



Research Article

Case Report: Chronic Maxillary Sinusitis, Osteoarthritis, Ankylosing Spondylitis, Vastus Notch and Sacral Hiatus

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Abstract

This case report provides an analysis and description of the skeleton of a 74-year-old Mexican male from the perspective of skeletal anthropology, osteology and osteopathology. The examined skeleton is part of the contemporary osteological collection of the Department of Innovation in Human Biological Material (DIMBIH) of the Faculty of Medicine of the Universidad Nacional Autónoma de México (UNAM) located in Ciudad Universitaria. In this individual, we identified the presence of chronic maxillary sinusitis, osteoarthritis, ankylosing spondylitis, and two anatomical variants. In order to evaluate and determine each disease, a morphometric and morphological analysis of the osteopathological structures was performed macroscopically and microscopically. This case is relevant for the opportunity it provides to investigate different diseases and anatomical variants, as well as for the information it provides. Studies such as this one provide physical anthropologists with new avenues for practical applications, such as documenting, analyzing and interpreting health-disease processes, etiology, ailments, and even treatments when there is access to the clinical history of individuals of contemporary populations, allowing for comparisons with osteological evidence through bone remains.

Keywords: Osteology; Osteopathology; Physical Anthropology; Sinusitis; Osteoarthritis; Ankylosing Spondylitis; Anatomical Variants

Introduction

According to Goodman and Bogin, the study of the biocultural adaptation of prehistoric, historical, and contemporary human populations allows us to delve into the impact of environmental interactions on human biology through indicators of adaptability, health-disease, and quality of life [1]. Thus, physical anthropology provides theoretical and methodological elements to approach the study of our species and incorporates the analysis of the evolutionary, ecological, or biocultural aspects intending to generate knowledge about the biology, origins, and

variability of past and present human populations. The knowledge acquired from the studies of the new physical anthropology is applied to very diverse fields such as public health, nutrition, sport and physical activity, and epidemiology [2].

With these theoretical and methodological tools, it is possible to analyze general and particular cases from skeletal remains and thereby interpret conditions and histories of life and disease in this specific case. The etiology, symptomatology, and consequences that the diseases generate in individuals must be detailed. In the following study, specific information is provided about chronic maxillary sinusitis, osteoarthritis, ankylosing spondylitis, diseases detected in the individual studied, as well as the two anatomical variants that were also recognized from the osteological analysis performed.

Chronic maxillary sinusitis (CMS)

Definition: Sinusitis is the acute or chronic inflammation of the paranasal sinuses. When sinusitis keeps recurring or continues for a prolonged period, it is considered chronic [3]. Anatomically, the paranasal sinuses are air-filled spaces in the bones of the face and skull, communicating with the nasal cavity through openings known as “ostia.” There are four sinuses: maxillary (also known as Highmore’s antrum), frontal, ethmoidal, and sphenoidal. However, the maxillary and frontal sinuses are the ones frequently disrupted by sinusitis.

Maxillary sinusitis becomes chronic because a bacterial infection may prevail. In addition, three-quarters of all chronic infections are caused mainly by three organisms: *Streptococcus pneumoniae*, *Haemophilus influenzae*, and *Moraxella catarrhalis* [4]. At the same time, chronic sinusitis is a common contemporary condition and accounts for substantial morbidity, especially in areas with heavy air pollution [5]. Chronic sinusitis has been diagnosed with higher prevalence in individuals living in urban areas. This condition can appear at any age, and allergies can cause chronic inflammation and even damage the bone structure.

Osteoarthritis (OA)

Definition: Osteoarthritis (OA) is a chronic, degenerative disease with a multifactorial etiology. The gradual loss of articular cartilage characterizes it and is the most common cause of pain and disability in older adults (Peña et al. 2007) [6]. Age, genetics, sex, and excessive use of repeated movements are factors that influence the disease. In advanced OA, the cartilage begins to degrade, and the bone surface erodes, causing the release of collagen and proteoglycan fragments into the joint cavity. This release causes an inflammatory response in the synovial membrane leading to the production of inflammatory cytokines; thus, the articular region of the bone responds with overproduction in an attempt to repair.

