

## Three cases of developmental dysplasia of the hip on partially mummified human remains (Roccapelago, Modena, 18th Century): a study of palaeopathological indicators through direct analysis and 3D virtual models

Trois cas de dysplasie développementale de la hanche chez des individus partiellement momifiés (Roccapelago, Modène, 18<sup>e</sup> siècle) : étude des indicateurs paléopathologiques par analyses directe et virtuelle

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**Abstract** Developmental dysplasia of the hip (DDH) is caused by a delay in the development of the acetabular cavity, leading to an anomaly in the angle of the acetabular roof. As a result, the femoral head and the acetabular cavity do not interact normally. The identification of three cases of DDH among the remains discovered in the most recent unit from the crypt at Roccapelago (Modena, Italy) enabled us to discuss the presence of DDH in this region during the 18<sup>th</sup> century. All three cases are bilateral and Dunn class I, two have been identified as male and one as female. These DDH cases are very likely mainly due to mechanical factors such as the breech position of the foetus, perhaps associated with birth order. But given their overall similarity, they may also have resulted from the practice of swaddling new-borns. In addition, current data from the Italian Ministry of Health suggests that the incidence of DDH has remained stable in this region from the 18<sup>th</sup> century to the present. The ongoing study of remains from earlier units should bring a better understanding of the particular prevalence of DDH at Roccapelago.

**Keywords** Palaeopathology · DDH · Joint pathology · Natural mummies · Palaeoradiology · Risk factors

**Résumé** La dysplasie développementale de la hanche (DDH) résulte d'un retard dans le développement de la cavité acétabulaire conduisant à une altération de l'inclinaison du toit acétabulaire. En conséquence, la tête fémorale et la cavité cotyloïdienne n'interagissent pas normalement. L'identification de trois cas de DDH parmi les restes découverts dans l'unité stratigraphique la plus récente de la crypte de Roccapelago (Modène, Italie) nous permet de discuter de la présence de DDH dans cette région au cours du 18<sup>e</sup> siècle. Dans les trois cas, identifiés comme deux hommes et une femme, la pathologie est bilatérale et de classe Dunn I. Ces cas de DDH sont vraisemblablement principalement dus à des facteurs mécaniques tels que la position en siège du fœtus, probablement associée au rang de naissance. Mais compte tenu de leur similitude globale, ils peuvent avoir été causés par l'utilisation de langes chez les nouveau-nés. En outre, les données actuelles du ministère italien de la Santé suggèrent que l'incidence de la DDH est demeurée stable dans cette région, du 18<sup>e</sup> siècle jusqu'à nos jours. L'étude en cours des restes des unités stratigraphiques antérieures favorisera une compréhension plus profonde de la particularité de la prévalence de DDH à Roccapelago.

**Mots clés** Paléopathologie · DDH · Pathologie articulaire · Momies naturelles · Paléoradiologie · Facteurs de risque

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### Introduction

Developmental dysplasia of the hip (DDH) substantially characterizes an anomalous conformation of the acetabulum

to varying degrees, resulting from a delay in development of the acetabular cavity (Fig. 1). This condition has regularly been described in palaeopathology [1-7]. DDH is due to a change in the normal relationship between the acetabulum and the femoral head. In 98% of cases, it is diagnosed as the result of an event occurring in the later phase of intrauterine life, in a hip that had developed regularly until then. The aetiology of DDH is more likely to be found among external or mechanical factors [8] than among genetic factors, which can only induce greater susceptibility to articular hyperlaxity (reduced mechanical resistance of the capsule) or to a shallow acetabulum (acetabular dysmorphia) [9]. DDH generally tends to affect females more than males (5:1 ratio), because females usually have greater ligament laxity than males [4-8]. In 45% of cases, DDH is bilateral and more frequent in first-borns, especially in the case of a breech delivery [10]. Infant swaddling in the first months of life is also considered a risk factor [6,11].

In this study, we examined the partially mummified or skeletonised human remains discovered in the crypt of Saint Paul's conversion church in Roccapelago (Pievepelago district, Modena). The archaeological excavation, which began in 2009, yielded 7 multiple graves and uncovered a forgotten crypt. The latter was used as a cemetery by the small village community between the 16th and the 18th centuries and yielded more than 300 individuals, including infants, subadults and adults. The bodies were probably placed one on top of the other in the crypt from an opening in the pavement of the church (Fig. 2), thus creating a sort of pyramid in which the archaeologists identified three stratigraphic units (Fig. 3). In a significant number of individuals (n=47), especially those from the most recent unit (US 23, see Fig. 3), the anatomic connections are preserved and only partially skeletonised, with substantial parts of mummified soft tissue remaining [12]. The study of the textile remains suggests that the bodies were dressed in tunics and heavy

