± 0.17 mm mean thickness) (Al-Ali et al., 2002) and therefore more vulnerable to degeneration. Subtalar OD is rarely mentioned in modern clinical literature. Two case reports mention a 10-year-old boy (Vialle et al., 2007) and 14-year-old girl (Kadakia and Sarkar, 2007) both suffering from subtalar OD without any history of trauma. Both reports agree that diagnosis of subtalar lesions are difficult and require additional imaging, rather than a plain radiograph, to be accurately identified. There is no consensus on the etiology of subtalar osteochondritic lesions. Kadakia and Sarkar (2007) proposed traumatic dislocation of the subtalar joint as a probable cause, whereas Vialle et al. (2007) suggested the theory of repetitive microtrauma occurring within the joint to be more appropriate, as it leads to devascularization of the region thus causing the lesion. Finally, Choi and Ogilvie-Harris (2002) reported a series of 10 patients with subtalar OD due to acute trauma.

The 1st proximal phalanx demonstrates the most OD lesions followed by the MT1. The base of the 1st proximal phalanx was involved in all OD cases, whereas the MT1 exhibited five cases of OD in the head, with two of them being bilateral, and three in the base, two of which were bilateral. It should be noted that none of the OD lesions found in the head of the MT1s corresponded to the lesions in the base of the 1st proximal phalanx. Consequently, it appears that the MT1 joint, with either the MT1 or the 1st proximal phalanx being involved, is the site mostly affected in the Middenbeemster sample. Athanasiou et al. (1998) find that OD and hallux rigidus are the two most frequent articular cartilage disease processes affecting the MT1 joint due to limitation of its natural locomotion. Degenerative changes would be enhanced in an area that undergoes more mechanical stress. However, cartilage degeneration could occur even without traumatic incidents (Athanasiou et al., 1998). The mean cartilage thickness of the MT1 joint was calculated to be 0.75 ± 0.21 mm (Athanasiou et al., 1998). This value is close to that of the talocalcaneal joint (0.72 ± 0.11 mm) but larger than that of the talus-navicular joint (0.57 ± 0.08) (Al-Ali et al., 2002). However, considering the function of the MT1 joint as a lever through which bodyweight is transferred during forward motion (Bojsen-Møller and Lamoreux, 1979), it is prone to cartilage degeneration (Camasta et al., 1993) and subsequently to hallux limitus, which regularly leads to OD (Camasta et al., 1993).

OD of the tarsal navicular is rarely described in modern clinical literature numbering few case reports of one up to four individuals (Beil et al., 2012; Bui-Mansfield et al., 2000; Ozturk et al., 2008). Its etiology still remains uncertain (Beil et al., 2012; Bui-Mansfield et al., 2000; Ozturk et al., 2008) and is probably multifactorial (Beil et al., 2012). These reports describe the diagnosis and treatment of OD in the tarsal navicular bone in four women and two men with ages ranging from 19 to 42 years old. Two of them were professional athletes, one individual experienced pain after hiking for 21 miles and one patient suffered an ankle sprain which led to progressive pain and swelling. The latter used the stairs regularly by working as an assistant at a shop. She also danced ballet until her adolescence. The last two patients did not recall any ankle injury incident. Given the patients' background, these reports suggest repetitive trauma and mechanical stress as the main agents of OD in the navicular. In Middenbeemster, two individuals, a male and female, suffered with OD in the navicular. In fact, the male skeleton demonstrated OD lesions bilaterally. Naturally, it was impossible to investigate whether these individuals suffered any acute or repetitive (micro)trauma. However, given the historical background of the Middenbeemster rural community, it is postulated that strenuous activity imposed both mechanical stress and higher chances of trauma incidents on the pedal bones. In Wells' (1974) study of ancient British people, who were mainly agriculturists, the navicular was also affected in many cases. Wells (1974) suggested that this was caused by the change of biomechanics in the foot due to being used as a tool while cultivating land and poor footwear usage.