Conversely, genes are the most frequent risk factor for OA in the general population. Genetic predisposition has been demonstrated through heterozygous twin studies, which established the presence of a considerable amount of the variation observed in the development of OA [7].

Diagnostic data for the pathology are observable from macroscopic bone alterations, including periarticular labiation or osteophytosis, characterized by the formation of new bone around the joint margins. Osteophytes are characterized by new bone formation on the joint surface due to vascularization of the subchondral bone. They may occur in isolation at the joint margin or on the joint surface. Porosity in the form of pitting is also observed on the joint surface. Finally, eburnation may be observed, characterized by a highly polished area on the articular surface of the bones [2].

Eburnation is easily observable on macroscopic analysis because, in severe osteoarthritis, the bone rubs against the other bone, causing a shiny surface [8].

Ankylosing Spondylitis (AS)

Definition: Ankylosing spondylitis (AS) is an arthropathy characterized by chronic inflammation of the axial skeleton, which causes progressive back pain, inflammation, and stiffness [9].

Ankylosing spondylitis (AS) is a systemic, chronic inflammatory disease that is classified as a seronegative spondyloarthropathy [10]. The disease has had many eponyms in the past, such as Bechterew’s syndrome, Marie-Strumpell spondylitis, rheumatoid spondylitis, and Von Bechterew’s disease; however, ankylosing spondylitis (AS) has become the most common term. OA was apparently first described by the Irish physician Bernard Connor (1666-1698), who reported an unusual skeleton found in a cemetery near where he taught anatomy in France [11].

Ankylosing spondylitis generates vertebral alterations called syndesmophytes, i.e., they are thin vertical exostoses and represent the calcification of the annulus fibrosus. Changes due to spondylitis and osteoarthritis predominate in the cervical and lumbar segment due to overloading at these levels. Syndesmophytes predominate in the anterior and lateral sides of the spine [12]. As syndesmophytes grow, ossification may affect the longitudinal ligament. In advanced stages of AS, extensive syndesmophytes produce an undulating vertebral contour called a “bamboo spine.” Osteophytes develop by disc displacement, causing traction on the insertion sites of the longitudinal ligaments. These elements have a horizontal orientation, unlike those produced in AS, which are vertical [13].

The disease may occur more frequently in the third decade of life, and men are affected two to three times more often than women. Approximately 90% of patients have one or other of the HLA-B27 subtypes. The prevalence of AD in Europe is about 1%; however, there are considerable variations in other populations, for example, in the Canadian Haida Indians who have a prevalence of 4.5%, and half of that population is antigen-positive [14]. The genetic factor is preponderant in AD, as most patients express the HLA B27 gene, which helps in the differential diagnosis [15].

When the disease advances, the vertebrae become stiff, immobile, and deformed, and flexion is limited; various neurological complications may also appear. The most frequent conditions are vertebral fractures with minimal trauma [16].

It is relevant to mention that ankylosing spondylitis (AS) is part of the seronegative inflammatory arthropathies that affect the lumbar spine, along with psoriatic arthritis, reactive arthritis, and arthritis accompanying inflammatory bowel disease. Other

systems are frequently affected, causing uveitis, mucocutaneous lesions, other peripheral localizations of arthritis, cardiac and digestive involvement, and enthesopathies with enthesophytes formation [17]. This condition results from the combination of genes, repetitive biomechanical stress, and an immune response to bacterial products leading to inflammatory changes at specific anatomical sites and joints [18].

The AS is characterized by sacroiliitis, which refers to the inflammation of one or both sacroiliac joints; the symptoms are a pain in the buttocks or lower back that may extend to one or both legs and characterize by the constant occurrence of pain in the lower back area that radiates to the buttocks and sacroiliac region. Because of the previously mentioned, a usual picture of morning stiffness occurs, and a limitation of mobility develops over time. Symptoms of sacroiliitis may improve with exercise.

Men are affected more often than women. It should be noted that sometimes the condition appears in young adults [19]. When sacroiliitis progresses, and AS is left untreated, it can lead to a functional disability, as bilateral ossification of the sacroiliac joints significantly limits movement. Correlation studies have found that the deterioration of functional capacity in patients with AS may be due to the lack of strict control of the disease, regardless of the time of evolution, lack of treatment, and in addition, a lack of attention to the depression and fatigue generated [20].