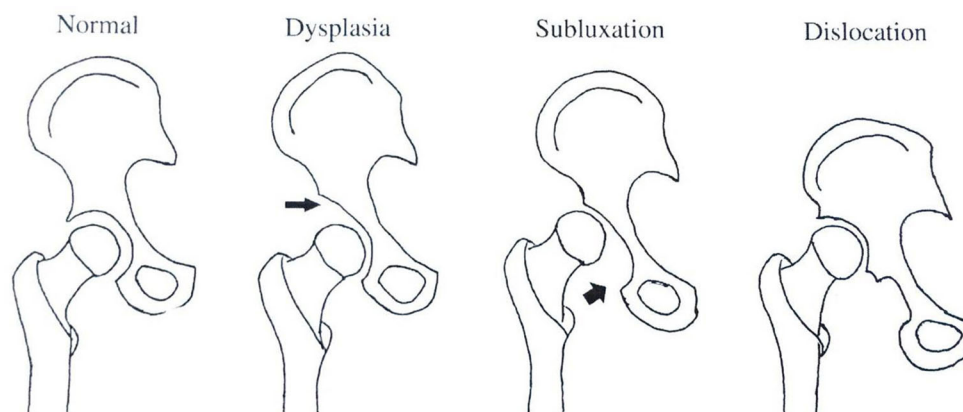
socks, and probably deposited in shrouds or sacks [13]. The mummification process was natural and for this reason, tissue preservation is generally partial and irregular among individuals. In general, the best preserved parts are tendons, skin and tissues with a lower percentage of water. This is why in some cases, what appeared to be a complete individual on site was found to be incomplete once the body was completely unclothed during the preliminary phases of the study: some anatomical regions were missing, probably due to the bones falling or rolling during body decomposition and while the crypt was being used as a cemetery. The secondary deposition of some individuals and the movements caused by the loss of body volume during decomposition probably facilitated the dislocation of bones whose balance was already unstable on the pyramid of bodies. Thus, the rest (and the majority) of the remains are comingled isolated bones that were re-associated when possible, based on pairing and joint articulation.

Preliminary genetic analysis of the remains suggests a genetic continuity between the ancient and modern populations of Roccapelago (Fixation index or  $F_{st}$  indexes). In the context of the variability of north-west Italian populations, Multi-Dimensional Scaling (MDS) analysis [14] indicates that Roccapelago is an isolated population.

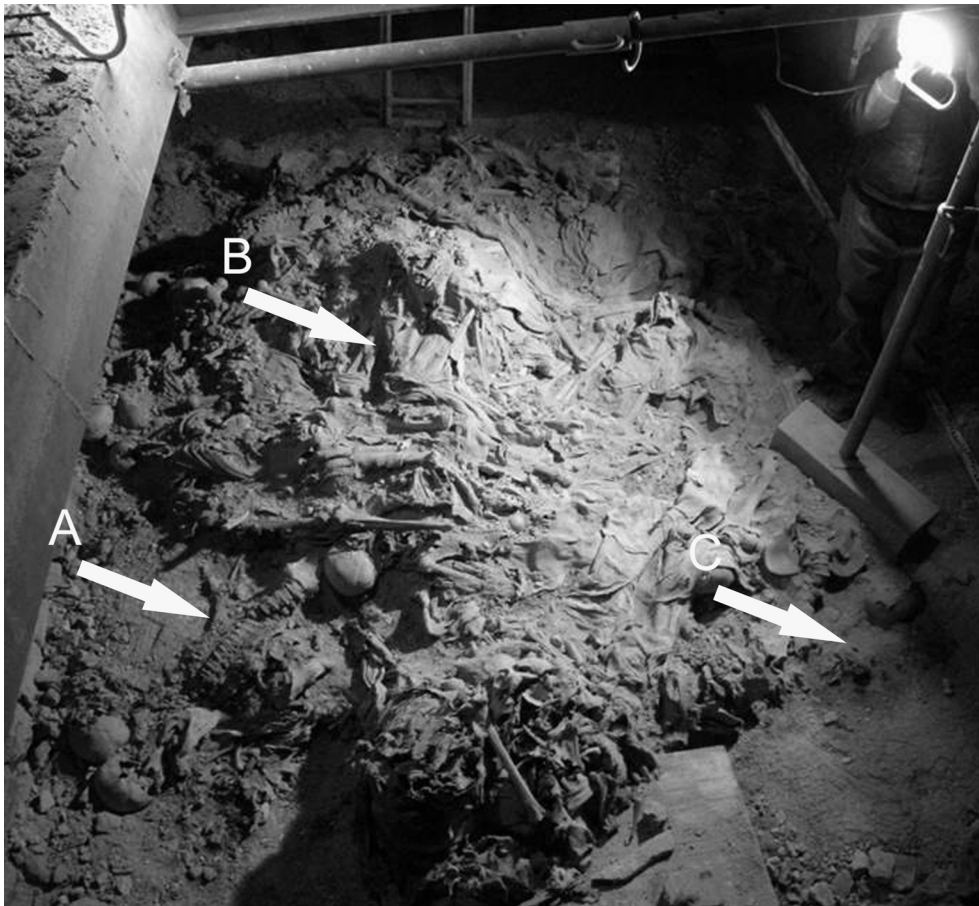
The aim of this note is to diagnose and identify DDH risk factors in this isolated 18th century mountain community. The identification of typological factors predisposing to and triggering this disease will cast new light on the aetiology of DDH.

