



Multiple osteochondromas in the archaeological record: a global review

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ABSTRACT

Multiple osteochondromas is an inherited autosomal dominant condition of enchondral bone growth. The paper undertakes the first synthesis study of the 16 known cases of the condition that have been identified in the international palaeopathological record. It also includes information derived from two newly discovered cases of the disease in two adult male individuals recovered from the Medieval cemetery at Ballyhanna, Co. Donegal, Ireland. The formation of multiple osteochondromas is the best known characteristic of the disease but it also involves the development of a suite of orthopaedic deformities. These deformities, which include disproportionate short stature, inequality of bone length, forearm deformities, tibiofibular diastasis, coxa valga of the hip and valgus deformity of the knee and ankle, are discussed in relation to the archaeological cases. Numerous synonyms for the disease have been used within the various publications produced by palaeopathologists, and this can generate confusion among readers. As such, the paper recommends that in future palaeopathologists should follow the guidance of the World Health Organization and use the term multiple osteochondromas when discussing the disease.

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1. Introduction

To date, all publications pertaining to palaeopathological examples of multiple osteochondromas (MO) have taken the form of case study papers. As such, the following paper represents the first attempt to undertake a review of all previous cases of MO identified in the palaeopathological record to see how the findings correlate with modern clinical knowledge of the condition. The disorder was first identified by the surgeon John Hunter in 1786 when he described a patient with multiple exostoses (Stieber et al., 2001). A confusing array of terms are now associated with MO and Stieber and Dormans (2005) have listed some 13 synonyms that have been used to refer to the disorder. In addition to MO, the following terms have been encountered in the palaeopathological literature – diaphyseal aclasia (Buckley et al., 1990; Ortner, 2003; White, 1988); diaphyseal aclisis (Aufderheide and Rodríguez-Martín, 1998; Lyall and Mann, 1993; Singer, 1962); multiple osseous exostoses (Gladykowska-Rzeczycka and Urbanowicz, 1970); exostosis multiplex (Sjøvold et al., 1974) and multiple cartilaginous exostoses (Coughlan, 2007). In 2002 the World Health Organization addressed the problem of the variation in general terminology applied to the condition, as well as the differences in

meaning attached to the term exostosis, and suggested that the preferred term for the disorder should be multiple osteochondromas (Bové and Hogendoorn, 2002) and this is the term that will be used throughout the current paper.

Multiple osteochondromas is an inherited autosomal dominant condition of enchondral bone growth which primarily involves the development of multiple osteochondromas – or exostoses capped with cartilage – at the metaphyseal regions of the long bones (Cook et al., 1993; Stieber and Dormans, 2005). Visible or palpable osteochondromas can be present at birth although they are more commonly identified during the first decade of life (Stieber et al., 2001). The osteochondromas have two main forms – sessile and pedunculated. Sessile osteochondromas are broad, bulbous lesions, whereas pedunculated osteochondromas are spur-like in appearance (Stieber and Dormans, 2005). While a person is still growing the osteochondromas continue to develop, resulting in thickening of the metaphyseal area and slowing of bone growth which contributes to the development of a range of other deformities, including disproportionate short stature; limb-length discrepancies; forearm deformities; coxa vara of the femora; valgus deformities of the knees and ankles; asymmetry of the pectoral and pelvic girdles; shortening of the foot and hand bones and scoliosis (Black et al., 1993; Cook et al., 1993; Shapiro et al., 1979; Stieber et al., 2001; Stieber and Dormans, 2005). Around 75% of affected individuals display an osseous deformity and these most frequently involve the forearm (50%), the ankle (45%) or the knee (20%), while some 40% have an abnormally short stature (Lichtenstein, 1973).

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Despite their benign nature osteochondromas are known to cause a variety of clinical and cosmetic problems. Pain can result from repeated soft tissue trauma or bursa formation over a large osteochondroma, while pedunculated osteochondromas can be susceptible to fracture and muscles overlying osteochondromas can be injured during certain movements. When the proximal femur or the forearm is extensively affected by osteochondromas a restricted range of motion (ROM) can occur. Other common side-effects include impingement, entrapment or damage to blood vessels, nerves and tendons. Less common complications as a result of osteochondroma formation include spinal cord compression, urinary and intestinal obstruction, problems with swallowing and bleeding within the chest cavity (Stieber and Dormans, 2005).

Recent genetic research has revealed that the two genes most strongly associated with MO are EXT1 on 8q24.1 and EXT2 on 11p13. It is thought that mutations in these genes result in the impairment of cell surface heparan sulfate proteoglycans synthesis, which in turn causes a reduction in the signaling of fibroblast growth factor and the proliferation of abnormal chondrocytes at the locations where the osteochondromas develop (Zak et al., 2002). It has proven difficult to ascertain an accurate prevalence rate of MO since individuals with a mild version of the condition may remain undiagnosed. The condition has been most thoroughly studied among white populations where its prevalence ranges from approximately 0.9 to 2 individuals per 100,000 (Stieber and Dormans, 2005). Strikingly higher prevalences, however, have been identified for more isolated populations such as the Chamorros of Guam (100 per 100,000 individuals) (Krooth et al., 1961) or the northern Ojibway community of Pauingassi, Manitoba, Canada (1310 per 100,000 individuals) (Black et al., 1993; Dooley et al., 1988).

2. Material

The research is based on data obtained from 14 published case studies of the condition in addition to the findings derived from two new examples of MO identified in individuals recovered from the Medieval cemetery at Ballyhanna, Co. Donegal, Ireland. A detailed overview of the specific lesions apparent in the two Ballyhanna individuals is provided as [Supplementary material](#). A summary of the demographic information obtained for all 16 individuals is included in [Table 1](#).

