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Palaeopathological and Palaeogenetic Study of 13 Cases of Developmental Dysplasia of the Hip with Dislocation in a Historical Population from Southern France

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ABSTRACT Dislocation of the hip can be consecutive to developmental dysplasia of the hip that is linked to a complex set of genetic and mechanical factors. The purpose of this report is to describe 13 cases of complete dysplastic hip dislocation observed in the skeletal remains of nine women exhumed from an historical gravesite dating from the 5th to 17th centuries in southern France. Despite the size of this palaeopathological series, which is the largest study published to date, findings indicate that the prevalence of hip dislocation in this historical sample was still lower than in some French areas at the beginning of the 20th century. Mitochondrial DNA sequencing demonstrated possible kinship only between two women, i.e. one who died in the period from the 11th to 13th centuries and another who died in the period from the 16th to 17th centuries. It is suggested that the tight swaddling of young infants in France up to the end of the 19th century could have been a predisposing factor for this highly debilitating disorder. Copyright © 2006 John Wiley & Sons, Ltd.

Key words: developmental dysplasia; hip dislocation; palaeopathology; palaeogenetic; DNA

Intrduction

Dislocation of the hip is usually linked to developmental dysplasia of the hip which has a variable geographical and ethnic prevalence. This severely handicapping deformity producing a highly characteristic waddling gait has left a lasting mark on history, folklore and legend, especially in rural societies. During the Middle Ages one French queen was nicknamed Joan the Lame (1293–1348). A number of family names have been derived from this disorder, with the use of a part of the French verb *boiter* that means to lame. So, the family names Boite and Boiteux, or in Breton, Le Cam, are directly linked to the waddling gait of an ancestor. The names were fixed between the 11th and 13th centuries, as the French kings issued the rules to choose a family name for registering the births, marriages and deaths with the Church. Many tales (i.e. the '*Waddling Devil*', written in 1707 by Le Sage) and legends have been developed in the West where hip dislocation has occurred sporadically in various regions. In Brittany in the 19th century, many sources were supposed to cure the dislocation of the hip. Lame young people were plunged into the water of magic fountains, and appeals

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were made to saints like Holy Corentin, one medieval bishop. The patients were often the object of rejection, and it was said that to avoid the wizards it was necessary to flee the lame one, the hunchbacked one, or the one-eyed one. The discovery of a method for early diagnosis and curative treatment at the beginning of the 20th century was a true medical revolution in the European countries where the disease was often regarded as a curse.

Few palaeopathological cases of congenital hip dislocation have been reported, with only around 30 cases in prehistoric and historical populations from the entire world (Brothwell & Sandison, 1967; Aufderheide & Rodriguez-Martin, 1997). Some diagnoses of congenital hip dislocation are doubtful (Gladykowska-Rzeczycka, 1980).

Pathophysiology of developmental dysplasia and consecutive dislocation of the hip

Dislocation of the hip is part of a spectrum of developmental dysplasia of the hip causing increasing disability due to disruption of the normal relationship of joint components. The aspect of the dysplastic acetabulum depends solely on whether or not contact is maintained with the femoral head. In minor dysplasia, the roof of the acetabulum presents an oval shape that leads to early arthritis of the hip. In dysplasia with dislocation, the femoral head slides out of the acetabulum and is retained only by the joint capsule and ligaments. When pressure is applied, the dislocated femoral head comes to bear against the external side of the ilium in the buttock, resulting in the formation of a pseudoacetabulum. When walking, the femur moves up and back with each step, thus producing the characteristic waddling gait.

Dislocation of the hip linked to dysplasia results from a complex process involving possible genetic susceptibility and mechanical factors occurring before and after birth. If dislocation occurs within the first months of life, the loss of contact between the femoral head and the acetabulum obstructs the normal formation of the articulation. Without reciprocal contact, neither the acetabulum nor the femoral head develops normally. The acetabulum assumes a triangular shape with three blunt edges, blocking reduction of the femoral head. At this point the dislocation becomes irreversible.