## Materials and methods

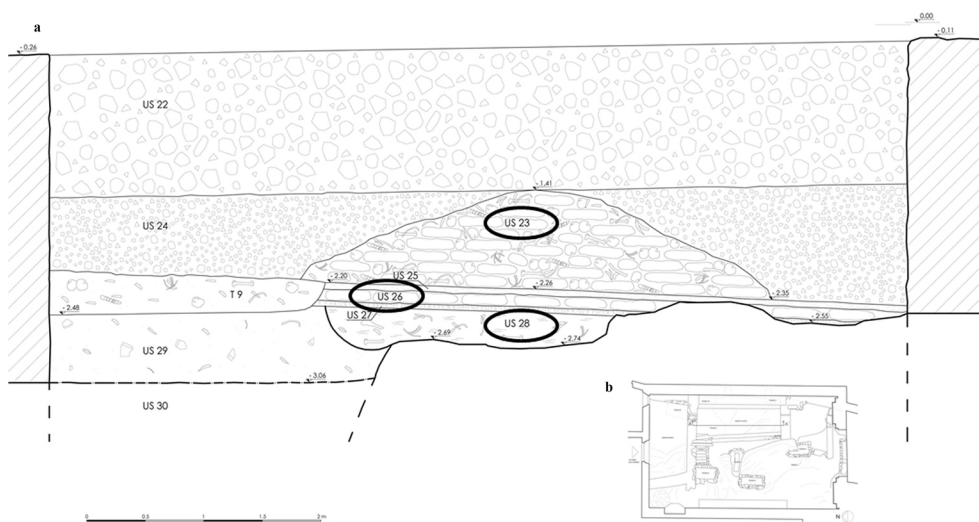
A total of 81 right and 79 left femurs and 81 right and 70 left coxal bones were analysed (Table 1). As indicated above, their state of preservation is irregular, with some coxal



**Fig. 1** Typological representation of acetabular dysplasia, subluxation, dislocation and DDH. From [7] / *Représentation typologique de la dysplasie acétabulaire, subluxation et dislocation dans le cadre de la DDH. D'après [7].*



**Fig. 2** Overall view of the crypt, showing the pyramid of bodies as they were recovered. Note the different states of preservation: A. fully skeletonised individual; B. heap of partially mummified bodies, including clothes; C. floor of the crypt / *Vue générale de la crypte, montrant la pyramide de corps telle qu'elle a été découverte. Notez la diversité de l'état de conservation : A. individu complètement squelettisé ; B. amas de corps partiellement momifiés, avec leurs vêtements ; C. fond de fouille.*



**Fig. 3** Stratigraphic units in the crypt. The layers that yielded human remains are US23, US26 and US28 / *Unités stratigraphiques de la crypte. Les couches ayant livré les restes humains sont libellées US23, US26 et US28.*

<b>Table 1</b> Sex distribution of the material analysed / <i>Distribution par sexe du matériel analysé.</i>					
	<b>Side</b>	<b>Female</b>	<b>Male</b>	<b>ND</b>	<b>Total</b>
Acetabulum	R	32	32	17	81
	L	28	27	15	70
Femur	R	26	27	28	81
	L	25	25	29	79

bones poorly preserved, incomplete or presenting mummified soft tissues. Among these, 10 individuals with exceptionally well preserved soft tissue and clothes that are destined for museum exhibition were examined via total body CT scans. All the bones analysed are from adult individuals from the more recent unit US23; the other remains are currently under study.

The CT study was carried out via a single volumetric MDCT scanner acquisition (CT 64 detector, Brilliance, Philips Medical System, Eindhoven, The Netherlands) according to the following acquisition parameters: helical CT scan, 1 mm thick layer, 1 mm reconstruction interval, 80kV and 100-200mA. The axial images obtained were re-processed using a 3D type workstation (Phillips Brilliance Portal Workspace) to obtain the multiplanar images and 3D reconstructions. The post-processing analysis was conducted in four phases. First, the axial images were assessed using different ranges of windows to identify structures of different density. A virtual 3D reconstruction of the skeleton was thus obtained. The reconstruction of each region of interest then proceeded further using different density filters, selection software and manual cuts in order to eliminate any waste material, fragments of clothes and soil. Finally, anthropometric observations were made on the multiplanar reconstructions.

The remaining material was examined by direct analysis. The individuals were sexed on the basis of the sexual characters of the hip-bones, using Probabilistic Sex Diagnosis procedure (DSP) [15]. The sex ratio is quite balanced and stands at 1:1 (39% male, 39% female, 21% not determined) (Table 1). The estimation of age-at-death combined analysis of the pubic symphysis [16,17] and of the auricular surface of the ilium [18]. The individuals were classified into three age classes (20-29 y., 30-49 y., >50 y.). Whereas there is an almost perfect balance between the sexes in the first and third classes (54% and 50% of male individuals respectively), females are clearly predominant (74%) in the second class.