3. Results and discussion

3.1. Demographic characteristics

The affected individuals range in date from the Middle Bronze Age (c. 1700 BC) for two cases from Jericho, Jordan (Lyll and Mann, 1993), through to the Post-Medieval period for the example from Que Que in Zimbabwe (Singer, 1962). It is interesting to note that the two early Jericho cases are the only prehistoric examples of the condition reported thus far, with all remaining 14 examples dating to historic times.

The cases have derived from a total of eight countries, with multiple examples having been reported from Gotland ($n=2$), England ($n=3$) Jordan ($n=3$) and Ireland ($n=4$). The majority of affected individuals ($n=9$) occurred as isolated cases within a burial ground but three sites have produced multiple cases of the condition. The two individuals with the condition recovered from the burial ground at St Clemens, Visby, on the island of Gotland, were clearly related – one being the mother, while the other was her unborn full-term infant. It was suggested that the presence of a large osteochondroma on the anterior aspect of the ilium may have made it impossible for the fetus to rotate during birth or that

the reduced size of the pelvic canal was incompatible with a normal delivery thereby resulting in the death of both the mother and child (Sjøvold et al., 1974). The two affected skeletons recovered from Middle Bronze Age contexts at Jericho, Jordan, were broadly contemporary, although they were recovered from two separate tombs – B50 and G73. A third individual from Jericho was notably more recent, dating to the 1st century AD (Lyll and Mann, 1993). Radiocarbon dating indicated that the two individuals recovered from the Medieval cemetery at Ballyhanna, Co. Donegal, Ireland, were separated in time by several hundred years. The occurrence of multiple, but temporally distinct, individuals from the same burial grounds may indicate that the genes responsible for the condition had existed in these groups over substantial periods of time. The recovery of four individuals from three separate sites in Ireland and three individuals from three separate sites in England is also of interest. It is possible that the island environments of Britain and Ireland are of significance here since they would have meant that there was relatively restricted movement for the majority of people. As discussed above higher prevalences of the condition have been found to occur among isolated modern populations, such as the Chamorros of Guam (Krooth et al., 1961).

It was possible to ascertain the sex of 12 of the individuals afflicted with MO. Six of the affected individuals were female (50%), while six of the skeletons were those of males (50%). Early clinical research was of the opinion that the disease was more common in males (e.g. Jaffe, 1943; Stocks and Barrington, 1925) but in their study of the northern Ojibway community of Manitoba, Canada, Black et al. (1993) found that females (60%) were affected to a greater extent than males (40%), while Solomon's (1964) study of patients from the Hospital for Sick Children, Great Ormond Street, and the Royal National Orthopaedic Hospital in London found that males (54%) and females (46%) were affected in near equal proportions. It is now generally accepted, on the basis of recent studies of nuclear families, that there is no evidence of predominance in either sex (Stieber and Dormans, 2005). The palaeopathological evidence would tend to support this assertion.

Only two of the 16 individuals with MO were young juveniles. As discussed above, the full-term fetus recovered from Gotland may have died as a consequence of obstructed labor resulting from MO in his/her mother (Sjøvold et al., 1974). The condition was also apparent in a four year old child recovered from Winchester, Hampshire. Apart from the presence of multiple osteochondromas there was nothing untoward apparent in the skeletal remains and the author was of the opinion that the condition was unlikely to have been the cause of death (Ortner, 2003). A further two of the individuals were classified as adolescents. The male skeleton recovered from Pruszcz Gdanski, Poland, had an age at death of 15–18 years (Gładkowska-Rzeczycka and Urbanowicz, 1970), while the Middle Bronze Age male skeleton recovered from Tomb G73 (Skeleton A) at Jericho, Jordan, was described as having been in late adolescence (Lyll and Mann, 1993). Following the adult age at death categorization system of Buikstra and Ubelaker (1994) the adult individuals were found to have included seven young and two young-middle adults. Due to the incomplete nature of the skeletal remains it was not possible to ascertain precise age at death details for the individuals from Uxbridge Ossuary, southern Ontario, Canada (Katzenberg et al., 1982); Chicama, Peru (Ortner, 2003) and Poundbury, Dorset, England (Farwell, 1993), although all three individuals appear to have been adults. The overall age at death profile for the corpus of 13 individuals with precise age at death assignments would tend to suggest that individuals with the condition were more susceptible to dying young – some 11 of these 13 people had died as juveniles or young adults, with only two having possibly survived to middle adulthood (c. 35–49 years).

Table 1

A summary of the archeological and paleodemographic characteristics of the 16 cases of multiple osteochondromas.