Genetic susceptibility

In most modern populations, the dysplastic and unstable hips are 4 to 8 times more common in females than males (Bjerkreim & Arseth, 1978; Higuchi *et al*., 1984; Mirdad, 2002). Family history has been noted in 3% to 12% of cases. In twin births the risk for the second infant, if the first infant has a dysplasia of the hip, is 42.7% in monozygotic sets and 2.8% for dizygotic sets (Coleman, 1978). The nature of hereditary factors is controversial. Most investigators have stressed the importance of excessive laxity of the coxofemoral ligament (Wynne-Davies, 1970).

Mechanical factors

Mechanical factors occurring during pregnancy and birth appear to play a major role in developmental dysplasia. Experimental observation has shown that hip dislocation can be induced by applying pressure to the femur of a full-term fetus for three hours, suggesting that intrauterine constraints could have a responsibility in hip dysplasia (Helmsted, 1983). A number of predisposing obstetric factors have been identified. Breech presentation has been implicated since it occurs in only 3% to 4% of deliveries but is associated with 10% to 20% of dislocations (Clausen & Nielsen, 1988). Other factors like first pregnancy and a deficiency of amniotic fluid can cause limited fetal mobility (Gunther *et al*., 1993). Developmental dysplasia of the hip is less frequent in multiparous women (Gunther *et al*., 1993). The highest risk for developmental dysplasia of the hip appears to occur when the intrauterine posture hinders or prevents abduction and external rotation of the femur or results in mechanical pressure on the grand trochanter (Seringe *et al*., 1990). The left hip is more frequently involved than the right hip. A possible explanation for this difference is that the fetus' back is usually on the left at the end of pregnancy so that the greater trochanter is blocked in external rotation against the mother's spine (Bracq, 1994). The role of femoral anteversion promoting exteriorisation of the femoral head in hip dysplasia remains controversial (Weinstein, 1987).

Another mechanical factor that has been implicated in the dislocation of the hip in case of developmental dysplasia involves traditional methods of handling newborns. Some investigators have suggested that tight swaddling, although not a direct cause, can maintain dislocation if it is already present. Use of cradleboards that maintain the infant's legs in adduction has also been associated with the higher incidence of hip dislocation in some populations, e.g. Navajos and Lapps (Holck, 1991). The use of tight swaddling that maintains the infant's legs pressed tightly together after birth has also been implicated in the development of dysplastic hip dislocation. Swaddling was widely used in Western Europe during the Middle Ages (Alexandre-Bidon, 1986). Newborns were wrapped from the feet up to the shoulders until walking age. Swaddling was terminated by a last turn from the right shoulder to left side of the waist. Despite the protests of the writer Jean-Jacques Rousseau in 1792 (Rousseau, 1999), the practice of swaddling persisted longer in France than in other countries. Indeed, swaddling was still widespread in France at the beginning of the 20th century, whereas it was abandoned in England during the first half of the 19th century following urging by writers including John Locke in 1693 (Adamson, 1922) and physicians including William Cadogan in 1749 (Cadogan, 1749). Conversely, initial treatment for congenital hip joint dysplasia in its early stages involves the use of traction bandages to maintain the femurs in adduction (Weinstein, 1987).

Epidemiology of developmental dysplasia of the hip

In contemporary populations, epidemiological surveys have identified several high-prevalence zones for developmental dysplasia of the hip, e.g. among island populations (Maricevic, 1995) and ethnic groups such as native North Americans (Walker, 1977; Coleman, 1978; Ghibely, 1990). Although the overall frequency of developmental dysplasia of the hip is similar in western, central or eastern France, various local and regional pockets have been noted. At the beginning of the 20th century, several high-prevalence zones for dislocation of the hip were identified in France. Le Damany (1912) reported a rate of 50% to 60% in the region around Douarnenez in Brittany. A rate of up to 70% was observed in Central France (Judet, 1964). Southern France was not considered a high-density zone for the condition.

Today the prevalence of childhood dislocation of the hip is low, thanks to routine screening in neonates to allow early diagnosis and treatment to prevent progression of unstable hips to dysplasia. In Europe the current incidence of dysplastic hip at birth varies considerably from 1.75% in Northern Ireland (Patterson *et al.*, 1995) and 4.4% in Spain (Padilla Estaban, 1990) to 22% in Norway (Bjerkreim & Arseth, 1978) and 22.5% in Sicily (Di Bella, 1997).

