

## Case Report

# Histological diagnosis of Bechterew's disease on exhumed female remains in Bosnia and Herzegovina: a case report

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## ABSTRACT

Ankylosing spondylitis is a serious ailment that affects people, and the first signs or symptoms usually occur between the ages of 15 and 45. While the condition is mostly prevalent in men, women are not immune to this disease. This problem is diagnosed with a combination of clinical history and X-rays, pathology and HLAB27 test. The aim of this case study is to demonstrate how macroscopic and microscopic analysis can be used for identification of the disease from a forensic point of view. In April 2018, we exhumed 11 remains near the city Višegrad, twenty-five years after the last war. All the remains were completely skeletonized. The skeleton of a female was specific and shaped like a bamboo branch, with a partial knitting of vertebral bodies in the lumbar region of spine and with total knitting in the thoracic part. The spinous processes were completely knitted. Her son gave informations for verbal autopsy that she had trouble walking and doing normal activities during life. Samples for analysis and pathological diagnostics were used to determine the real bone condition for forensic purposes. To our best knowledge our case is first one in the literature which combines macroscopic and microscopic analysis of AK in exhumed skeletal remains after 25 years of death in modern era of Europe.

**Keywords:** Bechterew's disease, Exhumation, Pathology, Identification

## INTRODUCTION

Ankylosing spondylitis (AS) is a chronic progressive inflammatory disease of the intervertebral joints which begins in the sacroiliac region and most often affects the lumbar and thoracic parts of the vertebral column. Pathological process first causes inflammation of the ligaments and joints around the spine with formation of syndesmophytes. Finally, fusion of vertebral bodies occurs. The disease usually begins in the second or third decade of life.<sup>1-5</sup>

There is large geographic variation in the prevalence of disease, being highest in North America, followed by Europe, Asia, Latin America, and Africa, respectively.<sup>6</sup> Although AS is nearly twice as common in men than

women, more recent studies suggest a more equal distribution among sexes.<sup>7</sup>

The diagnosis of AS is based on criteria determined by the Assessment of Spondyloarthritis International Society (ASAS) in 2009, which include the genetic biomarkers, response to non-steroid anti-inflammatory drugs, radiologic changes, and clinical presentation together with consideration of associated conditions.<sup>8</sup>

## CASE REPORT

In April 2018, we exhumed 11 remains near the city Višegrad, twenty-five years after the last war in Bosnia and Herzegovina. All the remains were completely skeletonized. A macroscopic examination was carried out,

with the analysis of the palaeopathological conditions of the remains. The skeleton of a 70-year-old female was specific, shaped like a bamboo stick, with a partial knitting of vertebral bodies in the lumbar region and with total knitting in the thoracic part of the spine. The spinous processes were completely knitted. Ribs showed degenerative changes. Her son gave information for verbal autopsy that she had trouble walking and doing normal activities during life. The specific appearance raised suspicion for ankylosing spondylitis. We sampled material for histopathology and found signs of osteoporosis, together with the formation of new non-mineralized bone with fibroconnective tissue in the lumbar part of the vertebral spine. Using trichrome histochemical staining we confirmed collagen transformation into osteoid.



**Figure 1:** After the exhumation, a complex skeleton and an autopsy were performed.



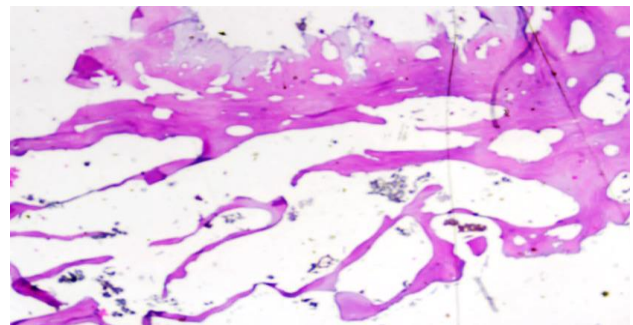
**Figure 2:** Coalescence and fusion of vertebral bodies, the spinous processes and kyphosis in thoracic part.



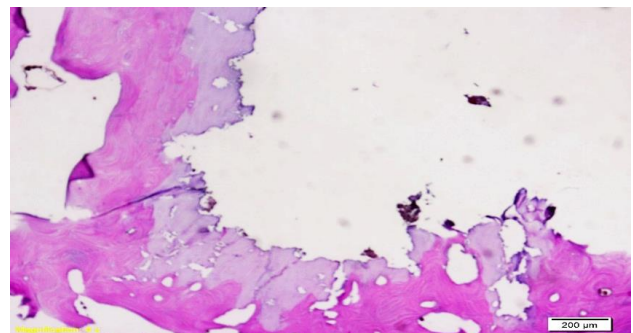
**Figure 3:** Degenerative changes in the ribs.



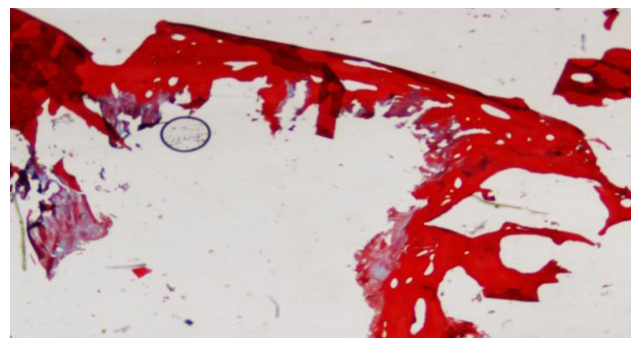
**Figure 4:** Macroscopic bone structure in Bechterew's disease.



**Figure 5:** Bechterew's disease; lumbar part. Osteoporosis, mineralized mature bone and also the new non-mineralized bone with fibroconnective tissue (HE, x20).



**Figure 6:** The new non-mineralized bone with fibroconnective tissue in lumbar part of spine (HE, x40).



**Figure 7:** Collagen production in Bechterew's disease (Trichrome, x20).

The aim of this case study was to demonstrate that macroscopic and microscopic analysis can be used for identification of AK on exhumed remains even more than 25 years after death.



**Figure 8: Collagen transformation into osteoid (Trichrome, x40).**

## DISCUSSION

Bone paleopathology is very important section in Bosnia and Herzegovina after the 1992-1995 war, because almost 7,000 people have still not been exhumed to this day. A lot of time has passed, so sometimes there are no more living relatives. The process, however, is very complex and involves the application of advanced forensic technology.

Although data from recent studies suggest that AS was found on the remains of mummies from ancient Egypt, there is little data regarding the occurrence of AK on human remains in modern era.<sup>9</sup> One of the cases described in Europe is from Slovakia from remains of European male, whose remains are dating from early modern times.<sup>10</sup>

In our case, the macroscopic analysis was very convincing for ankylosing spondylitis in addition of positive verbal autopsy from her son. In living human, histology of AK considers the inflammation of entheses of the vertebral bodies and the sacroiliac joint as the first sites of inflammation development. In entheses occurs remodelling of the bone with formation of new bone, which forms bony protuberances, known as syndesmophytes, in the lumbar, thoracic, and cervical spine.<sup>11-15</sup> The pathohistological analysis in our case is affirmative although the details (cellular elements) are not preserved, which is obvious after many years.

## CONCLUSION

To our best knowledge our case is first one in the literature which combines macroscopic and microscopic analysis of AK in exhumed skeletal remains after 25 years of death in modern era of Europe.

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