Given that the medical history of the Middenbeemster individuals can never be known, nor the full range of activities and events in which they engaged, it is impossible to reach a definitive conclusion regarding the etiology(ies) of the pedal OD. However, combining clinical literature and knowledge about Middenbeemster lifeways, a multifactorial etiology is most appropriate. Acute and/or repetitive microtrauma appear as the most probable factors causing OD, as brought about by a physically active and often laborious way of life. A possible additional factor is discussed below.

4.5. Occupation and physical activity in 19th century Netherlands

Life in Dutch rural communities of the 19th century, such as the Middenbeemster, was quite laborious. People engaged in physically demanding outdoor occupations often working in harsh weather conditions (Lindeboom et al., 2012). Rural women would regularly work in the fields, helping their husbands and leaving their children to be looked after by grandmothers (Lindeboom et al., 2012). Children over four years of age were expected to work with the livestock and in the fields with their parents (Gerard et al., 2002). An example of a daily practice that involved weight bearing in Middenbeemster and other Medieval and post-Medieval societies is the use of a wooden yoke to carry milk or water. Middenbeemster cemetery archives show the following list of occupations of the males who died between CE 1830–1835: cattle farmer, farm worker, gardener, garden aide, water miller, inn-keeper, tavern-keeper, saddle maker, cargo driver, baker, carpenter, tailor, tailor's assistant, preacher, artist, town council mailman, and policeman. For the females who died in this period, an occupation is often not listed but when information was specified, it was of the following: housewife, handmaiden, housekeeper, and servant girl. The most frequently listed occupations are related to dairy farming. It is possible that the specific activities associated with dairy farming would result in high levels of physical stress, acute trauma, and/or repetitive microtrauma, particularly in the feet. These factors' impact could be exacerbated by walking long distances on muddy, slippery and unpaved terrain in the countryside, an activity less common in urban centers where travelled distances were usually shorter and some thoroughfares were compacted by frequent traversing or paved with wood or stone. Daily activities may be the key factor leading to higher pedal OD frequencies in rural Middenbeemster compared to the urban sites of Alkmaar and Dordrecht.

The finding of high pedal OD in Middenbeemster is similar to that of Wells (1974) for the ancient British skeletal samples. Wells (1974) found that when the talus was affected, 75% of the cases were on the subtalar surface and not the dome, contrary to what clinical literature finds as the common site of occurrence (Bui-Mansfield et al., 2000; Kadakia and Sarkar, 2007; Santrock et al., 2003; Schuh et al., 2009; Steinhagen et al., 2001). The navicular, 1st cuneiform, MT1, and lastly, 1st proximal phalanx were the other common sites of OD occurrence (Wells 1974). Wells (1974) suggests that the lesions follow this pattern due to the fact that these ancient populations were predominately agriculturists and thus they were using their feet for daily heavy labour. He further continues that by doing so the 1st metatarsophalangeal joint is the first to suffer from impact, whose force will subsequently transfer along to the 1st cuneiform, the navicular, the head of the talus, and lastly the tibial plafond. However, Wells' report (1974) fails to mention the number of skeletons the sample is comprised of, as well as the number of individuals affected by either pedal or non-pedal OD. Thus, the prevalence of pedal OD is unknown. Nevertheless, the sites of OD lesions, nearly identical to the ones from Middenbeemster, may be indicative of a correlation between a laborious daily life and OD, in past populations.

The results of the statistical analysis of the Middenbeemster sample indicate that there is a significant difference between males and females in the number of bones affected by OD ($x^2 = 4.506$; $p = 0.034$) and particularly in the MT1 (Fisher's $x^2 = 8.8775$; $p = 0.003$). The reason for that could be that the males of Middenbeemster appeared to engage more than women in outdoor strenuous activities, thus having higher chances of suffering from acute trauma and/or repetitive microtrauma.

