



Two cases of osteoid osteoma in skulls dating from the 13–14th centuries from St. Elisabeth's Church in Wrocław, Poland

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ABSTRACT: In the study two human skulls recovered from archaeological excavations at St. Elizabeth's Church in Wrocław, dating from the 13–14th centuries were assessed. Direct measurements of each skull were recorded, and X-ray images in P-A, lateral and basal projections were taken. The skulls represented adult males. Large, bony, lobular tumours were found on the palatine bones of both skulls. X-ray examination identified these tumours as osteoid osteomas, which are benign bone tumours that may originate in the periosteum or may be located inside the bone, distorting the maxilla or mandible. However, osteoid osteoma of the palatine is very rare. This study extends our knowledge regarding the health and diseases of historical populations.

KEY WORDS: osteoid osteoma, skull, tumour of the palatine

Introduction

The skull's examination provides a lot of important information about the biological condition of the examined skeleton (Krenz and Piontek 1996; Gerszten

1998; Janas and Grzesiak-Janias 2005; Kwiatkowska 2005; Layadi et al. 2006; Dąbrowski and Gronkiewicz 2007). Some paleopathological investigations are based only on the skull's examinations (Woo 1931; Abler 1976; Teul et al.

2002; Gawlikowska-Sroka 2006; Gawlikowska et al. 2007; Vidal 2008), but of course the entire image of the health condition is reached when both elements – the appendicular and axial skeleton are examined (Piontek 1999; Kwiatkowska 2005;). Analysis of the skull can be very useful in assessing age, sex, nutritional status, developmental disorders and odontological diseases (Plenk and Gardner 1954; Gerszten 1998; Janas and Grzesiak-Janas 2005; Layadi et al. 2006; Dąbrowski and Gronkiewicz 2007). Analyzing bony remnants from different historical populations allows for the distinction between the structure of a disease and the level of a population's development. Using excavated material, we can investigate the history of disease development. Paleopathology, which describes the appearance, intensity and course of diseases that were prevalent in the past, allows for the prediction of the course of these diseases in the future, and can be useful for disease prevention and control (Plenk and Gardner 1954; Piontek 1999; Szydlowski et al. 2005; Layadi et al. 2006; Szostek et al. 2014). The most common human pathologies that have been described in excavated material include: posttraumatic changes, degeneration, intentional deformities of bones or teeth, inflammatory lesions, systemic diseases (i.e. rickets, anaemia, nutrient stress), bone necrosis, ailments of the masticatory apparatus, tumours and tumour-like lesions (Crawford 1973; Gładkowska-Rzeczycka 1978; Buraczewski 1987; Piontek 1999; Szydlowski et al. 2005; Gawlikowska et al. 2007; Tomczyk, Tomczyk-Gruza and Zalewska 2012; Gawlikowska-Sroka et al. 2013). Osteoid osteoma is a benign osteoblastic tumour first described by Bergstrand (1935). It can occur anywhere, but in 80–90% of

cases the pathological process affects the long bones. In the skull it is a rather rare process (Moon et al. 2006) which involves the temporal or frontal bone. The calvaria or facial part of the skull are not common locations (Moon et al. 2006). It can appear as a single change, or several bones can be involved. The lower extremities (especially the femur and tibia) and the vertebral column are the most common sites of osteoid osteomas (Barei et al. 1999; Sproule, Khan and Fogarty 2004). Osteoid osteoma is a common bone tumour which can affect a wide range of individuals, from newborns to 70 years (Moon et al. 2006). According to Barei et al. (1999), it is most common in people of about 20 years old. Men are affected more frequently than women (Barei et al. 1999; Moon et al. 2006). No ethnic predisposition is noted for osteoid osteoma. Osteoid osteomas are well-margined lesions with an osteoblastic nidus and a distinct surrounding zone of reactive bone sclerosis (Garrigosa, Ledesma and Servat 2013). The tumour does not invade adjacent tissues and has low growth potential. No malignant transformation has been reported (Grayeli, Redondo and Osterkers 1998). Generally, diagnosis is based on radiological methods and a characteristic appearance, i.e. the presence of a central nidus surrounded by dense sclerotic bone in radiological scans. All radiological methods can be used – radiographs, magnetic resonance imaging (MRI) or computed tomography (CT). CT is more accurate than MRI because it precisely delineates the nidus. Sans et al. (1999) reported that CT helped in confirming the diagnosis of osteoid osteoma in 74% of cases. Modern clinical research revealed that osteoid osteoma has no characteristic symptoms. Signs are instead size-related, and the

enlarged structure can press surrounding structures and provoke pain, which generally proves responsive to salicylates. When diagnosing osteoid osteomas, osteomyelitis, especially Brodie abscesses, eosinophilic granulomas, osteoblastoma, and other benign cysts should be considered (Moon et al. 2006).