Site	Country	Context	Date	Age (yrs)	Sex	Authors
Que, Que	Zimbabwe	–	Post-European invasion period (c. post 1800 AD)	25–35	F	Singer, 1962
Pruszcz Gdanski	Poland	Burial no. 204/68	2nd century AD	15–18	M	Gładkowska-Rzeczycka and Urbanowicz, 1970
St Clemens, Visby	Gotland	E corner of N aisle	1250–1525 AD	17–20	F	Sjøvold et al., 1974
St Clemens, Visby				Full-term fetus	–	Sjøvold et al., 1974
Uxbridge Ossuary, Southern Ontario	Canada	Bundle burial	1490 +/- 80 AD	Adult	?	Katzenberg et al., 1982
St Nicholas Shambles, London	England	Sk 5220	11–12th century AD	c. 30	M	White, 1988, 37–38
Poundbury, Dorset	England	Burial 1353	3–4th century AD	Adult	F	Molleson, 1993, 188
Winchester, Hampshire	England	BMNH G932, CG69, TR XL 1085	Anglo-Saxon	4	–	Ortner, 2003, 518–519
Chicama	Peru	NMNH, uncatalogued	?	Adult	?	Ortner, 2003, 518–519
Jericho	Jordan	Tomb B50, Sk H	1700 BC	Young adult	F	Lyall and Mann, 1993
Jericho		Tomb G73, Sk A	1740/1690 BC	Late adolescent	M	Lyall and Mann, 1993
Jericho		Trench 2, Sk 3	1st century AD	20–25	F	Lyall and Mann, 1993
Stephen Street, Dublin	Republic of Ireland	Burial XII and Disarticulated E	Medieval	Young-middle adult	F	Coughlan, 2007
Kilshane, Co. Dublin	Republic of Ireland	B93	Later Early Christian	25–35	M	Buckley et al., 1990; Buckley, 1991
Ballyhanna, Co. Donegal	Republic of Ireland	Sk 197	Medieval	Young-middle adult (c. 30–40)	M	–
Ballyhanna, Co. Donegal		Sk 331		Young adult (c. 18–25)	M	–

A probable cause of death was only apparent in two of the adults. Given the full-term nature of the associated fetus it is feasible that the young adult female from St Clemens, Visby, Gotland, may have died as a result of obstructed labour as a consequence of osteochondroma formation in the bones of the pelvis (Sjøvold et al., 1974). Coughlan (2007) was of the opinion that an osteochondroma on the right distal femur of the young-middle adult female from Stephen Street, Dublin, Ireland, had transformed into a malignant chondrosarcoma. A similar lesion may also have been present on the left distal femur but this was obscured as a consequence of post-mortem damage.

3.2. Palaeopathological characteristics

A limitation of the study is the difficulty of comparing the nature of the palaeopathological lesions apparent in the affected individuals. This has largely occurred as a result of the incomplete nature of a number of the skeletons or as a result of the manner in which the lesions were reported. A summary of the palaeopathological characteristics of each affected individual is presented in Table 2. It is recommended that future palaeopathological studies of MO take into consideration each of the features listed in Table 2 to enable a clearer understanding of the palaeopathology of the disease to be gained. A further limitation of the study is that the small number of individuals ($n = 16$) means that the percentage values can only be taken as approximate indicators of prevalence and may not be statistically significant. Nevertheless, despite these limitations a number of trends are apparent. While osteochondromas usually manifest during the first decade of life (Bové, 2008) many of the associated orthopaedic deformities caused by the condition are unlikely to have become apparent until older childhood and adolescence. As such, in the following discussion the lesions apparent in the two young children – the fetus recovered from St Clemens, Visby (Sjøvold et al., 1974) and the four year old child from Winchester, Hampshire – have largely been excluded from the discussion.

3.2.1. Nature of the osteochondromas

It was possible to ascertain the precise nature of the osteochondromas in 13 of the 16 cases. In the notable majority of cases (92.3%; $n = 12/13$) both sessile and pedunculated osteochondromas

were present. The only exception to this situation was Skeleton A from Tomb G73 at Jericho. This individual appeared to have been suffering from a very mild form of the condition since, despite the relatively complete nature of the skeleton, the osteochondromas appear to have been localised to the right distal humerus and proximal ulna (Lyall and Mann, 1993). A diagnosis of MO can be made on the basis of the presence of at least two osteochondromas of the metaphyseal regions of the long bones. Modern clinical studies have indicated that the size and number of lesions can vary significantly between affected individuals (Bové and Hogendoorn, 2002).

3.2.2. Distribution of the osteochondromas

Modern clinical research has indicated that the majority of osteochondromas develop at the extremities of the long bones, with the bones of the knee being most frequently affected (Bové and Hogendoorn, 2002). The lesions have also been identified on the scapulae, ribs and ilia as well as on the bones of the hands and the feet. They rarely affected the bones of the skull (Stieber and Dormans, 2005). A summary of the bones affected in the 16 archaeological cases is provided in Fig. 1.

No osteochondromas were recorded for either the cranium ($n = 0/6$) or mandible ($n = 0/7$). Despite their relatively infrequent preservation the bones of the pectoral girdle frequently displayed lesions, with frequencies of 85.7% ($n = 6/7$) for the scapulae, 100% ($n = 8/8$) for the sternal end of the clavicles and 75% ($n = 6/8$) for the acromial end of the clavicles. Similarly, the lesions occurred quite frequently on the ilia (64.7%; $n = 11/17$). Osteochondromas occurred relatively infrequently among individuals with preserved ribs (50%; $n = 3/6$) and vertebrae (33.3%; $n = 2/6$). Only a small number of individuals had observable hand bones present but all of those that did displayed the lesions (100%; $n = 4/4$), while lesions were evident in 57.1% ($n = 4/7$) of individuals with feet bones.

The elbow was the region of the appendicular skeleton least affected, with osteochondromas apparent in 16.7% of distal humeri ($n = 2/12$), 30% ($n = 3/10$) of proximal radii and 30.8% ($n = 4/13$) of proximal ulnae. The proximal femora at the hip displayed lesions with a frequency of 72.2% ($n = 13/18$), while the bones of the ankle displayed frequencies of 68.4% ($n = 13/19$) for the distal fibulae and 76.2% ($n = 16/21$) for the distal tibiae. The proximal humeri of the shoulders displayed osteochondromas with a frequency of 80%

Table 2
A summary of the palaeopathological characteristics of the 16 cases of Multiple Osteochondromas. + = affected; o = unaffected; – = unobservable/information not provided; +* = osteochondromas of uncertain form; Poss = Information not stated in reports but photographs suggest possible presence of lesion.

