

Two Cases of Joint Disease in Post-Medieval Church Cemetery of St. Ilija

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ABSTRACT

Evidence of disease was analyzed from the skeletal remains of 11 individuals dating to the post-Medieval period from church cemetery of St. Ilija in Serbia. Two individuals showed pathological condition affecting joints. It was supposed that first individual had been suffering from Legg-Calvé-Perthes disease. It seems that this condition remained untreated, with extensive bone remodeling, and that the deformity of femoral head and acetabulum caused secondary degenerative joint disease at a relatively early age of this individual. Second case was related to the bony akylosis of the hand finger, probably caused by Dupuytren's disease. In addition, we discussed development of differential diagnosis in both pathological conditions.

Key words: osteoarthritis, hip, ankylosis, contracture, cemetery, post-Medieval

Introduction

Osteoarthritis (OA) is a degenerative condition particularly affecting weight bearing joints. In the archaeological skeletal material it is the most frequently identified joint disease¹⁻³. In published paleopathological reports of the late Medieval church cemeteries in Serbia⁴⁻⁷, 15

out of 522 individuals examined had evidence of OA of the spine (3.57%), and 7 individuals showed signs of OA of the hip joint (1.34%). Majority of the reports do not specify the number of bones examined, and, consequently, calculation of prevalence will be different from the ac-

tual prevalence rate. Furthermore, it seems that in some reports only outstanding features of disease were scored, neglecting minor changes in the early stage of the disease. In this study we have concentrated on differential diagnosis of secondary osteoarthritis of the hip joint. Moreover, we are discussing possible causes of bony ankylosis found on interphalangeal hand joints. In terms of osteoarchaeological material, this type of lesion is very rare.

Material and Methods

The paleopathological evidence was recorded on the skeletal remains excavated in 1998, during the reconstruction of foundation of the Medieval church of St. Ilija in village Ba, near Valjevo, in western Serbia. The burials originated from the period between the sixteenth and the nineteenth century and were a part of the cemetery surrounding the church. The graves contained remains of people living in the parish at the time, and represented a settled, agricultural community. The sample consists of 11 skeletons, with four of them being infants and the remaining seven being adults (three males, three females and one of unknown sex). Age assessment of adults was based on Suchey-Brooks method⁸. Sex determination was based on dimorphic features of the os coxae⁹. The skeletons were studied in the Laboratory of Anthropology in the School of Medicine in Belgrade. The paleopathological analysis was based on macroscopic and radiological features.

Results and Discussion

Grave No 7.

In the male skeleton assessed to be between 30 and 40 years of age at the time of death (35.2 ± 9.4 years estimated by Suchey-Brooks method⁹), pathological



Fig. 1. Affected and unaffected femur of individual N° 7, frontal aspect.

condition of the left hip joint was identified (Figure 1, 2 and 3). Only postcranial skeleton was preserved and no other pathological changes were found on the bones.

Due to extensive new bone formation the affected acetabulum became shallow with irregular surface (Figure 2). New bone deposits within the central aspect of the acetabulum tend to over bridge the acetabular fossa. The thickness of this layer of deposit measured on its lower margin is 7 to 9 mm. Minute area of eburnated bone was preserved on the lower portion of the acetabular surface. Femoral head is flattened, enlarged and deformed, associated with thickening of the femoral neck (Figure 3). Large osteophytes were seen on joint surface and margins of femoral head, accompanied by areas of bone porosity.



Fig. 2. Acetabulum of affected and opposite side, frontal aspect.



Fig. 3. Femoral head of affected side, dorsal aspect.

Metric characteristics of pelvis and femora of affected and opposite (unaffected) side (Table 1) show that acetabulum is flattened and increased in size to accommodate the enlarged femoral head. There is shortening of the femoral neck but no significant changes of colliodiaphyseal angle.

Radiographic features of affected hip joint (Figure 4), in frontal projection (simulating clinical positioning) could be summarized as follows:

1. Irregular acetabular surface;
2. Shallow acetabulum and increased thickness of acetabular floor;
3. Osteophyte formation on the upper margin of acetabulum;
4. Subchondral sclerosis in juxtaarticular zones of coxae and femur;
5. Enlargement of femoral head;
6. Irregular surface of femoral head;
7. Cyst-like destruction in the subarticular part of femoral head, rounded in form with well-defined borders (i.e. subchondral cysts with sclerotic margins);
8. Shortening and widening of femoral neck.