Acetabular lesions in DDH were scored against three diagnostic criteria: the morphology of the acetabular roof, acetabular margin and femoral head (Table 2) [5,7]. Three additional items helped to confirm the diagnosis, although they do not identify which form of DDH is present: the morphology of the femoral neck and knee, and leg length

(Table 2) [5,7]. All cases were recorded according to Dunn's classification [19,20] following Mafart's recommendation [21]. Three grades were scored, based on the severity of the anatomical disorder: grade I: subluxation with an everted labrum; grade II: intermediate dislocation with a partially inverted labrum; grade III: complete dislocation with an inverted labrum. The same criteria were taken for the CT study, following the diagnostic indications of Freyschmidt et al. [22].

In order to exclude any other possible pathological condition, two parameters were examined: alignment of the femoral neck and of Klein's line [1,2]. Four pathologies were considered in particular in the differential diagnosis:

- Slipped Upper Femoral Epiphysis (SUFE), inferoposterior displacement of the femoral capital epiphysis with fusion of the neck in that position;
- Legg-Calvé-Perthes disease (LCP), a childhood osteochondrosis of the femoral head;
- congenital epiphyseal dysplasia, an abnormal development of the bone and cartilage of the femoral epiphysis;
- trauma.

## Results

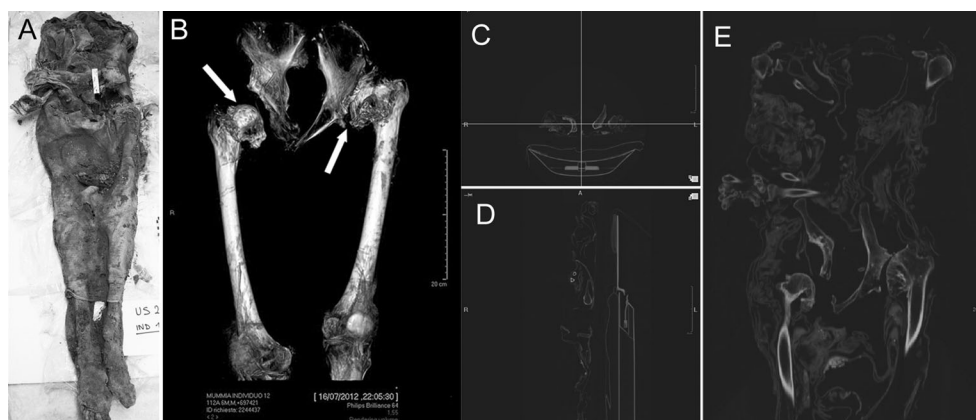
The anthropological study revealed 3 cases of DDH: Case 1 (US 23 id. 12), Case 2 (US 23 id. 47) and Case 3 (US 23 id. 6) (Table 2).

### Case 1

The anatomical connections are preserved and the remains appear to be only partially skeletonised, with substantial parts of mummified soft tissue and some textile remaining (Fig. 4A). This individual, represented by an almost complete skeleton, was classified as a female between 40 and 49 years old (range 30-49). Destined to be displayed in a site museum, she was analysed through CT scan reconstruction (Fig. 4B).

The relationship of the acetabulo-femoral joint components observed on the left side is consistent with a hip subluxation with acetabular dysplasia (Fig. 5B). The abnormal left femoral head is in a superolateral position with respect to

Table 2 Diagnostic items of DDH disease. From [7] / Critères diagnostiques associés à la DDH. D'après [7].				
Anatomical region	Indicators	Case n°1	Case n°2	Case n°3
Acetabular roof or sourcil	1) Shallow acetabulum with superolateral slope to roof (sourcil), Normal roof curves down beyond horizontal to slope inferiorly at the lateral margin	YES	YES	YES
	2) Degenerative change (cysts, eburnation, hard sclerotic bone) in the lateral part of the acetabular roof and not the medial part	YES	YES	YES
Acetabular margin	Osteophytes and cysts along acetabular rim, indicates labral tears. If acetabular dysplasia is also present but acetabular roof is smooth	YES	YES	YES
Femoral head	Eburnation, subchondral cysts and osteophytes on the medial part of the upper surface suggest subluxation if acetabular degenerative change is positioned laterally.	YES	YES	ND
Femoral neck	Femoral neck anteversion and valgus neck shaft angle	YES	YES	ND
Knee	Valgus angulation at the knee (knock knees) when femur and tibia oriented in anatomical position	ND	ND	ND
Leg length	The leg on the side with abnormal hip may be shorter than the contralateral leg	ND	ND	ND
Dunn's classification		I	I	I



**Fig. 4** Case 1 (US 23 id. 12): A. Partially skeletonised individual, with substantial parts of mummified soft tissue and some textile remaining. B. CT scan 3D reconstruction. Note the flattened and deformed femoral heads (white arrows); C. axial view; D. sagittal view; E. coronal view / Cas 1 (US 23 id. 12) : A. individu partiellement squelettisé, conservant une part importante de tissus mous momifiés et des restes textiles. B. Reconstruction tomodensitométrique tridimensionnelle : Notez les têtes fémorales aplaties et déformées (flèches blanches) ; C. vue axiale ; D. vue sagittale ; E. vue coronale.