4.6. Footwear contribution to development of OD

Clinical research shows that footwear with inflexible and thick soles can limit the natural locomotion of the foot (Bojsen-Møller and Lamoreux, 1979). Specifically, the harder the sole, the more constraint there is on the torsional (eversion/inversion) and adduction range of motion of the foot (Lieberman et al., 2010; Morio et al., 2009). Furthermore, shoes can affect the level of mechanical stress and strain our feet are subjected to and chronic use of certain types of shoes, such as those of narrow width and pointed tips in which the foot is constricted, can result or enhance the development of foot abnormalities such as hallux valgus (Trujillo-Mederos et al., 2014). Additionally, Menz and Morris (2005) correlated narrow shoes to hallux valgus, foot pain and corn development on the toes and women who wore more than 25 mm elevated heel shoes developed hallux valgus and plantar calluses. Sim-Fook and Hodgson (1958) observed that an unshod Chinese population demonstrated less foot deformities compared to the Chinese shod population. They concluded that stockings and footwear alter the foot's natural form which results in a decrease of flexibility and mobility. Lastly, it is worth pointing out that even shoes specifically designed with flexible soles for better shock attenuation, were not effective in preventing oversuse injuries of the foot such as stress fractures, arch pain, heel pain, and metatarsalgia (Milgrom et al., 1992).

This research suggests that wooden clogs, the everyday footwear of rural Dutch populations, known as *klompen* in Dutch, may have contributed in the development of OD in the Middenbeemster people. The clog is an example of limitation to the natural pedal motion and imposes movement constraints that require the walking motion to be carried out via a kind of kicking motion. The entire lower leg is kicked forward in order to generate forward momentum as it cannot be generated by the clog bound foot. This alteration may be a factor in the formation of OD.

Clogs come in various forms and shapes. The main design was made entirely out of wood (Fig. 7). Sometimes they are stuffed with straw or cotton and worn with wool socks, or made up of an upper layer or hood of leather (called *kap* in Dutch), but in every case the sole was still made of wood (De Boer-Olij, 2002), thus providing a rather hard and inflexible surface for the foot. *Klompen* in the Netherlands have been worn since at least as early as the beginning of the 13th century, as the oldest wooden shoe ever retrieved and kept in Amsterdam, dates back to 1213 CE. De Boer-Olij (2002) provides numerous examples of wooden shoes that accompanied every event of daily life, in the Netherlands as well as in other regions of northern Europe. Wooden shoes were not only worn by 'common' people but also by nobility, with their status being indicated from the beautiful carvings and decorations on the front of the shoe (De Boer-Olij, 2002). Apart, from the nicely decorated *klompen*, there were other kinds of special wooden shoes, such as the rider's wooden shoe, a type attached to stirrups and made especially for use while riding during winter to protect the foot from cold. Another example is dikers, who wore special square-nosed wooden shoes that facilitated working on their knees when they put basalt blocks onto the sea dikes (De Boer-Olij, 2002).

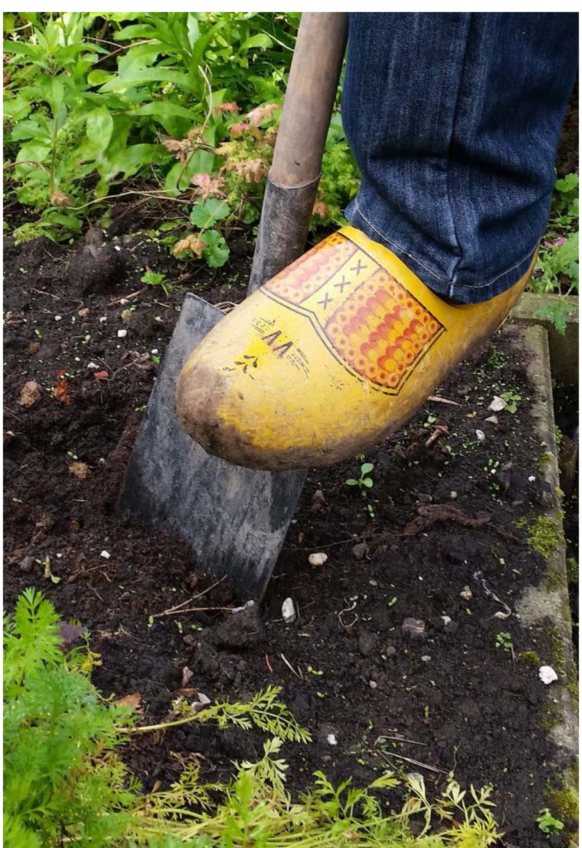

Fig. 7. Allotment picture of amateur greenkeeper from Leiden University using wooden clogs.