Site	Context	Sessile osteochondromas	Pedunculated osteochondromas	Short stature	Inequality in bone length
Que Que	–	+	+	+ (146 cm)	–
Pruszcz Gdanski	Burial no. 204/68	+	+	–	R radius and ulna
St. Clemens, Visby	E corner of N aisle	+	+	+ (145 cm)	R humerus, L radius and ulna
St. Clemens, Visby	E corner of N aisle	+*	+*	–	–
Uxbridge Ossuary, southern Ontario	Bundle burial	+	+	–	R femur
St. Nicholas Shambles, London	Sk 5220	+	+	+ (not provided)	–
Poundbury, Dorset	Burial 1353	+	+	–	–
Winchester, Hampshire	BMNH G932, CG69, TR XL 1085	+*	+*	–	–
Chicama	NMNH, uncatalogued	+	+	–	–
Jericho	Tomb B50, Sk H	+	+	–	o
Jericho	Tomb G73, Sk A	+	o	–	o
Jericho	Trench 2, Sk 3	+*	+*	–	o
Stephen Street, Dublin	Burial XII and Disarticulated E	+	+	–	–
Kilshane, Co. Dublin	B93	+	+	o (161 cm)	–
Ballyhanna, Co. Donegal	Sk 197	+	+	o (166.8 cm)	L radius
Ballyhanna, Co. Donegal	Sk 331	+	+	+ (158.3 cm)	L clavicle, L humerus, L ulna and radius, L fibula

($n = 12/15$; Fig. 2). As such, the knee joint was most frequently affected and the lesions were evident in 81.3% ($n = 13/16$) of proximal fibulae, 82.6% ($n = 19/23$) of distal femora and 90.9% ($n = 20/22$) of proximal tibiae. This trend largely mirrors the clinical findings of Shapiro et al. (1979) in which the proximal humeri (98%) and the bones of the knee (distal femora = 98%; proximal tibiae = 98%; proximal fibulae = 97%) were particularly frequently affected. In their study osteochondromas were also frequently apparent on the proximal femora (90%) and the distal tibiae (85%). An interesting discrepancy was apparent at the wrist joint where 100% ($n = 10/10$) of distal radii displayed the lesions, whereas they were only present in 66.7% ($n = 8/12$) of distal ulnae. In their study of modern patients Shapiro et al. (1979) recorded that 85% of distal radii displayed osteochondromas, while lesions were present in 80% of distal ulnae. The reason for the discrepancy between the archaeological and modern data is unclear.

3.2.3. Disproportionate short stature

Information on estimated living stature was available for six of the 14 adults and older adolescents. The adult female recovered from Que, Que, Zimbabwe, had an estimated living stature of 146 cm, and was reported as having had an abnormally short stature that may have been related to MO (Singer, 1962). The woman retrieved from St Clemens, Visby, was estimated to have had a living stature of approximately 145 cm (Sjøvold et al., 1974). Although no comparative stature information was provided it would seem probable that this would have been an abnormally small stature for a Medieval Gotlander. The adult male retrieved from St Nicholas' Shambles, London, was described as having been of 'beneath average height for the population', but the precise stature value was not provided (White, 1988). The adult male recovered from Kilshane, Co. Dublin, Ireland, had an estimated living stature of 161 cm. This individual was the shortest adult male recovered from the site and his stature was notably lower than the average male value of 173 cm (Buckley, 1991). The estimated living statures of adult males at Ballyhanna, Co. Donegal, Ireland, had a range of 147.7–181.6 cm, with a mean of 167.1 cm. As such, it is evident that Skeleton 197, with an estimated living stature of

166.8 cm, was of average height, while Skeleton 331, with an estimated living stature of 158.3 cm, was shorter than average. Clinical work has indicated that a disproportionate short stature is found in 37–44% of affected individuals (Bové and Hogendoorn, 2002); five of the six archaeological individuals with the condition appear to have had abnormally short statures (83.3%; $n = 5/6$). The stature is considered to be disproportionate because limb involvement is greater than spinal involvement (Shapiro et al., 1979). It is interesting to note that Skeleton 197 from Ballyhanna displayed a relatively mild manifestation of the condition and it is possible that as a consequence the individual's stature had been unaffected. In a study of 32 modern patients Shapiro et al. (1979) reported that, while the majority of the affected individuals were below average in height, all but one of them remained within the normal range.

3.2.4. Inequality in long bone length

Evidence pertaining to the presence of unequal long bones was available for eight of the 14 adults or older adolescents. The data available for the individual recovered from Que Que, Zimbabwe, was suggestive that there may have been discrepancies of long bone length but the information provided was not sufficiently complete for this conclusion to be drawn with certainty (see Singer, 1962). In Burial no. 204/68 from the cemetery at Pruszcz Gdanski, Poland, the right radius and ulna were described as having a 'distinctly shortened' appearance when compared with the other long bones, although the bones of the right forearm were not present (Gładkowska-Rzeczycka and Urbanowicz, 1970). In the adult female recovered from St Clemens at Visby, Gotland, the right humerus (247 mm), left radius (175 mm) and left ulna (199 mm) were shorter than their opposing bones which measured 271 mm, 194 mm and 206 mm respectively (Sjøvold et al., 1974). The bundle burial from Uxbridge Ossuary, southern Ontario, Canada, displayed an abnormally short right femur, which measured 407 mm as opposed to 431 mm for the corresponding left bone (Katzenberg et al., 1982). In Skeleton 197 from Ballyhanna, Co. Donegal, Ireland, the radius was the only bone with obvious length discrepancies, with the left bone measuring 229 mm and the right having a length of 249 mm. Skeleton 331 from the same cemetery