Developmental dysplasia of the hip is much less frequent outside Europe. In China the prevalence is 10-fold lower than in European populations (Hoaglund, 1981). The number of cases is also low in Japan, where family history appears to be implicated (Higuchi *et al.*, 1984). In sub-Saharan Africa congenital hip dislocation is almost unknown (Roper, 1976; Skirving & Scadden, 1979).

Few cases of dislocation of the hip linked to dysplasia have been observed in prehistoric (Baudoin, 1909) and historical populations from France. One case was observed in the churchyard (12th to 14th century) of St Pierre de l'Almanarre in Hyeres (Var) located 60 km south of Digne (Mafart, 2004). This case was the only occurrence in a total of 200 skeletons examined in this sample. Since previous examination of 200 other skeletal remains from Provence had not revealed any cases of this condition, the total prevalence in our experience in Provence is 1 in 400 (0.25%) (Mafart, 1983). This is close to the figure observed for the same period in other Middle Ages populations from northern and eastern France (2/700, i.e. 0.28%) (Blondiaux & Millot, 1991).

However, we have observed several cases of hip dislocation in the burials from a cemetery dating from the 5th to 17th centuries in southern France. Considering the possible involvement of kinship between individuals, it was decided to undertake a genetic study based on recent ancient DNA (aDNA) analysis techniques (Higuchi *et al.*, 1984; Pääbo, 1985). Indeed, the scope of application for aDNA analysis in palaeopathology (Salo *et al.*, 1994; Filon *et al.*, 1995; Donoghue *et al.*, 2004) has been greatly enlarged thanks to advances in molecular biology (Pääbo *et al.*, 1989). In most studies the DNA content from mitochondria has been used for analysis. Mitochondrial DNA (mtDNA) has demonstrated several documented advantages for phylogenetic assessment of populations, including a high number of copies per cell, a high mutation rate, and single-parent and exclusively maternal transmission (Macaulay *et al.*, 1999).

The purposes of this study were (i) to perform palaeopathological examination of the cases of hip dislocation in relation to developmental dysplasia of the hip; (ii) to assess the kinship between subjects by analysing mtDNA; and (iii) to assess resulting data in a sociocultural context.

Material

All cases of dislocation of the hip were specifically identified by systematic screening during the bioarchaeological assessment (supervisor B. Mafart) of 1450 skeletons removed from the Notre-Dame-du-Bourg cathedral gravesite in Digne, France (Alpes-de-Hautes-Provence). The site was used for the burial of local inhabitants with no apparent social class distinction from the 5th to 17th centuries. Based on archaeological stratigraphy and numerous radiocarbon dates, use of the site can be divided into five time periods: 4th to 7th centuries, 8th to 11th centuries, 11th to beginning of the 13th century, end of the 13th to the 15th century, and 16th to 17th centuries (Démians d'Archimbaud, 1989). Most skeletons were classified into one of these periods, but dating of some graves was less precise (e.g. 11th to 15th centuries).

Methods

All the subadult and adult hip bones with a well preserved acetabulum were considered; a total of 990 such hip bones was collected.

Determination of sex and estimation of age at time of death

Sex was determined according to the method described by Bruzek (2002). Age at time of death was estimated by studying the epiphyseal union of long bones in the extremities of young subjects (Krogman & Iscan, 1986), and the alteration of sacroiliac joint surfaces in adults (Schmitt & Broqua, 2000).

Palaeopathological study

Acetabular lesions in developmental dysplasia of the hip are graded according to the classification system described by Dunn (1976). The stage I of Dunn is a dysplasia with subluxation. In medicine, the diagnosis is based on the relative position of the femoral head in the hip as observed by radiography. The diagnosis of stage I is difficult on dry bones. It is based on the relative development of the roof of the acetabulum and on the depth of the cavity (Mafart, 1983). The limits between normal and pathologic acetabulum are not clear. Dislocation is much more easily diagnosed. For this reason, stage I was not included in this study, which focused on stages II and III of developmental dysplasia of the hip. Stage II is defined as non-permanent luxation with the presence of a neo-articulation contiguous with the acetabulum, and stage III as permanent luxation with a well-developed false acetabulum in the iliac fossa and transformation of the acetabulum into a shallow triangular depression. Whenever possible, depending on the state of conservation, palaeopathological study included assessment of the morphology of the femoral head and measurement of the neck-shaft angle (Anderson & Trinkavs 1998) and femoral neck anteversion angle (Olivier, 1960). Measurement of the femoral neck anteversion angle is not easy on dry bones, with a lack of reproductibility (Stirland, 1994), so the angles were measured three times and the mean value was used for each bone. The purpose in the measurement of these angles in developmental dysplasia of the hip is to determine whether the neck-shaft angle is reduced (angle $< 120°$) and/or the femoral neck has an anterior position (angle $> 25°$). These anomalies are frequently observed in developmental dysplasia of the hip and are currently measured in medical practice.