Osteoid osteoma in the skull is rather rare, especially in the facial region (Moon et al. 2006), and in our research we present two cases of osteoid osteoma in a medieval population from the city of Wrocław.

Materials and methods

Two human skulls attached to skeletons recovered from an archaeological excavation at St. Elizabeth's Church in Wrocław were examined. The church was originally built in the Roman style in the first half of the 13th c. and rebuilt in the Gothic style after subsequent city fires in the 14th c. The church operated a parish school and later a lower secondary school. Because of this, and numerous precious gravestones found on the church site, we can suppose that this was a burial place for wealthy residents of Wrocław. In 1975 and 1976 two fires almost completely destroyed the church. In 1977 archaeological investigations associated with the reconstruction of the church revealed many graves with skeletons. Excavated skeletons were dated to the 13–14th centuries (Lasota and Piekalski 1986; Lasota 1996; Kwiatkowska 2005). This material is currently kept in the collection of the Polish Academy of Sciences in Poland, Anthropology Unit in Wrocław.

The sex of individuals was assessed based on skull and hip bone morphology, applying standards that are common-

ly accepted and used in anthropology (Acsádi and Nemeskéri 1970; Buikstra and Ubelaker 1994; Piontek 1999). The age of each individual was determined based on the obliteration of cranial sutures, the degree of tooth crown wear, and changes in the morphology of the acetabulum and the surface of the pubic symphysis (Buikstra and Ubelaker 1994; Dąbrowski and Gronkiewicz 1997; Piontek 1999; Kautzenberg and Saunders 2008; Latham and Finnegan 2010). Direct measurements of the skull were recorded, and X-ray images in P-A, lateral and basal projections were taken.

Results

The excavated material was well preserved. The examined skulls were from males, of an estimated age-at-death of 40–55 years. The skulls lacked a mandible; their state of preservation was described as *cranium*. Both skulls had large, bony, lobular smooth tumours on the palatine process of the maxilla. In skull no. SK1, the tumour size was 25×17×7 mm (Fig. 1), and in skull no. 11A, the tumour size was 32×18×12 mm (Fig. 1). X-ray examination revealed a radiolucent nidus surrounded by a reactive sclero-

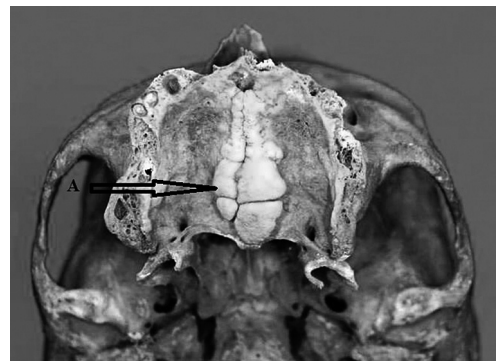


Fig. 1. Male skull No 11 A. A – bony tumour on the palatine (A). Scale 1:2,5.

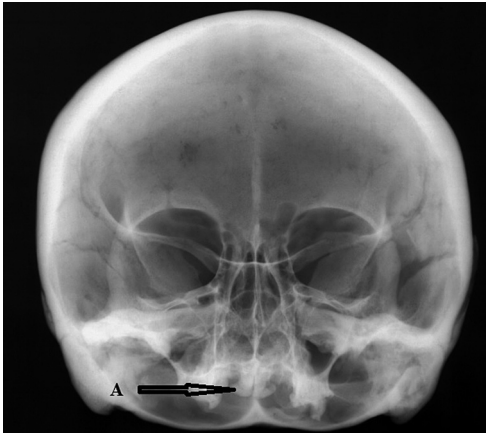


Fig. 2. X-ray photo of male skull No SK 1 in P-A projection presents bony structure of tumour which is described as a osteoid osteoma (A).