Forearm deformities (Masada classification)		Hand involvement	Tibiofibular diastasis		Coxa valga of hip		Valgus deformity of knee		Valgus deformity of ankle		Assymetry of pectoral girdle	Assymetry of pelvic girdle	Malignant transformation
R	L		R	L	R	L	R	L	R	L			
Type III	Prob Type III	+ (RMC1)	Prox and dist	o	–	–	o	o	Poss	o	–	–	o
Type III	–	–	o	o	–	–	o	o	o	o	–	–	o
Type III	Type III	–	o	Prox and dist	o	o	Poss	–	o	o	+	+	o
–	–	–	–	–	–	–	–	–	–	–	–	–	o
o	–	–	o	o	o	o	Poss	Poss	–	–	–	–	o
–	–	–	–	o	–	–	–	–	–	–	–	–	o
–	–	–	o	–	–	–	–	–	–	–	–	–	o
–	–	–	–	–	–	–	–	–	–	–	–	–	o
o	o	o	o	o	o	o	o	o	+	+	o	o	o
o	o	o	o	o	o	o	o	o	o	o	o	o	o
o	o	o	o	o	o	o	o	o	o	o	o	o	o
–	–	–	Prox and dist	Prox and dist	–	–	–	–	–	–	–	+	+
–	–	–	Dist	Prox	–	–	–	–	–	–	–	–	o
Type III	Type III	o	o	o	+	+	+	+	o	o	o	+	o
Type III	Type I	+ (L carpals, RMC5)	Prox and dist	Prox and dist	+	+	+	+	o	+	+	o	o

displayed clear length discrepancies of the left clavicle (L = 128.3 mm; R = 134.2 mm), the left humerus (L = 271 mm; R = 281 mm), the left ulna (L = 196 mm; R = 249 mm), the left radius (L = 214 mm; R = 217 mm) and the left fibula (L = c. 271 mm; R = c. 296 mm). Modern clinical research has indicated that inequality in limb length can arise in 10–50% of cases (Bové and Hogendoorn, 2002); the inequalities were apparent in some 62.5% (n = 5/8) of the affected archaeological individuals. Among the archaeological cases the 12 bones affected comprised four radii, three ulnae, two humeri, one clavicle, one femur and one fibula. All four of the affected individuals with inequality in the lengths of the bones of the forearms also displayed forearm deformities which will be described below. It is probable that the shortening of the forearm bones is directly related to the deformities apparent in these skeletal elements (see Masada et al., 1989; Stieber and Dormans, 2005 for discussion). Inequality in long bone length would have resulted in limb-length discrepancy during life – a common feature of MO (Stieber and Dormans, 2005).

3.2.5. Forearm deformities

Clinical literature has reported that forearm deformities can arise in as many as 30% to 60% of patients with MO (Masada et al., 1989). Forearm deformities were observable in nine of the adults and older adolescents, 55.6% (n = 5/9) of which displayed the lesions. A total of nine out of the 16 observable forearms had been affected (56.3%). The deformities were classified according to the scheme developed by Masada et al. (1989) and it was found that in eight of the affected cases Type III deformation was apparent. In Type III deformation the focus of osteochondroma formation is on the metaphysis of the distal radius and it is associated with shortening of the radius. Almost normal movement of the forearm and elbow is possible, but ulnar deviation of the wrist is slightly impaired (Masada et al., 1989). Both forearms were affected in the adult individuals recovered from Que Que, Zimbabwe (Singer, 1962); St Clemens, Visby, Gotland (Sjøvold et al., 1974) and Skeleton 197 from Ballyhanna, Co. Donegal, Ireland. It was only possible to observe the right forearm in Burial no. 204/68 from Pruszcz Gdanski, Poland, and this displayed the Type III deformity (Gładkowska-Rzeczycka and Urbanowicz, 1970). In Skeleton 331 from Ballyhanna Type III deformity was evident in the right forearm, while the more severe Type I deformity was visible in the left

forearm (Fig. 3). In Type I deformity the osteochondroma formation arises predominantly on the distal portion of the ulna. The ulna is relatively short and is associated with bowing and ulnar tilt of the radius, although the radial head is not dislocated. In this type of deformity extensive impairment of forearm rotation is evident, although movement of the elbow is normal (Masada et al., 1989). In a study of MO patients with untreated forearm deformities it was reported that only 13% of the individuals experienced pain or physical limitations (Noonan et al., 2002a). It was also reported, however, that out of 77 upper limbs 40 displayed notably poor function in terms of grip, strength, pinch, range of motion and hand strength (Noonan et al., 2002a).

3.2.6. Hand involvement

Defects of the hand bones were only reported for two individuals and would only have been observable for a total of six of the adults and older adolescents (33.3%; n = 2/6). The right first metacarpal was reported as having been abnormally short in the skeleton from Que Que, Zimbabwe. It is not clear if the corresponding left bone was affected. Osteochondromas were also present on a number of metacarpals (Singer, 1962). In Skeleton 331 from Ballyhanna, Co. Donegal, Ireland, the right fifth metacarpal was notably shorter than its left counterpart. In addition, all of the left carpals displayed a normal morphology but were notably smaller than their right counterparts. Osteochondromas were visible throughout the metacarpals and hand phalanges. Involvement of the hand has been reported in 30–79% of modern patients with MO. Osteochondroma formation is believed to contribute to the shortening of the metacarpals and phalanges but brachydactyly has also been observed without any associated lesions (Stieber and Dormans, 2005).