Anatomical variables

Definition: These are singularities in the morphological arrangement of anatomical structures that do not have a pathological cause nor represent a danger to the person with them. In the following, only those that concern the case in question will be mentioned, i.e., vastus notch and sacral hiatus.

The Vastus notch (VM)

Definition: It is an anatomical feature characterized by a concavity in the superolateral angle of the patella, where the tendon of the vastus lateralis muscle inserts, and is called a non-metric variant. In order to distinguish it, an indentation or depression in the superolateral or superomedial corner of the patella must be identified [21]. The MV was named and described by Finnegan (1978) in his studies of the Terry Collection (USA). It is worth mentioning that this anatomical variable is also referred to as the “emarginate patella.”

Sacral Hiatus (HS)

Definition: It is an opening at the lower end of the sacrum, i.e., an incomplete fusion of one or more neural arches in the midline. It may present with vertebral hypoplasia, hemivertebral, laminar, pedicular fusion, scoliosis, and kyphosis. All extensions of the sacral hiatus are conditioned by an arrest in the fusion process

of the two lateral segments of the vertebral arch [22]. These congenital anomalies are anatomical variables, also called non-metric traits or characters, that have been described since antiquity and are produced by pathological (or not) modifications in the development during intrauterine life. In most cases, these non-metric variants have no effects, although there may be clinical symptoms such as low back pain or genetic conditions linked to the presence of a non-metric trait.}

Based on the previous information, it was possible to evaluate the bone remains of the individual, and this will allow us to present the differential diagnosis made from the general pathological evidence of the skeleton, as well as the interpretation of the following evidence: chronic maxillary sinusitis with inflamed Pott’s tumor, accompanied by osteoarthritis, possibly associated with ankylosing spondylitis and two anatomical variants (Table 1).

Material and Methods

In the study of anthropological osteopathology, there is a descriptive system for analyzing bone conditions. There are essential elements that must be established, such as (1) an unambiguous terminology, (2) precise identification of the location and distribution of the damaged bone, and (3) a descriptive summary of the morphology of the damaged bone. Therefore, this work contemplated these elements, presenting a descriptive summary of the altered or damaged morphology [23].

In the present work, a description and analysis of the skeleton of a 74-year-old Mexican male who presents: Chronic Maxillary Sinusitis (CMS), Ankylosing Spondylitis (AS), Advanced Osteoarthritis (AO), and two anatomical variants. In order to evaluate and determine these conditions, a morphological analysis -macroscopic and microscopic- of the bone structures was performed, showing pathological conditions. The individual comes from the contemporary osteological collection of the Department of Innovation in Human Biological Material (DIMBIH) at the Faculty of Medicine of the Universidad Nacional Autónoma de México (UNAM), located in Ciudad Universitaria, Mexico City.

The osteological collection of the DIMBIH is made up of the bodies of individuals who, in life, decided to donate them through the UNAM’s Body Donation Program and others that are in safekeeping through the Ministry of Health. After the death of the individuals, the bodies received embalming treatment in order to be used for teaching and research. It is worth mentioning that some of them, once the objective is achieved, are destined to be part of the contemporary osteological collection, and for this purpose, they must undergo osteotechnics. A body can only be part of the contemporary osteological collection as long as it has been previously authorized by the Secretaría de Salud based on Art: 347, 329, and 350 Bis 3 of the General Health Law.

The case of the man reported here consists of a complete skeleton with its respective death certificate and death record; it does not have a clinical history even though the hospital of origin was consulted.

The following is a description of the information obtained from the osteological analysis and the aforementioned documents. The documents were cross-checked to correlate the skeletal lesions observed during the anthropological analysis and the documentary information.

Materials and Equipment

A registration form was used to document demographic, morphological data (macroscopic and microscopic), and anthropometric measurements. Nikon D3300 camera, GPM vernier or slide compass and Leica S9i microscope.