Palaeogenetic study

Extraction of aDNA from long bone fragments (femur and tibia) was performed using the technique described by Béraud-Colomb *et al.*, (1995; Kéfi *et al.*, 2003).

The aDNA sequences obtained after the sequencing reaction were aligned with the mtDNA reference sequence, i.e. the Anderson sequence or the Cambridge reference sequence (CRS) (Anderson *et al.*, 1989), using the Blast 2 sequence (Tatusova & Madden, 1999) and ClustalX (Thompson, 1997) software packages. Variations in each specimen were checked by comparing numerous sequences. Substitutions were validated only if they were found by two separate PCR (Polymerase Chain Reaction) procedures on specimens from two different extractions. Individual haplotypes were defined based on the mtDNA polymorphism characterising each sequence. Haplotypes were classified into continent-specific haplogroups (Torroni *et al.*, 1996).

Statistical analysis

Categorical variables were compared using the chi-square test and Fisher's exact test. Differences were considered as significant at the conventional level of $p = 0.05$.

Results

Developmental dysplasia of the hip with dislocation was noted 13 times in 9 individuals (table 1). Data and descriptions for each individual follow below.

Individual 1

This case involved the left hip bone of a mature adult woman dated to the 8th to 11th centuries. The acetabulum is triangular with no cup and there is a shallow depression in the middle of the external part of the ilium. Because this specimen was found in a tomb containing several individuals, reconstitution of the complete skeleton and examination of the femurs was not possible. No evidence of dislocation was found in the other hip bones in the tomb.

Individual 2

This unilateral left case involved the remains of a mature adult woman dated to the 11th to 13th centuries. Examination of the complete hip bone showed a normal acetabulum on the right. The left joint had a triangular acetabulum with a large false acetabular cavity on the external aspect of the iliac bone (Dunn III) (Figure 1). This cavity had a vertical extension to the external surface of the ilium. The femoral head had a flattening deformity with a large, highly arthritic defect in the lower posterior aspect. The shape of the false acetabular cavity was consistent with loose movement of the femoral head. The femoral neck anteversion was 35° and the neck-shaft angle was 100°.

Individual 3

This bilateral case involved the remains of a female adolescent dated between the 11th and

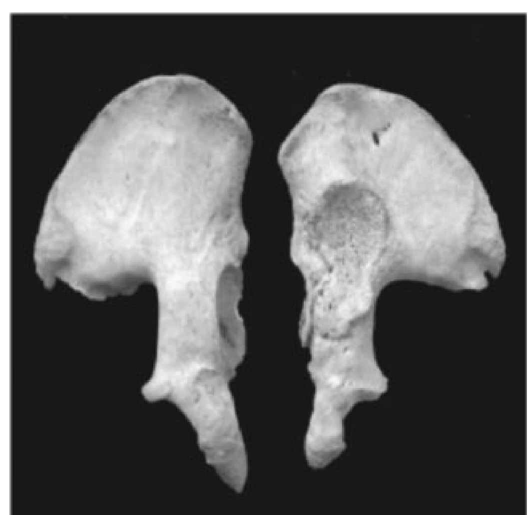

Figure 1. Unilateral (left) dislocation of the hip, case 2.

15th centuries. The right acetabulum had a triangular shape completely incompatible with reduction of the femoral head (Dunn type III). A slight indentation corresponding to the contact site of the dislocated femur was visible in the middle of the external surface of the ilium. The posterior part of the head of the right femur exhibited a flattened uneven surface. The neck-shaft angle was 112°. The left acetabulum was also dysplastic (type III) and the external side of the right ilium displayed a pronounced indentation caused by the femoral head. The middle area of articular surface of the head of the femur was flattened. The neck-shaft angle of the left femur was approximately 118°.