Presence of osteophytes, reactive new bone formation on joint surface, pitting of the bone surface, deformation of the articular surface, no tendency of ankylosis as well as subchondral sclerosis and cysts on radiography, suggest that the left hip joint is affected by severe osteoarthritis^{10–14}. Osteoarthritis (OA) is associated with migration of the femoral head and joint space narrowing, which influences the radiographic and pathologic characteristics of the disease. On the routine frontal radiographs of the hip, three basic patterns of femoral head migration can be observed: superior, medial and axial¹⁴. In our case, new bone is laid down on inferior surfaces of the acetabulum and, as a result, there is a developed so-called »traveling acetabulum«, while the femoral head is extruded. Despite the limitation caused by the lack of soft tissue, and consequently the real position of the elements in the hip joint (on frontal radiography of specimen with simulated clinical position) it seems that the femoral head moved superolaterally. Although both superomedial and superolateral migration

TABLE 1
METRIC CHARACTERISTICS OF PELVIS AND FEMORA IN INDIVIDUAL FROM GRAVE NO. 7

Metric characteristics	Affected side (mm)	Opposite side (mm)
Cotyloischadic diameter ¹	3.5	3.9
Pubic length ²	7.3	7.3
Ischial length ³	12.3	11.3
Maximum acetabular depth	2.3	3.2
Maximum acetabular diameter	7.4	5.9
tlparCollodiaphyseal angle	137	135
Maximum diameter of femoral head	7.1	4.9
Maximum diameter of femoral neck	3.7	3.3
Maximum length of femoral neck ⁴	2.2	3.4

- 1 – direct and projective distance between the anterior border of the great sciatic notch and the adjacent border of the acetabulum;
 2 – distance between the most prominent point of the symphyseal surface and the nearest point of the acetabular rim;
 3 – the greatest distance between the midpoint of the ischial tuberosity and the acetabular rim;
 4 – direct distance between the base of the lesser trochanter and the periphery of the femoral head

are accompanied by osteophytes along the lateral aspect of acetabulum and femoral head, subchondral cysts, subluxation of the femoral head and shallow acetabulum is indicative for/of superolateral migration. This pattern is the most common type of femoral head migration, and occurs usually unilaterally, producing pro-

gressive loss of medial rotation and abduction^{13,14}. In our case, there is no damage of the opposite joint that may have happened due to additional stress in the attempts to spare the patient pain and disability resulting from affected joint. It may suggest that symptoms, which sometimes do not correlate with



Fig. 4. Radiography of pelvic girdle, frontal view.

radiological finding, were not that painful and disabling and do not correlate with severe pathological changes visible macroscopically.

Individual from grave No 7 is assessed to be a relatively young male and there are no signs of degenerative changes found on the other parts of its skeleton. Considering that OA is usually found in people beyond middle age, in this case of premature OA it is most likely that arthritis was superimposed on a pre-existing pathology. Diseased hips are prone to develop osteoarthritis at earlier age period because the mechanical defect, due to the deformity, subjects the articular surfaces to oft-repeated trauma. Murray and Jacobson¹² cited that pre-existing anatomical abnormalities appear to be responsible for approximately two-thirds of degenerative hip joint disease, while among the rest at least a half may be attributed to former undiagnosed rheumatoid process. Greenfield¹¹ quoted detailed etiology of secondary OA of the hip, which could be summarized in following groups:

1. Mechanical (fractures, athletic activity in adolescence, disturbances of stress forces);
2. Vascular (Legg-Perthes disease, slipped capital femoral epiphysis, ischemic necrosis of the femoral head);

3. Congenital (dislocation of the hip, acetabular dysplasia, multiple epiphyseal dysplasia, coxa vara);
4. Endocrine (acromegaly, obesity, hypo- and hyperparathyroidism);
5. Previous arthritis (infective, rheumatoid, psoriatic, tuberculous).