the acetabulum, in strict anatomical contact with it but only partially overlapping. The right femoral head is slightly displaced distally relative to the acetabulum, but this is the result of taphonomic post-mortem displacements (Fig. 5B). Both proximal femora have a femoral neck in normal alignment (their heads are not displaced inferiorly but only deformed); the Klein's line along the upper border of the femoral neck intersects with the femoral head (especially on the left). We can therefore exclude SUFE with avascular

necrosis or with a congenital epiphyseal dysplasia since they are rarely bilateral [1], and also LCP disease since we observed neither specific flattening of the cephalic core nor any delay in the development of osteoarthritis. Both acetabula, especially the left one, are shallow, medially oriented and elliptical, with a deficient and hypoplastic acetabular roof (Dunn I). The left femoral head, associated with a short femoral neck, is flattened and deformed and shows signs of severe osteoarthritis, one of the most common

causes of secondary osteoarthritis in young adults [23]: numerous coarse osteophytes are present and the subcortical cavity is rough, with pores penetrating the surface (Figs. 4B and 5B). No false acetabulum can be seen on the lateral aspect of the iliac wings of the pelvis (Figs. 5A and C).

These characteristics are consistent with an advanced stage of DDH associated with secondary osteoarthritis of the hip [22]. The spine, absent due to taphonomic processes, could not be used to confirm the diagnosis.

## Case 2

This individual, consisting of a fairly complete skeleton, is a male aged over 50 with some anatomical connections preserved and only partially skeletonised (Fig. 6). The acetabulo-femoral joint was not found in anatomical connection; however, it was possible to re-associate the femoral heads functionally with their respective acetabula (Figs. 7A and C).

The articulations thus restored presented the characteristics of hip subluxation (Dunn I). On both sides, the acetabulum is shallow, medially oriented and elliptical, with a deficient and hypoplastic acetabular roof and a large eburnation zone (Fig. 7E). The femoral heads, associated with a short neck, are flattened and deformed and exhibit numerous coarse osteophytes (Figs 7A and C). The subcortical cavity is rough, with pores penetrating the surface, and shows signs of substantial osseous re-modelling and large eburnated surfaces. No false acetabulum could be observed on the lateral aspect of the iliac wings of the pelvis (Fig. 7B and D). These

characteristics are consistent with an advanced stage of acetabular dysplasia associated with a secondary acetabular coxarthrosis [19,20,23]. Analysis of the rest of the skeleton revealed a scoliotic curve to the right on the spine and muscular insertions with biomechanical stress indicators, especially at the insertions of the *gluteus minor*, *medius* and *iliopsoas* muscles. These observations suggest that despite scoliosis and dysplasia, the individual experienced moderate exercise. Moreover, the scoliotic curve observed may be the result of infantile scoliosis, a neuromuscular abnormality that can be considered as a mechanical risk factor which makes the development of DDH 10 times more likely [24].