Analysis

Documentary analysis

A search was made in the archives of the Department of Innovation in Human Biological Material to obtain the documentation corresponding to the entry of the body to the department, i.e.: death certificate and death record, birth certificate, the official letter from the director of the hospital of origin which allows storing the body and the permission issued by the Secretaría de Salud to authorize the DIMBIH as a depository.

Anthropological Analysis

In the present case, a thorough anthropological and osteopathological analysis was performed, emphasizing the

morphoscopic and macroscopic observation of the bone tissue. The macroscopic analysis was performed according to the disease and the bone element affected, namely:

Chronic Maxillary Sinusitis

Skull. It was identified that chronic maxillary sinusitis generated the following characteristics in the skull: bone destruction of the right maxillary sinus, just in the central part of the structure. The perforation has the following dimensions: 145 mm in diameter, with thinning of the bony structure in all its contour as a consequence of an advanced and prolonged infectious process, as indicated by the characteristics of the lesion. In the left maxillary sinus, there is also a thinning of the anterior wall without perforation.

In the transillumination test, the bony tissue's thinning was observed in the roof, floor, and walls of both orbits, as well as in both maxillary sinuses, being more pronounced on the right side (See Figure 1 B and C).

Osteomyelitis, sometimes called Pott's inflamed tumor, was observed in the frontal bone (See Figure 1 A). The chronic maxillary sinusitis directly impacted the frontal area, causing the osteomyelitis that manifested with the formation of Pott's inflamed tumor or also called Pott's puffy tumor, as this can be caused by *Streptococcus pneumoniae*, a microorganism present in prolonged bacterial infections and creating subperiosteal abscesses [24].

According to Hagedorn (2004), the above characteristics have already been reported in other cases of chronic maxillary sinusitis [25].

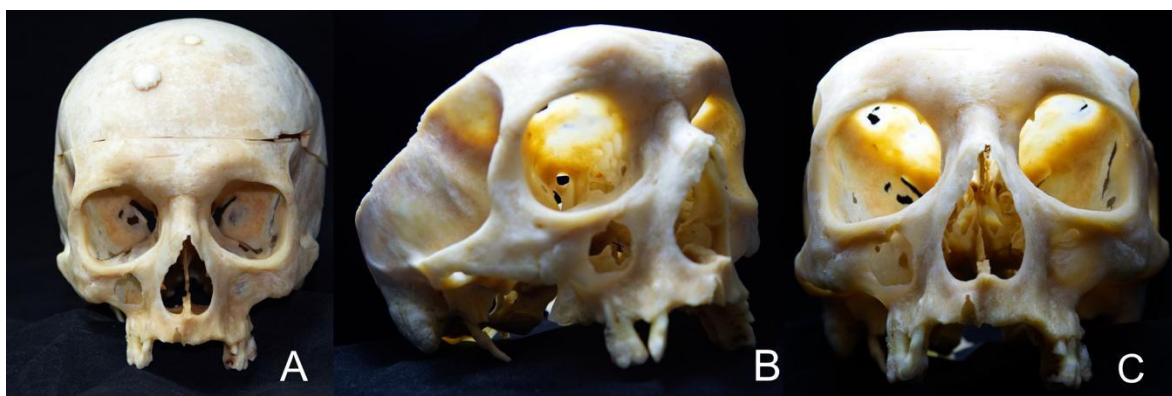


Figure 1: A) Frontal view of the skull showing the lesion in the right maxillary sinus and two osteoid osteomas called “Pott’s puffy tumor.” B) Right frontal, lateral view showing an infectious process with necrosis of 145 mm diameter in the right maxillary sinus. C) Transillumination test to observe the thinning of the bony tissue, identifying a thinning projected towards the four faces of the maxillary sinus.

Osteoarthritis (OA)

The lesions caused by osteoarthritis were located in the following skeletal structures: the atlanto-occipital structure, both scapulae, humerus, radius, ulnae, vertebrae, femurs, patellae, and only on the right side in the tibia, fibula, calcaneus, and talus.