Distribution on the skeleton, lack of ankylosis, type of femoral migration, extensive new bone formation, and lack of osseous erosions and absence of other skeletal manifestations allow differentiation of pathological changes of individual from No 7 from the deformity associated with pre-existing mechanical, endocrine and inflammatory hip disorders. Monoarticular involvement, macroscopic findings and radiographic features of individual from grave No 7 suggest that several conditions may be associated with osteoarthritis of this individual: acetabular dysplasia, Perthes disease, and old congenital dislocation of the hip. The distinctive radiological features of each disease are given in Table 2, in order to make the differential diagnosis.

Acetabular dysplasia is the one of the diagnoses that should be taken into consideration in the developed osteoarthritis in this case. Superolateral migration of femoral head has often been attributed to

TABLE 2
OUTSTANDING FEATURES ENABLING TO DIFFERENTIATE THREE PATHOLOGICAL CONDITIONS THAT SECONDARY OSTEOARTHRITIS OF INDIVIDUAL FROM GRAVE NO. 17 MAY BE SUPERIMPOSED ON

Osteoarthritis associated with acetabular dysplasia	Osteoarthritis associated with Perthes disease	Osteoarthritis associated with old congenital dislocation of the hip
- Lateral subluxation of the femoral head	- Enlargement of the femoral head	- Flattened femoral head
- Shallow acetabulum	- Widening and shortening of the femoral neck	- Lateral subluxation of the femoral head
		- Shallow acetabulum
		- Shortening and anteversion of the femoral neck
		- Contralateral stress sclerosis in iliac component

acetabular displasia. However, it must be viewed with caution because it may be difficult to discern whether the acetabular flattening antedated the appearance of OA or the shallowness of the acetabulum was related to collapse and flattening of the outer aspect of the bone¹⁴. It is more likely that in this case the acetabular changes are secondary to femur head changes.

Developmental dysplasia of the hip, is a group of disorders that are mostly asymptomatic in childhood, but in adults usually become manifested due to continued mechanical stress and consequential development of osteoarthritis. This entity comprises not a complete dislocation, but a slight disturbance in anatomical position of articulated bones in hip joint. There are several internal (neonatal hip instability, acetabular hypoplasia, anteversion of femoral head, and laxity of articular capsule and ligaments) and external (intrauterine fetal position, birth mechanical trauma etc.) factors causing this disorder^{16,17}. Congenital dislocation of the hip is responsible for approximately a quarter of the so-called »idiopathic osteoarthritises« of the hip¹². Untreated deformity ultimately forms pseudoarthroses on the ilium causing premature OA. Shallow acetabulum together with deformed and subluxated femoral head is the original deformity on which degenerative changes could superimpose on. Although the findings in our case are basically similar to degenerative changes of the hip previously affected by congenital dislocation, relation between femoral head and the great trochanter, and the absence of manifestations of prolonged stress, such as contralateral bone sclerosis or cysts, do not suggest such a diagnosis.

Legg-Calvé-Perthes disease is the necrosis of immature femoral head of unknown etiology, with 10 to 20% of patients showing bilateral involvement (Resnik, 2002). The pathological process

is characterized by subsequent infarctions, bone necrosis, crushing of necrotic bone, reossification, and remodeling of femoral epiphysis that continues until skeletal maturity¹⁸. Coxa magna is the most frequent (58%) pattern of deformity in adults with Perthes disease¹⁵. Other forms are less common: coxa brevis (21%), coxa irregularis (18%), and osteochondritis dissecans (3%). Coxa magna, the deformity showing a lot of similarity with our case, is characterized by enlargement of femoral head, widening of femoral neck and nearly normal relationship between the height of the greater trochanter and the femoral head. Absence of radiodense area on the inner aspect of the femoral head representing the original zone of articular cartilage, on the radiographies of our case, and incompletely obliterated the fovea of the femoral head, argues against the diagnosis of Perthes disease. However, other characteristics such as enlarged and malformed femoral head, and thickening and shortening of the femoral neck, suggest Perthes disease as the most likely diagnosis in this case.

Grave No 2.