Individual 4

This unilateral left case was observed in the remains of a 50+-year-old woman dated to the 16th to 17th centuries. Only a fragment of the hip bone was found. The preserved upper edge of the acetabulum was incompatible with reduction of the femur head and a false acetabulum was clearly visible on the external side of the ilium that had a polished effect consistent with Dunn type III dislocation. The fragment of the preserved femoral head showed complete destruction of the joint surface and part of the femoral neck, with presence of an eburnated bone plate.

Individual 5

This unilateral left dislocation was observed in the remains of a young woman dated to the 16th to 17th centuries. A shallow depression could be seen on the ilium as evidence of contact with the femoral head. The left femoral epiphysis was dysplastic with a triangular, flattened head. The femoral neck anteversion was 20° and the neck-shaft angle was 109°.

Individual 6

This bilateral hip dislocation was observed in the remains of an adolescent female dated to the 16th to 17th centuries. There was no visible mark on the ilium. Fusion of the hip bones was still incomplete but the acetabulum was shallow and presented a clear-cut triangular shape (type III). Both femoral heads were deformed with uneven flattening of the lower posterior surfaces. The neck-shaft angle was approximately 110° on both sides. The femoral neck anteversion was 45° on the right.

Individual 7

This bilateral case was observed in the remains of an adult woman dated to the 16th to 17th centuries. The right acetabulum had a triangular shape preventing reduction of the femoral head. A neo-acetabular cavity could be seen on the external aspect of the ilium (Dunn type III). The femoral head was flattened and presented signs of major arthritis, suggestive of poor mobility. The neck-shaft angle was 115° and femoral neck anteversion was 18°. The left acetabulum was also unformed with a triangular aspect (Figure 2). There was a neo-acetabular cavity that extended upwards with flattening of the upper external edge of the acetabulum (Dunn type III). The head of the femur displayed a flattened arthritic appearance with clear-cut polishing of the upper

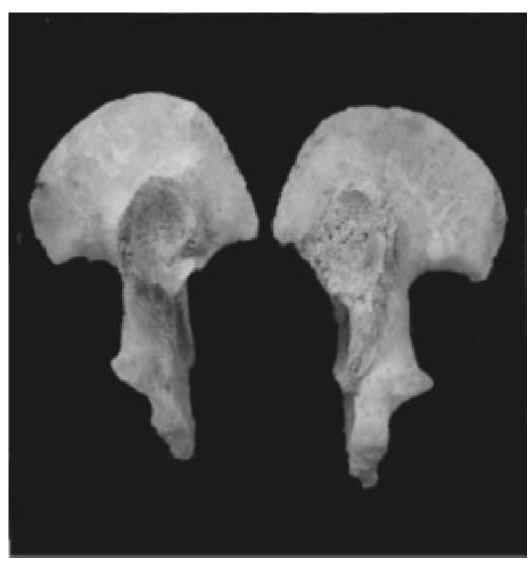

Figure 2. Bilateral developmental dysplasia, case 7.

anterior surface. This finding implies that the femoral head was mobile in the false acetabulum during walking. The neck-shaft angle was 130° and femoral neck anteversion was 27°.

Individual 8

This case was observed in the remains of a young adult woman dated to the 16th to 17th centuries. Dislocation was bilateral type III. The right acetabulum was triangular. Contact between the femoral head and the iliac bone was evidenced by the presence of a small depression in the middle of the external surface of the ilium. The femoral head presented a flattened aspect. The left acetabulum presented the same dysplastic lesions. Contact between the femoral head and the iliac bone was demonstrated by a small depression in the lower third of the bone on the left. The femoral head that had been damaged after death showed evidence of a flattened deformity.

Individual 9

This case was observed in the remains of a mature adult woman dated to the 16th to 17th centuries. Dislocation was unilateral on the right. The right acetabulum had a dysplastic triangular appearance (Dunn type III). Fragmentation of the ilium prevented examination to detect marking by the femoral head. The medial tibial plateau presented major arthritis with eburnation, probably due to difficulty in remaining upright resulting from hip dislocation on the same side. The femur was not available for examination. The left acetabulum and femur were normal.