3.2.7. Tibiofibular diastasis

It was possible to examine 13 of the adults and older adolescents for evidence of tibiofibular diastasis in one or both legs. The lesions were visible in five individuals (38.5% – n = 5/13 individuals; n = 8/24 legs). In two of the cases the diastasis had only affected a single leg, while in the remaining three individuals both legs had been fused. Of the eight affected legs, in six cases both the proximal and distal ends were affected, while in one leg only the proximal end had been fused and in another only the distal end was affected. In

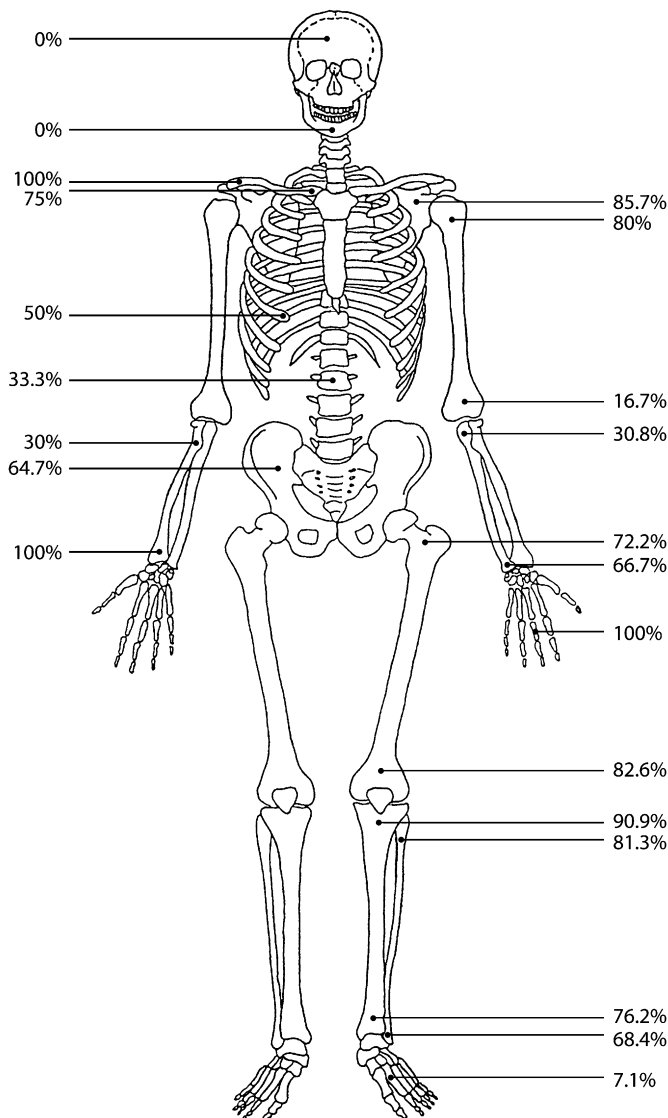


Fig. 1. Summary of the distribution of osteochondromas in the 16 archaeological cases.

both the individual from Que Que, Zimbabwe (Singer, 1962) and the adult female from St Clemens, Visby, Gotland (Sjøvold et al., 1974), only a single leg had been affected. In both cases the proximal and distal ends of the tibiae and fibulae were involved. In Skeleton 331 from Ballyhanna, Co. Donegal, Ireland (Fig. 4), and the individual recovered from Stephen Street, Dublin, Ireland (Coughlan, 2007), tibiofibular diastasis was evident in the proximal and distal ends of both legs. Skeleton B93 from Kilshane, Co. Dublin, Ireland (Buckley, 1991), displayed fusion at the proximal end of the left tibia and fibula and at the distal end of the corresponding right bones. Peterson (1989) stated that tibiofibular diastasis often occurs among modern patients as a result of the longstanding osteochondroma growth at the proximal and/or distal ends of the bones. He also reported that the diastasis was asymptomatic and did not result in functional impairment. Nevertheless, surgical intervention, which involves the removal of large osteochondromas, is often undertaken in modern sufferers of MO thereby preventing tibiofibular diastasis (see e.g. Chin et al., 2000).

3.2.8. Coxa valga of the hip

It was possible to assess the proximal femora of seven adults and older adolescents (14 femora) for coxa valga. The deformity was

apparent in both hips of Skeleton 197 and Skeleton 331 from Ballyhanna, Co. Donegal, Ireland (28.6% – $n = 2/7$ individuals; $n = 4/14$ femora; Fig. 5). In a study of 32 modern patients with MO it was found that 25% of femora displayed coxa valga but this did not present clinical problems (Shapiro et al., 1979).

3.2.9. Valgus deformity of the knee

The distal femora and proximal tibiae of nine adults and older adolescents (17 legs) were examined for valgus deformity. Femora vara was visible in the distal femora and tibia vara in the proximal tibiae of the two individuals from Ballyhanna, Co. Donegal, Ireland (see Fig. 5), and it is probable that both men would have displayed genu valgum (knock-knees) in life. Photographs of the bones of the individuals recovered from St Clemens, Visby, Gotland (Sjøvold et al., 1974), and Uxbridge Ossuary, southern Ontario, Canada (Katzenberg et al., 1982), were considered to show possible evidence of femora vara and tibia valgus in these two individuals. Stieber and Dormans (2005) reported that valgus knee deformities arise in 8% to 33% of modern patients with MO; within the nine archaeological individuals the condition arose with an approximate prevalence of 44% ($n = 4/9$; 41.2% – $n = 7/17$ legs). Among modern sufferers some patients undergo corrective surgery to produce acceptable alignment of the knees.