The skeletal remains belonged to the male individual aged between 40 and 50 years at the time of death (45.6 ± 10.4 years estimated by Suchey-Brooks method⁸). On the partially preserved skeleton marginal osteophyte were detected on the lower thoracic, and first and second lumbar vertebra. On the bones of the right arm (the left is missing) there were no pathological changes found except for bony ankylosis of the joints of the little finger (Figure 5). Proximal (PIP) and distal (DIP) interphalangeal joints are completely bony ankylosed in the position of flexed proximal and extended distal interphalangeal joint (Figure 6). Radiological features (Figure 7) of IP joints (in frontal and lateral views) comprise intraarticular osseous fusion, joint space



Fig. 5. Dorsal aspect of the hand with ankylosis of the little finger.

loss, osteoporosis particularly on the middle phalanx, absence of signs of lytic lesions and/or new bone formation. Radiograph also reveals no pathological changes of the articular surface for MP joint.

The pattern of the deformity of the little finger, suggests several possible diagnoses: Dupuytren's disease, psoriatic arthropathy, buttonhole deformity, and septic joint (Table 3). Dupuytren's disease is a condition of genetic predisposition but of unclear etiology, and is characterized by progressive digital deformity as a consequence of cord formation in the region of palmar and digital fascia. Disease usually affects individuals over the age of 50 years, and is more common in males, with reported ratios varying between 4:1 and 10:1¹⁹. The PIP joint fixed flexion deformity with MCP joint in flexion indicates severity of the disease and poor prognosis. In the advanced stage of the disease flexion deformity of DIP joint or boutonniere deformity may be observed¹⁹, and long term flexion contracture of PIP joint could lead to bony ankylosis.



Fig. 6. Ankylosed finger: a) lateral aspect, b) palmar aspect.

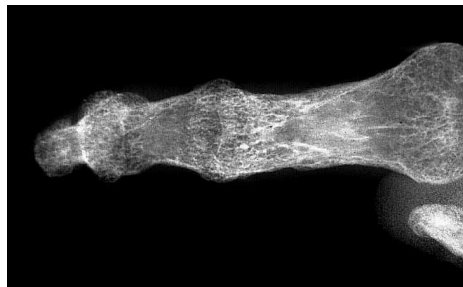


Fig. 7. Radiography of the ankylosed finger (lateral and AP projection).

McFarlane (1974)²⁰ explains the concept that in Dupuytren's disease normal anatomical fascia and sheaths contract into pathological cords that could be classified into several categories: pretendinous, central, spiral, natatory, lateral, and retrovascular cord (Figure 8). The

TABLE 3
 OUTSTANDING FEATURES ENABLING TO DIFFERENTIATE ADVANCED STAGES
 OF FIVE PATHOLOGICAL CONDITIONS THAT COULD BE ASSOCIATED WITH DEFORMITY
 OF INDIVIDUAL FROM GRAVE NO. 2

Dupuytren's disease	Psoriatic arthropathy	Buttonhole deformity	Septic joint	Rheumatoid arthritis
– Flexion deformity of the MCP and PIP joint	– Asymmetric distribution – Involvement of IP joints – Usually involved more than one finger – Marginal bony erosions with adjacent proliferation – Intraarticular osseous fusion – Destruction of phalangeas tufts – Pencil-an-cup appearance on radiography	– DIP hyperextension and – PIP flexion deformity.	– Marginal and central bone erosions – Signs of osteomyelitis on adjacent bones – Fibrous and bony ankylosis – Osteoporosis	– Symmetric involvement of MCP and PIP joints – Cystic lesions of the phalanx – Bony erosions – Fibrous ankylosis and occasional intra-articular osseous fusion of IP joints – Osteoporosis – Mallet finger, swan-neck and boutonniere, and other characteristic deformities

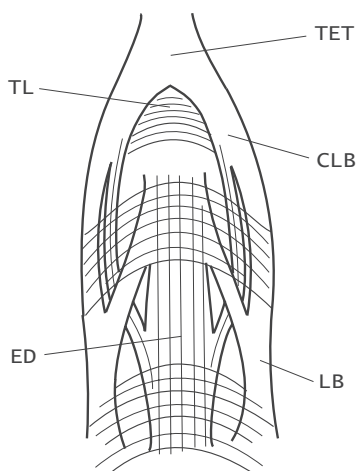


Fig. 8. Scheme of pathologic cords in Dupuytren's disease: PC – pretendinous, CC – central, SC – spiral, LC – lateral. (Adopted from McFarlane, RM.: 1974. Patterns of the diseased fasciae in the fingers in Dupuytren's contracture. *Plast. Reconstr. Surg.* 54:31–35).

majority of them are the extensions of the palmar fascia which terminate superficially in the skin distal to the metacarpophalangeal (MP) joint, and are connected with flexion deformity of both MP and PIP joints. However, the lateral cords, which are adherent to the skin and flexor sheath at the area of the PIP joint, and isolated fascial cords within digits without proximal palmar connections²¹ are associated with significant PIP joint deformity, as it was in our case.