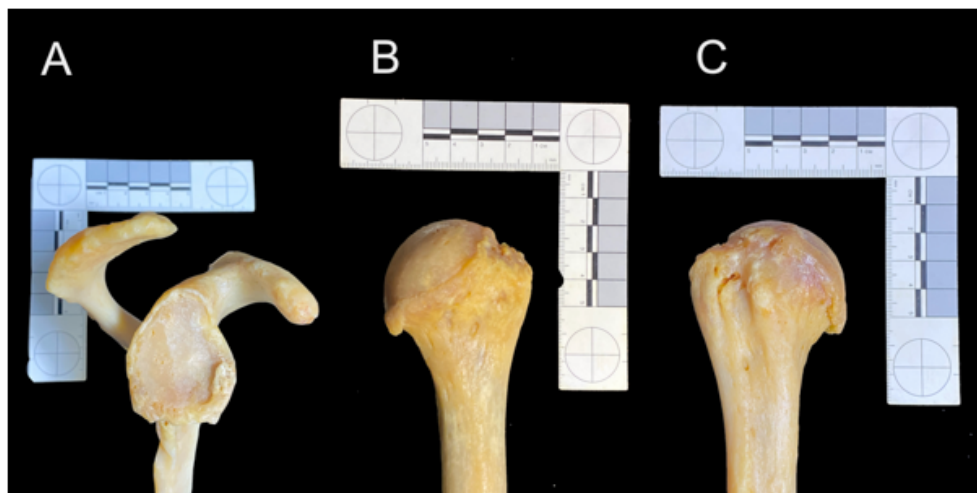


Figure 2: A) Superolateral view of the right scapula with osteophytes at the margins of the glenoid cavity characteristic of OA. B) Anterior view of the upper proximal third of the right humerus affected by OA, formation of marginal osteophytes. C) Posterior view of the upper proximal third affected by OA, formation of marginal osteophytes at the periarticular borders of the humeral head.

Patellae: Normal patellae exhibit smooth and sometimes wavy facets. However, in the studied man, osteophytes with eburation were observed, which are characteristic of OA.

Scapula: In the right scapula, marginal osteophytes are observed around the edge of the glenoid cavity, with a more significant presence in the infraglenoid and supraglenoid tubercles (Figure 2. A).

Humerus: On the right humerus, marginal periarticular osteophytes are shown proliferating around the anatomic neck, anteriorly and posteriorly, as well as on the greater tubercle in the anterior view (Figure 2. B and C).

Radius and Ulna: In the tuberosity of the radius, enthesophytes are observed with marked entheses where the tendon of the biceps

brachii muscle inserts. In contrast, the head of the radius shows mild marginal osteophytes. The right ulna shows erosion and osteophyte formation around the margins of the trochlear notch.

Vertebrae: In the axis (C2), there is a new bone formation or marginal osteophytes in the upper part of the odontoid process and localized eburation in the anterior articular cartilage, a characteristic of osteoarthritis (Figure 3. A). Marginal osteophytes were also located in the C2 vertebrae, in the facet and inferior articular process of C3, and in C4, there is an asymmetry on the right side. The most affected structures at the cervical level are C2, C3, C4, and C5, and at lumbar level L4 - L5 (Figure 3. D and C). Osteophytes can form syndesmophytes, as seen in Figure 3. B and E, and their presence indicates instability. Syndesmophytes present at T6 - T7 and T8.