Overall assessment of lesions

The deformity was bilateral in at least four cases. In patients with unilateral lesions, the side of involvement was the left in three cases and the right in only one.

All dysplastic lesions were Dunn type III with permanent dislocation of the hip. The acetabulum was never anatomically formed. The acetabulum was triangular in shape and of variable depth, sometimes with an indentation in the upper posterior edge caused by pressure from the hip muscles and loose ligaments. These structures held the femoral head in place to various degrees in each case. These differences account for the variability in the location of the false femoral head articulation socket on the ilium. A false articulation site was observed in all but one case involving an adolescent, and was located more or less behind and above the acetabular cavity on the external side of the ilium. The false articulation varied according to age, from a simple indentation in young individuals, to a full-fledged false acetabulum with deformation of the ilium in older individuals (Figures 4 & 5). Mobility of the neo-articulation could be more or less conserved with upwards extension of the false articulation.

Deformation of the femoral heads was observed in young subjects, and degenerative arthritis with major osteophytic formation in mature adults (Figures 3, 4 & 5). The neck-shaft angle of the dislocated femur was reduced (<120°) in seven out of eight cases. In the remaining case involving unilateral dislocation on the left, the neck-shaft angle was large but the neck of the femur was severely deformed as a result of almost complete destruction of the head. Femoral neck anteversion varied, but dysplastic values greater than 25° were observed in two of the four femurs that could be measured, and in one normal femur contralateral to the dislocated hip.

All nine cases involved women including two adolescents and seven adults (Table 1). The prevalence of dislocation was significantly higher in women compared with the overall population (Chi squared, $P = 0.047$).

From an historical standpoint, developmental dysplasia of the hip with dislocation was observed during all time periods. The greatest prevalence occurred during period of the 16th to 17th centuries, which also included the largest sample, but differences in prevalence between the different historical periods were not significant.

Given the number of evaluable hip bones per historical period, the prevalence of dislocation at the gravesite was low (range 7.3‰ to 21.3‰), with a mean value of 13.1‰ (Table 2). The

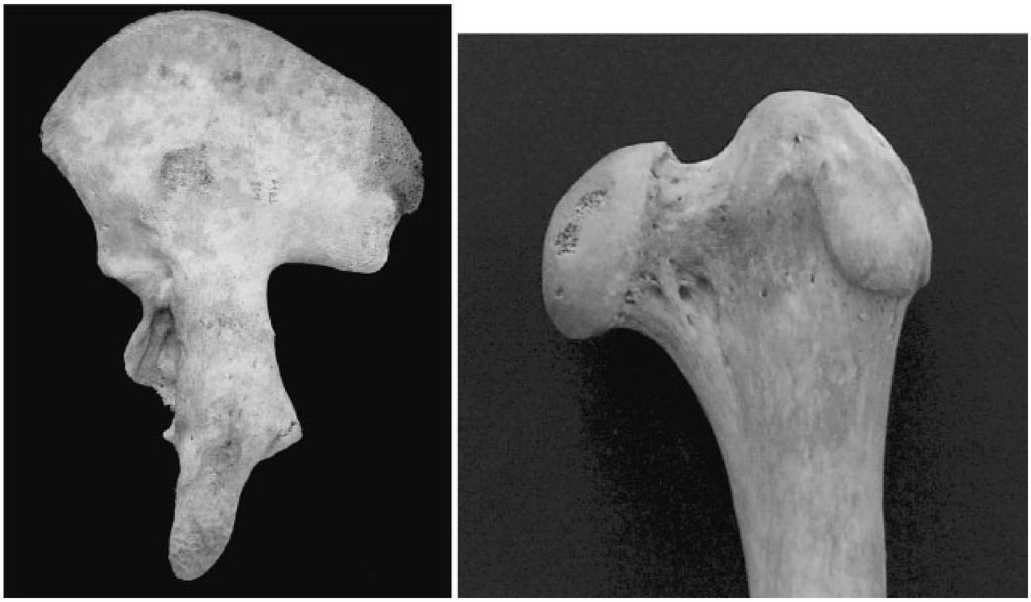

Figure 3. Dislocation of the hip, coxal bone and femoral head, juvenile female, case 3.

overall prevalence of hip dislocation in the population from Notre-Dame-du-Bourg (i.e. 1.3%) was higher than at other gravesites in Provence and in northern and eastern regions of France, but the differences were not significant.