Buttonhole (Boutonniere) deformity is characterized by DIP hyperextension and PIP flexion. It is usually results from untreated laceration of the central slip of extensor digitorum communis on the dorsal aspect of PIP joint (Figure 9), although other forms of this deformity could accompany dorsal burns of hand with flexion contracture and psoriatic arthritis. In the buttonhole deformity, flexi-

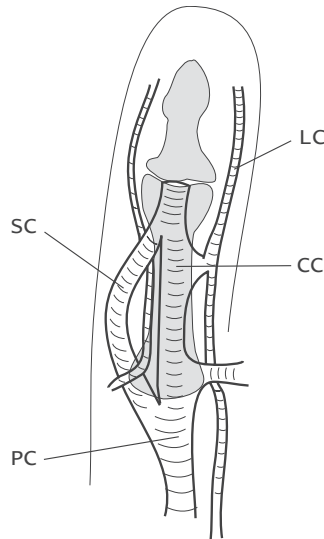


Fig. 9. Scheme of the complex extensor mechanism of the finger:

As the extensor digitorum (ED) tendon reaches the dorsal aspect of proximal phalanx, it begins its trifurcation into central slip (which ends by insertion on the base of the middle phalanx), and two lateral slips. At the level of PIP joint, tendinous fibers of interosseal and lumbrical muscle form two lateral bands, which exchange the fibers with central and lateral slips of ED tendon, forming two conjoined lateral bands (CLB); these bands merge over the dorsum of the middle phalanx to form terminal extensor tendon (TET), which inserts into dorsal base of the distal phalanx. The central and lateral bands are anchored to each other and to the adjacent skeletal elements by groups of fibers: the oblique cord, extending from the palmar aspect of proximal phalanx to the lateral band; the triangular ligament (TL) composed of transverse fibers between lateral bands and central slip insertion. Distal to the zone of convergence of tendons (at the level of PIP) the central slip and conjoined lateral bands represent dual extensor mechanism with both intrinsing and extrinsing muscles involved in IP extension.

on of the middle phalanx due to unopposed superficial flexor digitorum produces herniation of the condyles of the proximal phalanx through the central slip²². As the deformity progresses, the conjoined lateral bands slide to the axis of PIP joint and allow tightening of the terminal extensor tendon followed by DIP hyperextension. Ankylosis with significant degree of flexion deformity in PIP joint, as it was found in our case, as well as absence of hyperextension in DIP joint are not indicative for diagnosis of the buttonhole deformity. In regard with skeletal distri-

bution and morphological features seen on radiography, psoriatic and septic arthritis (Table 3) are less probable diagnoses.

In conclusion, individual from grave No. 2 probably suffered from Dupuytren's disease with long term flexion contracture of PIP joint that resulted in bony ankylosis. Even to date, no surgical treatment has been effective enough in the management of Dupuytren's disease, so we could expect to recognize similar pathologies in osteoarchaeological material.

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DVA SLUČAJA BOLESTI ZGLOBOVA S POST-SREDNJOVJEKOVNOG GROBLJA SVETI ILIJA

S A Ž E T A K

U radu su analizirani skeletni ostaci 11 osoba s groblja oko crkve Svetog Ilije u Srbiji, datiranog u kasni srednji vijek kako bi se istražila prisutnost bolesti. Dvije osobe imale su patološke promjene koje su zahvatile zglobove, a jedna od njih najvjerojatnije je patila od Legg-Calvé-Perthesove bolesti. Izgleda da bolest nije bila liječena sa znatnim remodeliranjem same kosti, a deformiranost glave femura i acetabuluma uzrokovalo je sekundarne degenerativne promjene na zglobovima u relativno ranoj životnoj dobi ove individue. Drugi slučaj odnosi se na koštanu ankilozu prsta ruke vjerojatno uzrokovanu Dupuytrenovom bolešću. Ujedno su raspravljane i moguće diferencijalne dijagnoze oba patološka stanja.