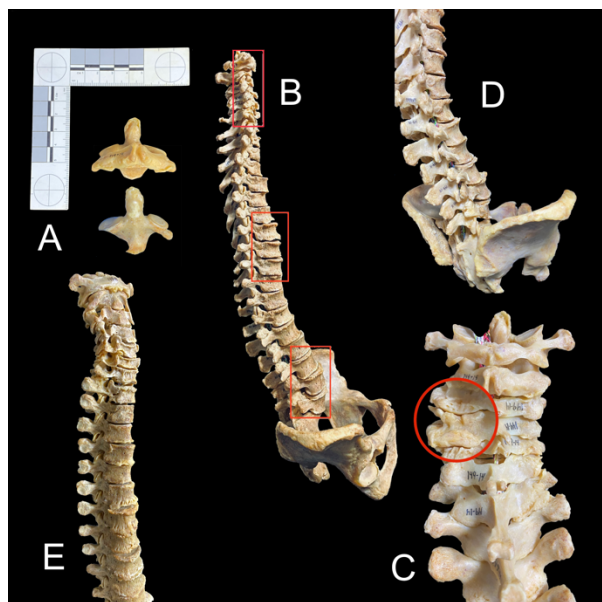


Figure 3: A) Axis (CII), the upper part of the odontoid processes, with the presence of eburnation. B) Lateral view of the spine; the most affected areas are shown in red. The most affected levels at the cervical level are C2 - C3 - C4 and C5, and at the lumbar level are L4 - L5. Osteophytes can form syndesmophytes, as seen in T6 - T7 and T8. C) Close-up of the formation of marginal osteophytes in the cervical area. Marginal osteophytes were located in the C2 vertebrae, in the facet and inferior articular process of C3, C4, asymmetrical right side. D) Posterolateral view of the lumbar area of the spine, the formation of the slight bamboo-spine shape characteristic of AS can be seen, which is due to the waistband formed in the vertebral body and the protruding osteophytes edges of the articular areas of the intervertebral discs. E) Anterolateral view of the cervical and thoracic areas of the spine showing the formation of osteophytes and syndesmophytes.

Ankylosing Spondylitis (AS)

Pelvis: Both iliac bones are fused with the sacrum and show osteophytes (Figures 4 and 5). Bilateral sacrolitis indicates ankylosing spondylitis (AS) or degenerative hypertrophic spondylitis.

Sacrum: The sacrum is completely fused to the iliac bones, i.e., there is a total hip ankylosis (fusion).

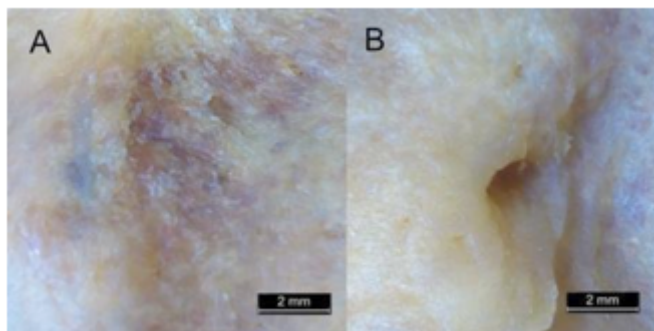


Figure 4: Close-up with Leica Model S9D Microsystems microscope, distance or depth of field of 122 mm, integrated 10 Mp CMOS camera. Total ankylosis with synostosis of the right and left sacroiliac joints are observed.



Figure 5: Symmetric fusion of both sacroiliac joints due to ankylosing spondylitis (AS).



Figure 6: Fusion defect in the lower sacral hiatus between S5 and S3, type II of Testut (1979).

Femur. Enthesophytes, outgrowths, and new bone are present in the rough line as irregular spurs and ridges at muscle attachment sites along the rough line (Figure 7). Enthesophytes, known as traction spurs, reflect tensile stresses and inflammation at ligament and tendon insertions.

Right foot. It presents ankylosis of the ankle and severe fusion of the tibia and fibula with the talus and calcaneus; this frequently occurs in ankylosing spondylitis and spondyloarthropathies (Figure 8). The metatarsals and phalanges are free of involvement.



Figure 7: Right femur with very pronounced enthesophytes in the rough line. Moderate OA with osteophyte formation in the periarticular area. The femur is mainly related to the quadriceps crural muscle since three of its four muscular bodies (crural, vastus internus, and vastus externus) insert into it, making the leg extension movement possible.



Figure 8: Ankylosis of the right ankle, severe fusion of the tibia and fibula with the talus and calcaneus frequently occurs in ankylosing spondylitis.

Anatomical Variants

Sacral hiatus: A fusion defect was found in the lower sacral hiatus between S5 and S3, type II of Testut (1979) [26]. We can confirm the presence of sacral hiatus by ruling out spina bifida, as spina bifida is differentiated by incomplete development of the neural arch elements in one or more vertebrae [27] (Figure 6). The lower sacral hiatus is classified as type II [22].