Palaeogenetic study

Palaeogenetic analysis was feasible for seven of the nine individuals in our historical sample. In the remaining two cases palaeogenetic study was

Figure 4. Dislocation of the hip, coxal bone and femoral head, mature woman, case 2.

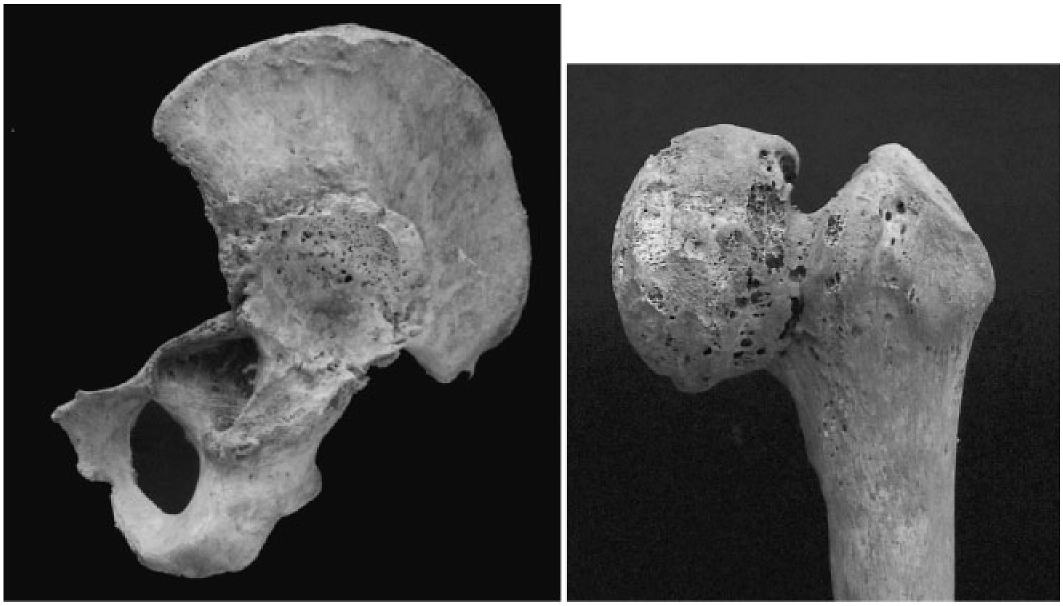

Figure 5. Dislocation of the hip, coxal bone and femoral head, old woman, case 7.

prevented by the lack of sufficient bone material from locations other than the pelvis. The alignment of the sequences with the mitochondrial DNA references is described in Table 3. Specimens 2 and 8 presented a similar sequence with the same 16192T substitution. The other individuals and the investigator presented different sequences. In comparison with the Anderson reference sample, subject 4 differed by two substitutions (16153A and 16298C), subject 5 by one substitution (16129A), subject 6 by no substitution, and subject 7 by one substitution (16126C). Subject 9 presented three mutations (16153A, 16159T and 16186T).

The haplotypes of seven specimens from the Notre-Dame-du-Bourg population were classified into haplogroups frequently detected in European populations (Torroni *et al*., 1996), i.e.:

- haplogroup H for specimens 2, 5, 8 and 9
- haplogroup T for specimen 7
- haplogroup V for specimen 4
- haplogroup H, U or R (uncertain) for specimen 6.

Table 1. Classification of hip dislocations according to Dunn, and femur angles

Cases	Period	Sex	Age	Right			Left		
				Hip bone Dunn	Femur		Hip bone Dunn	Femur	
					FNA	NSA		FNA	NSA
1	9th–10th	F	Adult	III	—	—	—	—	—
2	11th–13th	F	Adult	Normal	30	120	III	35	100
3	11th–15th	F	14–16	III	—	112	III	—	118
4	16th–17th	F	Adult	Normal	25	120	III	—	—
5	16th–17th	F	Adult	Normal	—	—	III	20	109
6	16th–17th	F	14–16	III	—	110	III	—	110
7	16th–17th	F	Adult	III	18	115	III	27	130
8	16th–17th	F	Adult	III	—	—	III	—	—
9	16th–17th	F	Adult	III	—	—	normal	30	125

NSA, neck shape angle; FNA, femoral neck anteversion angle.