3.2.10. Valgus deformity of the ankle

It was possible to assess the distal tibiae of eight adults and older adolescents (16 tibiae) for evidence of valgus deformity of the ankle. It was considered possible, on the basis of photographs and the associated description, that the deformity may have been present in the right ankle of the individual recovered from Que Que, Zimbabwe (Singer, 1962). In Skeleton H, recovered from Tomb B50 at Jericho, Jordan, both of the ankles displayed the deformity (Lyll and Mann, 1993). Skeleton 331 retrieved from Ballyhanna, Co. Donegal, Ireland, displayed a clear valgus deformity of the left ankle, while his right ankle was normal. Following the guidelines of Shapiro et al. (1979), it was possible to classify the deformity as a Type III (severe) deformity in which the subchondral surface of the distal tibial epiphysis had slanted into the epiphyseal growth plate at its lateral third rather than at its lateral margin. Degenerative changes in the form of marginal osteophytes and macroporosity were apparent and were undoubtedly secondary to the deformity of the ankle joint. Noonan et al. (2002b) reported in their study of untreated adult individuals that, among individuals aged less than 40 years, 12% ($n = 5/43$) of ankles had signs of degenerative joint disease. Deformity of the ankle tends to arise in 2% to 54% of modern patients with MO (Bové and Hogendoorn, 2002). Among the archaeological cases it occurred with an approximate frequency of 37.5% ($n = 3/8$; 25% – $n = 4/16$ distal tibiae).

3.2.11. Asymmetry of the pectoral girdle

Asymmetry of the pectoral girdle is a recognized characteristic of MO (Peterson, 1989). The morphology of the pectoral girdle was observable in six of the adults and older adolescents, two of whom appeared to have displayed asymmetry (33.3%; $n = 2/6$). In the adult female skeleton recovered from St Clemens, Visby, Gotland, osteochondromas were visible on the clavicles and scapulae (Sjøvold et al., 1974). In Skeleton 331 from Ballyhanna, Co. Donegal, Ireland, the left clavicle was 6 mm shorter than its right counterpart, and both bones displayed pronounced osteochondromas.

3.2.12. Asymmetry of the pelvic girdle

It was possible to assess seven of the adults and older adolescents for asymmetry of the pelvic girdle, three of whom were considered to display lesions that may have caused such asymmetry (42.9%; $n = 3/7$). Both sacroiliac joints appear to have been



Fig. 2. View of the anterior aspects of the humeri of Sk 331 from Ballyhanna, Co. Donegal, Ireland, with evidence of sessile and pedunculated osteochondromas at the proximal ends.

fused in the individual recovered from Stephen Street, Dublin, Ireland. In Skeleton 197 from Ballyhanna, Co. Donegal, Ireland, the right auricular surface of the ilium appears to have been fully ankylosed to the sacrum, while the left sacroiliac joint displayed

partial ankylosis at its anterior margin. The pregnant adult female skeleton retrieved from St Clemens, Visby, Gotland, displayed the most pronounced pelvic changes and numerous osteochondromas were evident (Sjøvold et al., 1974). A number of the



Fig. 3. Anterior view of the left forearm bones of Sk 331 from Ballyhanna, Co. Donegal, Ireland, with evidence of Masada et al.'s (1989) Type I deformity.



Fig. 4. Anterior view of the lower leg bones of Sk 331 from Ballyhanna, Co. Donegal, Ireland, which were ankylosed at their proximal and distal ends as a consequence of large sessile osteochondroma formation. Valgus deformity was evident at the left distal tibia.



Fig. 5. View of the anterior surfaces of the femora of Sk 331 from Ballyhanna, Co. Donegal, Ireland, with evidence of sessile and pedunculated osteochondromas as well as coxa valga and femora vara.