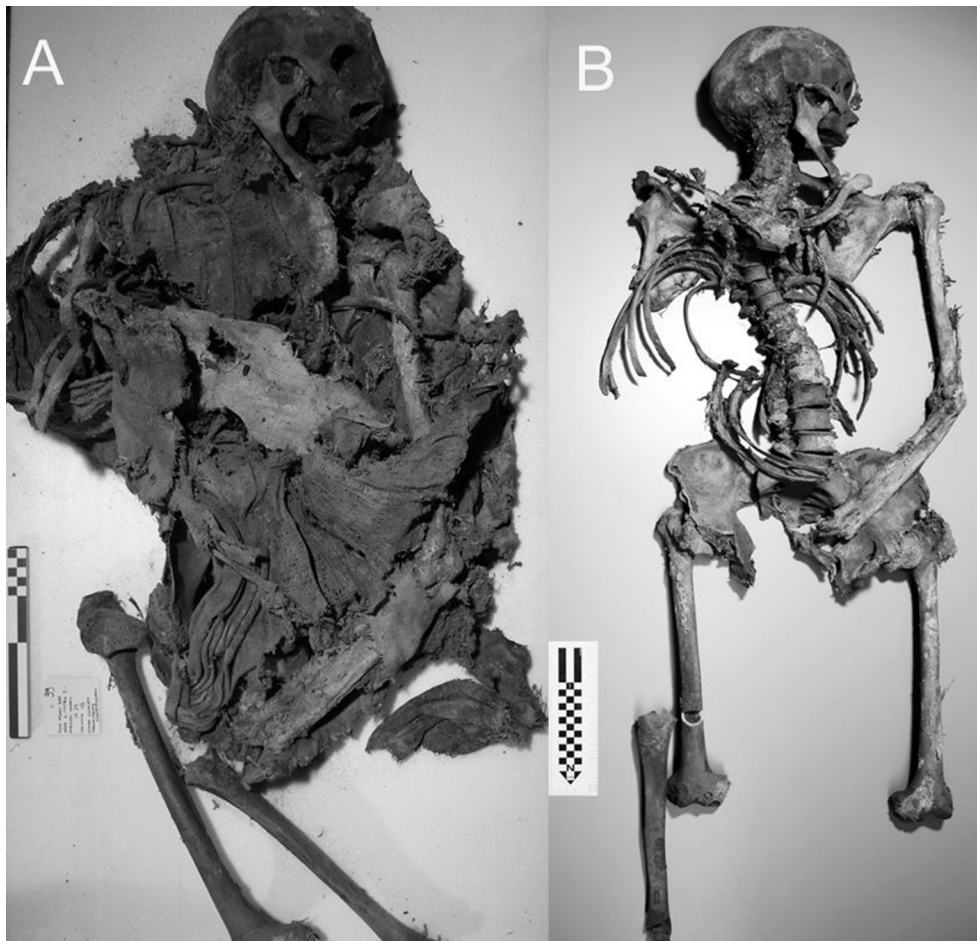
## Case 3

This case is an isolated pair of fully skeletonised innominate bones for which it was not possible to reattribute the corresponding femurs (Fig. 8A). They were classified as a 40-49 year-old male (range 30-49).

Again, both acetabula are affected, especially the left one. They are shallow, medially oriented and elliptical, with a deficient and hypoplastic acetabular roof and large eburnated regions (Dunn I). There is no false acetabulum on the lateral aspect of the iliac wings of the coxal bones (Figs. 8B and C). These observations could be consistent with an advanced stage of DDH and coxarthrosis [20,21]. However, without the femoral heads, it is difficult to safely say whether this abnormal acetabulum is a result of DDH, healed septic arthritis or hip trauma in childhood leading to avascular necrosis of the femoral head. The morphological similarity



**Fig. 5** Case 1: A. Physiologically oriented right acetabulum; B. Coronal view of the innominate bones showing the relationship of the acetabulo-femoral joint components (white circles); C. Physiologically oriented left acetabulum / *Cas 1* : A. *acétabulum droit orienté physiologiquement* ; B. *coupe frontale des bassins montrant la relation entre les différents éléments de l'articulation acétabulo-fémorale (cercles blancs)* ; C. *acétabulum gauche orienté physiologiquement*.



**Fig. 6** Case 2 (US 23 id. 47): individual with some anatomical connections preserved and only partially skeletonised, A. before unclenching, B. after unclenching / *Cas 2 (US 23 id. 47) : individu partiellement squelettisé conservant quelques connexions anatomiques, A. avant le déshabillage, B. après le déshabillage.*

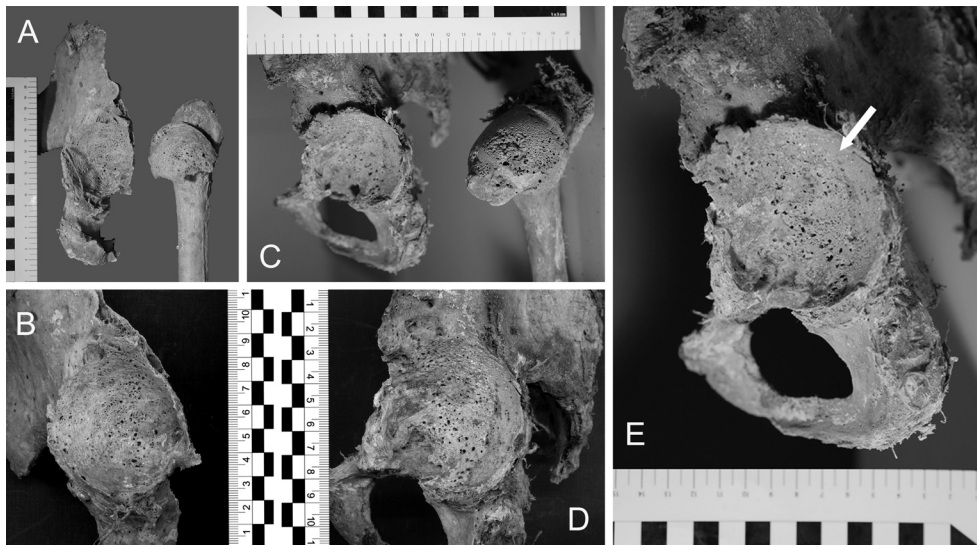
of this case with Case 2 especially (the shape of the roof, acetabular margin, eburnation zone) contributed to the diagnosis of DDH in Case 3.