Table 2. Prevalence of hip dislocations during different time periods in the overall sample, and in the female samples (the 11th to 13th century and 13th to 15th century periods were combined)

Period	Hip dislocation	Overall sample		Female sample	
		n	%	*n*	%
5th–8th	0	19	0	8	0
9th–10°th	1	47	2.12	16	6.25
11th–15th	3	412	0.72	152	1.97
16th–17th	9	465	1.93	197	4.57
Unknown date	0	47	0	14	0
Total	13	990	1.31	387	3.35

Discussion

Exclusive female involvement and the predominant left-side location noted in this study are consistent with contemporary epidemiological data on dysplastic hip dislocation. Although the lesions observed here varied, the basic diagnostic criteria as described by Aufderheide & Rodriguez-Martin (1997) were always present, i.e. a small triangular-shaped acetabulum, a depression or false acetabulum on the external side of the ilium, and variable mobility of the femoral head against the ilium. Residual mobility was ascertained based on progressive development of arthritis in the false socket, sometimes associated with arthritis and destruction of the femoral head.

The highest prevalence of dysplastic hip dislocation was noted during the most recent time period, that is the 16th to 17th centuries, but was still lower than the prevalence reported regionally at the beginning of the 20th century in the high-prevalence zones in France (Le Damany,1912) and in the Po plains area of Italy (Ortolani, 1948).

The palaeogenetic data were indicative of a possible maternal family link between two women who lived five centuries apart, with a unilateral left dislocation for case 2 and a bilateral one for case 8. The palaeogenetic testing requires great caution not only to prevent contamination by contemporary exogenous DNA, but also to verify the reproducibility of results using different extracts and separate PCR for each specimen. The possibility of contamination by investigator DNA was ruled out by searching the investigator's sequences to detect the presence of the polymorphisms found in the different sequences of test samples. No correspondences were found.

It can be excluded that these similar sequences are a result of chance. However, the prevalence of the mutation 16192T in the modern human population is low (rank 15) (Meyer *et al.*, 1999). The prevalence of this mutation is lower than the mutation 16129A (rank 1) and the mutation 16126C (rank 8). It was not possible to study the prevalence of these mutations in the rest of the archaeological sample but the haplotypes observed in the seven specimens are similar to those observed in modern European populations.

Table 3. Alignment of sequences of the different individuals and the investigator, compared with the Anderson reference

Specimens	Period	16054		16121					16126			16129		16153		16159		16186	16192	16223
Anderson		A	...	T	A	T	T	G	T	A	C	G	...	G	...	C	...	C	C	C
2	11th–13th	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	T	—
4	16th–17th	—	—	—	—	—	—	—	—	—	—	—	—	A	—	—	—	—	—	—
5	16th–17th	—	—	—	—	—	—	—	—	—	—	A	—	—	—	—	—	—	—	—
6	16th–17th	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—
7	16th–17th	—	—	—	—	—	—	—	C	—	—	—	—	—	—	—	—	—	—	—
8	16th–17th	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	T	—
9	16th–17th	—	—	—	—	—	—	—	—	—	—	—	—	A	—	T	—	T	—	—
Investigator		—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—	—

The fact that the mutation 16192T and this possible family link only concerns two cases out of 9, and not the whole sample, is consistent with the low prevalence of familial forms currently observed in modern populations, except in homozygous twins.

Swaddling of newborns probably practised by mothers in Digne, as elsewhere in medieval France, could have been a factor in worsening dysplastic hips already existing at birth.

Due to its low prevalence, dysplastic hip dislocation has rarely been reported in archaeological populations. Although the 13 cases of complete hip dislocation observed in the skeletal remains of nine women in this sample make up the largest archaeological series of hip dislocations ever published, this study does not provide sufficient epidemiological evidence to support the hypothesis of a focus of developmental dysplasia of the hip between the 5th and 17th centuries in the city of Digne comparable to the foci described in other regions of France and northern Italy at the beginning of the 20th century.

Application of palaeogenetic methods has great potential for the study of hereditary diseases and deserves further development.

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