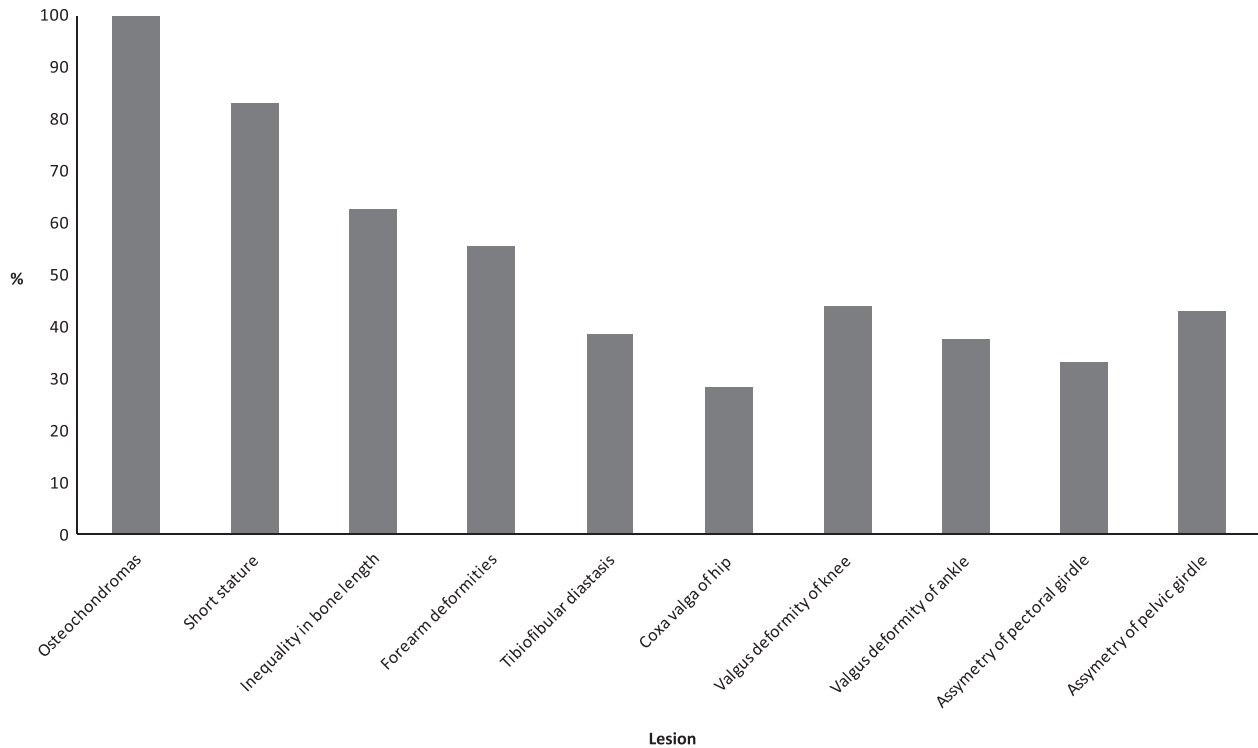


Fig. 6. Summary of the prevalence rates of the main lesions and orthopaedic deformities apparent among the 16 archaeological individuals.

osteochondromas were considered to have obstructed the pelvic space and it was suggested that these lesions would have hindered the normal rotation of the fetus and prevented its head from passing through the pelvic canal. Given the full-term nature of the fetus it was proposed that the woman had probably died during childbirth (Sjøvold et al., 1974). The presence of osteochondromas within the pelvis is known to interfere with the normal birth process and pregnancies in modern day women with MO often end in caesarean sections (Wicklund et al., 1995).

3.2.13. Malignant transformation

The only possible example of the malignant transformation of an osteochondroma was reported for the individual recovered from Stephen Street, Dublin, Ireland. A large sessile osteochondroma on the antero-lateral aspect of the right distal femur was considered to have transformed into a chondrosarcoma, presumably because of its nodular appearance and substantial size (Coughlan, 2007). Among modern cases of MO malignant transformation is the most important complication that patients might face. This situation is relatively rare, however, and arises in 0.5–3% of individuals (Bové and Hogendoorn, 2002); among the archaeological adults and older adolescents it would have had an approximate prevalence of 7.1% ($n = 1/14$). A study of 59 individuals with MO in whom malignancy had occurred found that the mean age for the diagnosis of the malignancy was 31 years. The transformation was found to have rarely arisen in individuals of less than 10 years of age or in those aged greater than 50 years (Stieber and Dormans, 2005).

3.2.14. Summary of lesions

A summary of the prevalence rates of the main lesions and orthopaedic deformities apparent among the 16 archaeological individuals with MO is provided in Fig. 6. Despite the small numbers, it is clear that osteochondroma formation was the most frequent characteristic of the condition, having occurred in all 16 cases (100%). Short stature (83.3%; $n = 5/6$), inequality of bone

length (62.5%; $n = 5/8$) and forearm deformities (55.6%; $n = 5/9$) also occurred frequently. Valgus deformity of the knee (44%; $n = 4/9$), asymmetry of the pelvic girdle (42.9%; $n = 3/7$), tibiofibular diastasis (38.5%; $n = 5/13$), valgus deformity of the ankle (37.5%; $n = 3/8$), asymmetry of the pectoral girdle (33.3%; $n = 2/6$) and coxa valga of the hip (28.6%; $n = 2/7$) were all reasonably well represented among the cohort of individuals.

4. Conclusions

Since its first identification in 1786 a variety of names for Multiple Osteochondromas have been used. This can and does lead to confusion within the literature. As such, it has been suggested that to avoid confusion palaeopathologists should follow the example of the World Health Organization and refer to the condition as Multiple Osteochondromas (Bové and Hogendoorn, 2002). The paper has also provided a checklist of the main characteristics of MO that are frequently observed in modern day sufferers and it is envisaged that this should help enable standardisation in the recording of future palaeopathological cases of the condition.

All previous publications pertaining to MO have taken the form of case study papers and no attempt has previously been made to gain an overview of the nature of the lesions apparent in archaeological individuals. The paper has shown that the manifestation of MO apparent in the 16 archaeological cases generally corresponds to the pattern of lesions evident among modern sufferers of the condition, with osteochondroma formation being one of the most obvious characteristics of the disease. It is clear that the archaeological individuals would also have suffered from the full range of orthopaedic deformities that are characteristic of the disease today. The archaeological individuals, however, would obviously not have been able to have availed of the range of surgical procedures that are undertaken today to help correct these deformities and alleviate the many associated unpleasant side-effects. As such, the study of archaeological cases of MO can be of benefit to clinicians

working with modern cases of MO since it affords them with the opportunity to gain information about the natural progression of the condition in untreated individuals.

Life must have been difficult for certainly the most severely affected of the archaeological individuals with MO and no doubt they would have struggled with pain, fatigue and mobility problems throughout their lives. Indeed, it has been reported that most modern patients with MO experience pain – approximately half of which is generalised (Bové, 2008). The reality of the difficulties involved with being a disabled individual in the past can perhaps be illustrated through the case of an 18 year old girl from the Ojibway community in Manitoba who had a severe forearm deformity and had been brought to the nursing station for help by her mother because of her inability to help with the housework (Dooley et al., 1988). Everyday life would potentially have been much more physically active and demanding for all individuals who lived in the past and it may well have been particularly challenging for those with the most serious physical manifestations of MO.

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Appendix A. Supplementary material

Supplementary data associated with this article can be found, in the online version, at [doi:10.1016/j.jas.2010.03.023](https://doi.org/10.1016/j.jas.2010.03.023).

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