## Discussion

The three partially mummified and skeletonised cases discovered in the more recent unit in the Roccapelago crypt (US23, 18<sup>th</sup> century) are bilateral and Dunn class I. Among these remains, two were identified as male and one as female; all three were adults over 40. The shape of the roof and the margin of the acetabulum excluded a diagnosis of the pathology identified as a degenerative age-related joint pathology. The presence of a probable case of infantile scoliosis on the Case 2 male, which multiplies the probability of developing DDH by 10 [24], might alter the normal incidence of DDH. The oldest unit (US28, late 16<sup>th</sup> century to early 17<sup>th</sup> century)

only yielded 11 unpaired coxal bones that showed no evidence of DDH [25]. The ongoing examination of the material from US26 (second half of the 17<sup>th</sup> century, beginning of the 18<sup>th</sup> century) will produce a better assessment of the sex ratio and could bring a possible secular trend to light.

According to our analysis, the recorded DDH condition of the acetabulo-femoral joint allowed a certain degree of flexion, whereas abduction was severely limited. The fact that the condition was also associated with severe osteoarthritis (observed in 69% of the sample analysed) in all three cases probably restricted their movement even further. However cortical bone resorption in the femurs is limited and the insertions of the short and large adductor muscles are robust, just as observed in the other individuals that do not show DDH. All these observations suggest that, despite the articular degeneration due to DDH, these individuals experienced normal muscular activity of the lower limb, and thus had normal habitual mobility.



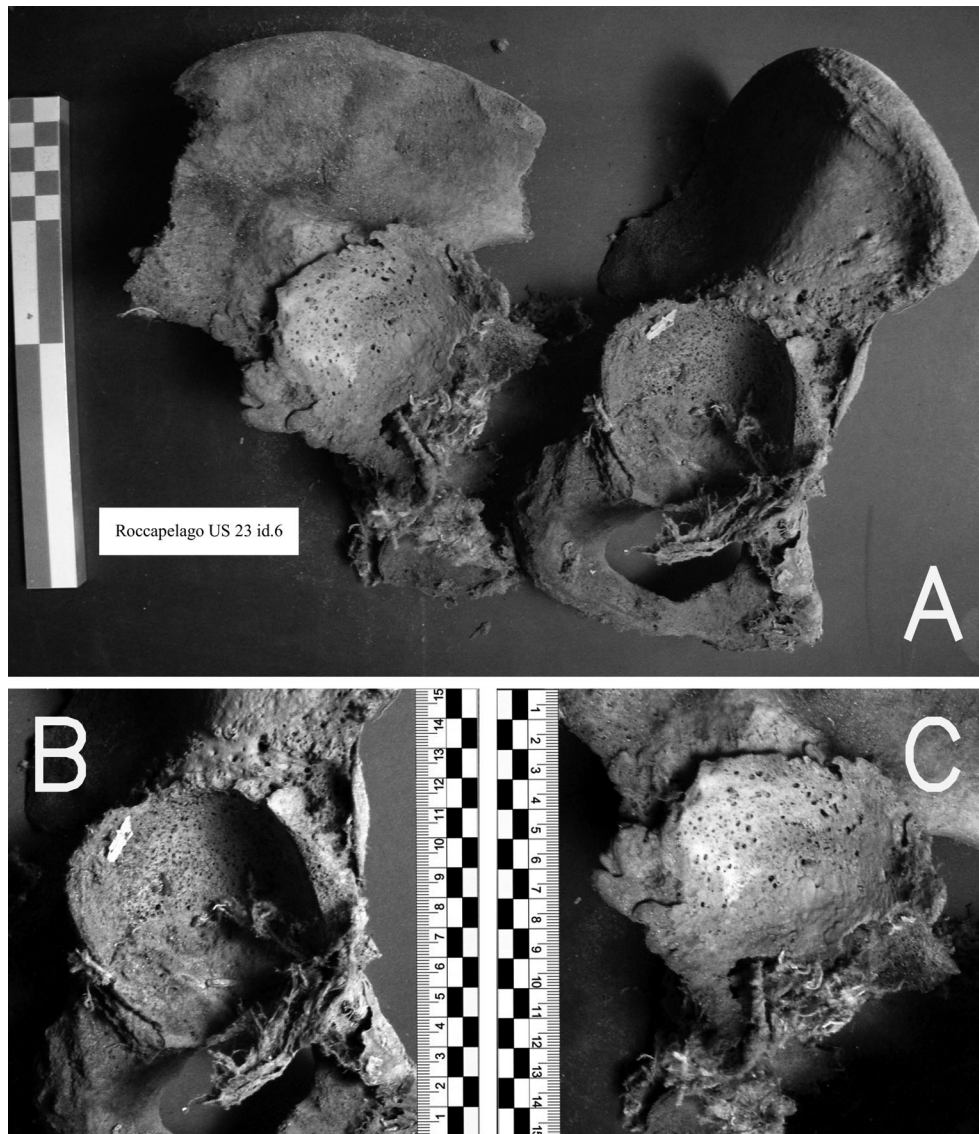
**Fig. 7** Case 2: A. Right femoral head with its acetabulum; B. right acetabulum; C. Left femoral head with its acetabulum; D. Physiologically oriented left acetabulum; E. Left acetabular roof with a large eburnation zone (white arrow) / *Cas 2* : A. tête fémorale droite et acetabulum correspondant ; B. acetabulum droit orienté physiologiquement ; C. tête fémorale gauche et acetabulum correspondant ; D. acetabulum gauche orienté physiologiquement ; E. toit de l'acetabulum gauche présentant une importante zone d'éburnation (flèche blanche).