In the Netherlands during the time-period of CE 1815–1920 men and women, regardless of class, used to wear *klompen* on a near daily basis, reserving their 'buckle shoes', if they had any, for festivities or Sundays. This practice becomes more common after the 1840s and does not really change until the 1920s (De Leeuw, 1992). Thus, the time period of the majority of skeletons from Middenbeemster fits well into a period of very regular clog use. Clogs were not usually worn in the house. In a typical farmstead, there was a specific area inside the stable next to the entrance of the house for the storage of clogs, and inside the house, slippers were usually worn. Even though wooden clogs were usually stuffed with some sort of malleable lining or worn with thick socks, their hard and inflexible surface can contribute to soft tissue conditions such as achillodynia and heel pad shock absorbency loss (Jørgensen, 1985). It might be possible that regular clog usage, especially in circumstances where the feet are prone to repetitive trauma due to outdoor activities, leads to pathological conditions in the foot bones. The fact that both men and women wore wooden shoes, and that in this rural community also women regularly participated in outdoor dairy farming activities (Lindeboom et al., 2012), may explain why OD lesions were more common in the Middenbeemster sample compared to the ones from the urban sites of Alkmaar and Dordrecht.

Feet undergo a significant level of mechanical stress on a daily basis as they carry the weight of the entire body. The type of footwear one wears can either limit or enhance this amount of stress (Jørgensen, 1985; Trujillo-Mederos et al., 2014; Wells, 1974). Therefore, it is proposed that the comparatively high prevalence of OD in the feet of Middenbeemster peoples is due to a high level of strenuous activity combined with the use of hard and inflexible footwear.

5. Conclusions

The predominately 19th century Middenbeemster skeletal sample exhibited definitive OD lesions only in the foot bones. The knee or other non-pedal sites did not demonstrate lesions that could be assessed as definitive OD. This yielded a non-pedal OD prevalence of 0%, whereas the prevalence of pedal OD was 12.9%. Etiological factors for these results may be attributed to acute trauma and/or repetitive micro-trauma to the foot, a conclusion derived from the rural and outdoor nature of the population's occupation related mostly to dairy farming. Dutch skeletal samples from the urban sites of Alkmaar (1448–1572 CE) and Dordrecht (1275–1572 CE), demonstrate overall OD frequencies of 4.8% (Schats, 2012) and 3.5% (Maat et al., 1998), respectively. Alkmaar exhibits a pedal OD frequency of 2.9% (this value is not known for Dordrecht), much lower than that of Middenbeemster's 12.9%. A possible explanation for this may lie in urban Alkmaar citizens engaging in less frequent strenuous activity and thus experiencing less trauma in pedal and non-pedal sites.

Since agricultural industrialization did not come to the Netherlands until the late 1800′s (Drukker and Tassenaar, 1997; Van Zander, 1996), the Middenbeemster dairy farmers were using traditional tools, methods, and footwear such as clogs. Considering clinical literature and historical data about the occupations of the Middenbeemster community, it is suggested that a laborious way of life may have contributed to OD development. The fact that definitive OD was only observed on skeletal elements of the foot indicates that a common agent might be responsible. Thus, the hypothesis of wooden shoe usage is suggested. Hard and inflexible shoe surfaces limit natural movement and cause mechanical stress and strain, which are both considered possible causes in OD development. Further research should be conducted between populations originating from Northern and Southern Europe to investigate the impact of different footwear, in conjunction with the level of activity, on the skeletal elements of the foot. This research provides a poignant example of the effect of cultural practices on the skeleton.

Acknowledgements

We would like to thank members of the Historisch Genootschap Beemster (HGB) for their help with the cleaning of skeletons and archival research. Also, special thanks to Dr. Rachel Schats, from Leiden University, Faculty of Archaeology, Human Osteoarchaeology Laboratory, for providing useful data from the Alkmaar report and to Laurens van Maren for providing a clear map of the Netherlands indicating the location of Middenbeemster.

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