Vastus notch: The presence of superficial osteophytes or porosity, irregular margins, and marginal osteophytes with a severe degree due to osteoarthritis were identified. The bipartite patella was ruled out as the vastus notch was distinguished from it by having a smooth, nonporous surface.

Results

The records that could be consulted for this man were only the death certificate and death record, from which the age, sex, date of death, and cause of death (right basal pneumonia) were obtained.

A thorough anthropological analysis of the skeleton was carried out through morphoscopic, macroscopic and microscopic

observation of the bony structures, and chronic maxillary sinusitis (CMS), osteoarthritis (OA), ankylosing spondylitis (AS) as well as two anatomical variants were identified: sacral hiatus and vastus notch. After analysis, it was identified that the diagnostic criteria for AS were met [23]. Also, OA was determined to be grade 2 to 3, i.e., moderate to severe.

The following table is based on the macroscopic and microscopic analysis of the individual (Table 1).

Distribution of lesions								
Skull	Scapula	Humerus	Radium and ulna	Vertebrae	Sacrum	Pelvis	Calcaneus and astragalus	Patella
	OA	OA	OA	OA		OA	OA	OA
CMS								
				AS	AS	AS	AS	
CMS: Chronic maxillary sinusitis OA: Osteoarthritis AS: Ankylosing spondylitis								

Table 1: Distribution of altered morphology due to osteopathological lesions.

An infectious process was identified in the bone material, indicating the presence of chronic sinus disease, also called chronic maxillary sinusitis. Two osteoid osteomas due to chronic maxillary sinusitis were observed in the frontal portion of the skull, which caused an intracranial abscess, also called Pott's puffy tumor [24]. Untreated catarrhal processes can lead to chronic sinusitis as inflammation prevents adequate clearance of bacteria in these cavities, increasing the likelihood of developing sinusitis [4].

This case is relevant since advanced osteoarthritis lesions were located in the scapula, humerus, radius, ulna, and all spine segments. The identified vertebral alterations extend along the margin of the intervertebral discs, showing total sacroiliac ossifications, which is one of the typical findings of ankylosing spondylitis (AS). For the detection and characterization of vertebral degenerative changes, in this case, where the main feature is the formation of osteophytes, an adequate localization of the lesion sites was performed.

AS was fully identified (Figure 5), since the sacroiliac joint was fused bilaterally, without extended fusion to the spine. In turn, in this case, the disease progressed, and ankylosis occurred in the right foot, ankylosis of the ankle, and severe fusion of the tibia and fibula with the talus and calcaneus attributed to ankylosing spondylitis (Figure 7).

Discussion

According to Akhaddar (2017) maxillary sinuses can be analyzed macroscopically because their anterior walls are thin, air-filled spaces in the bones of the face and skull, which communicate through openings known as ostia with the nasal cavity. For this reason, the most destructive lesion is usually located in the maxillary sinus since it is the largest paranasal sinus, and its drainage orifice is high on the medial wall, below the middle turbinate. These

anatomical conditions allow infections to advance deeply through the adjacent structures. In the case study, the indicated lesion was evidenced in the right maxillary bone caused by sinusitis.

Regarding the other pathological conditions identified, further radiological and molecular studies are considered necessary to confirm what was observed during the analysis of the evidence. In this case, possible diseases were identified and diagnosed by analyzing dry bones; no soft tissue histology, further history, or records of the individual as a patient are available.

It should be noted that although OA is slightly more common in women than in men, it is clear that the lesions identified in this male individual correspond to this multifactorial pathology that is related to advanced age. Bone loss and degenerative joint changes accumulate with age, making it difficult to differentiate between secondary and age-related osteopenia.

Future work will continue to explore progressive bone fusion that affected the sacroiliac joint, vertebrae, and ribs in ankylosing spondylitis, as it may lead to secondary osteopenia related to immobility, studies have shown vertebral osteopenia in the early stages of the disease, indicating that limited mobility is not the only cause of bone loss in this condition [28,29].

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Author contributions: Julia Stephan Sánchez Torrijos and Lorena Valencia Caballero: concept and research design. Julia Stephan Sánchez Torrijos and Lorena Valencia Caballero: analysis and

interpretation of data: writing of the manuscript. Translation: Brenda Sandoval Meza.

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