The minimum number of elements (MNE) used in the present study is 81, based on the number of coxal bones that yielded US23; the frequency of DDH is 3.70%. According to the latest data from the Italian Ministry of Health, DDH has a 3 to 4% rate of incidence in Italy (incidence of all forms of the disease: acetabular dysplasia, subluxation and dislocation based on ultra-sound scans of babies), although some regions seem to be more severely affected, such as Emilia (up to 5%), Marche, the Aosta Valley, Basilicata (4%) and Lombardy [26]. In Europe, Poland, Germany, Spain and France also have a high rate of DDH incidence. Unfortunately, there are no statistics over a longer period of time because in the past, patients were diagnosed for DDH only as adults, so neonatal cases (97% of which tend to be cured spontaneously within the first 6 months of life [27]) were not recorded. Because of these substantial methodological differences, the frequency we observe at Roccapelago cannot be considered as a strong indicator, despite apparently agreeing with current data on DDH prevalence in the modern population living in the same area [4]. Despite the existence of a comprehensive range of archaeological studies [1-7], it is difficult to place Roccapelago within a broader historical context. Differences in methodological approaches, sampling and demographic profiles prevent any accurate comparison. Each study presents very specific cases that may reveal only little about frequency. Moreover, the criteria employed are extremely heterogeneous: some authors do not take dysplastic DDH into consideration, others only consider dislocations and some focus

on specific historical populations (e.g. Middle Ages). For all these reasons, the percentages published are very variable, ranging from 0.1% to 20% [1-7].

Analysis of the parish registers indicates that 63% of all mothers only gave birth once, which means that many inhabitants were first-borns. Unfortunately, no information on the type of delivery was recorded, so we do not know whether any births were breech. During breech births, the greater trochanter is under considerable pressure and this can often result in bilateral dislocation, especially in first-borns [10]. When infants are born head first, they are under pressure from the lumbar region of the maternal spinal cord. A higher percentage of these infants are born with a unilateral dislocation of the left hip, since most are born with the back facing left [19]. Hip dislocation may develop towards the end of gestation (the last couple of weeks, days, and possibly even during labour) due to various mechanical factors, often in combination with genetic susceptibility. After birth, the hip tends to recover from the pressure applied during gestation and develops normally. Usually, infants born with a dislocated hip are diagnosed with hip instability; if this instability continues as they grow (e.g. due to clothing), it will persist and dislocation will become permanent. In 50% of cases, the hip recovers, and as a result, either the hip reverts to normal, or anomalies such as dysplasia and dislocation persist [8]. The current incidence of breech births in Italy is between 2.4% and 4.9%, so if we apply this data to the Roccapelago remains, we would expect that between 1.9 and 3.9





**Fig. 8** Case 3 (US 23 id. 6): A. Fully skeletonised isolated innominate without corresponding femurs; B. Physiologically oriented right acetabulum; C. Physiologically oriented left acetabulum / *Cas 3 (US 23 id. 6) : A. os coxae isolés complètement squelettisés, sans fémurs correspondants ; B. acétabulum droit orienté physiologiquement ; C. acétabulum gauche orienté physiologiquement.*

individuals were born with breech presentation; this estimation is entirely consistent with the number of cases we identified. Moreover, since swaddling infants was common practice at the time, clothing may have reinforced the dislocation, thus being a contributing factor to permanent DDH in infants that may have grown up without this condition if their hip instability had not been maintained. Clinical studies tend to confirm an increase in cases of infants born with DDH during the winter, with acetabular hypoplasia either in depth or in the angles [28]. The only probable factor that could explain this data is the use of heavier or tighter fabric to protect the infant from colder temperatures [11,29-31].

## Conclusion

The most recent unit (US23, 18<sup>th</sup> century) in the Roccapelago crypt delivered three cases of DDH that seem fairly unique since they are partly consistent with the clinical statistics of the pathology: all three are bilateral cases and Dunn's grade I; two have been identified as male and one as female. The cases of DDH recorded at Roccapelago are very likely mainly due to mechanical factors such as the breech position of the foetus, perhaps associated with birth order, since first-borns represented at least 63% of the cases we were able to detect from the parish registers. The three Roccapelago cases, given their overall similarity,

may also have resulted from the practice of swaddling new-borns, a common practice that probably survived up to the pre-modern era. The prevalence of DDH (3.70%) at Roccapelago, apparently in line with the regional trend currently available for new-borns, suggests a constant incidence of the pathology from the 18<sup>th</sup> century until recent times, a condition that would most likely be prevented from persisting in adulthood nowadays. The ongoing study of the remains from earlier units should bring a better understanding of the particular prevalence of DDH at Roccapelago.

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Conflict of interest : no conflict of interest to declare.

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