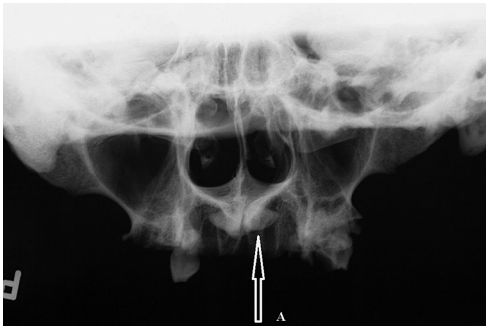


Fig. 3. X-ray photo of male skull No 11 A in P-A projection presents bony structure of tumour which is described as a osteoid osteoma (A).

sis in both cases of skulls (Fig. 2 and 3). This determined that the bone irregularities were osteoid osteoma.

Discussion

Many physicians and historians of medicine have studied when tumours first occur in the human population (Gerszten 1998; Ortner 2011; Ahmadi, Ahmadi and Dehghan 2014). Paleopathology plays an important role in establishing the origin, evolution and spread of tumours through time and in populations (Gładkowska-

-Rzeczycka 1978; Gładkowska-Rzeczycka, Prejzner and Iwanek 1993; Gerszten 1998; Layadi et al. 2006; Kozłowski 2012; Gawlikowska-Sroka et al. 2013). The oldest known description of tumours was found in two medical papyruses, specifically the E. Smith and Ebers papyrus, which date from 3,000 years BCE (Nicolaidis 2013). The oldest tumour found in archaeological specimens was seen in the remains of *Homo erectus* from the middle of the Pleistocene, and was a posttraumatic osteoma. Difficulties with identification and classification are two of the reasons for the lack of a clear history of tumours. This incomplete historical record of tumour development and spread is also due to the age at which tumours typically develop. Tumours usually form after 50 years of age, and become more frequent in older individuals. The average lifespan of historical populations was shorter than that of today. This may be a reason for the low number of tumours identified in historical specimens (Gładkowska-Rzeczycka 1978; Gładkowska-Rzeczycka et al. 1993; Layadi et al. 2006; Moon et al 2006; Marwah et al 2008; Kozłowski 2012). Only some tumours appear in younger people, such as osteoid osteoma, which classically develops at about 20–30 years of age. In our case, both skulls were from adults of approximately 50 years old.

The use of radiological methods for paleopathological studies allows for the assessment of the internal bone structure, as well as the examination of the structure of bony exostoses (Gładkowska-Rzeczycka, Prejzner and Iwanek 1993; Gerszten 1998; Bloch and Silcox 2006; Layadi et al. 2006; Gawlikowska et al. 2007; Marwah et al. 2008; Vidal 2008; Gawlikowska-Sroka et al. 2013). In the current report, the tumours were diag-

nosed as osteoid osteomas on the basis of radiographic evaluation. It is the most common method for the identification of this type of tumour, not only in the analysis of historical material, but also in contemporary clinical cases. Osteoid osteoma has a characteristic radiological image – a translucent central area surrounded by a larger area of dense reactive bone. Such an image was observed in our cases. Of course, the final diagnosis in the case of patients is confirmed by histological examination, but in the case of anthropological excavated material it should be discussed because preparing histological specimens requires damaging the skull. It is too valuable a material for such confirmation.

Osteoid osteoma is a benign bone tumour that is seen more often in the vertebrae and long bones than the skull (Sayan et al. 2002; Janas and Grzesiak-Janias 2005; Jones 2006; Layadi et al. 2006; Kashkouli et al. 2008; Marwah et al. 2008; Kozłowski 2012; Strong, Tate and Borys 2012; Ahmadi, Ahmadi and Dehghan 2014; Singh, Srivastava and Singh 2015), so our findings are interesting not only for paleoanthropologists but also for the medical doctors. Today, it accounts for approximately 11% of all benign tumours (Strong, Tate and Borys 2012), but in historical material it is a rarer phenomenon. Jones (2006) observed that this tumour is more common in females, but Moon et al. (2006) reported that it was more common in males. In our study both cases represented males. Osteomas may originate from the periosteum and appear on the bone as an external mass. The tumour can exist inside the bone and lead to a distortion of the maxilla or mandible. Osteoid osteomas of the skull are usually located on the frontal bone (Strong, Tate and Borys 2012;

Garrigosa, Ledesma and Servat 2013). The tumour has an oval or round shape, with a smooth or lobular surface, and is slow-growing, as in our research. It is asymptomatic unless the alveolar nerve is involved (Sayan et al. 2002; Janas and Grzesiak-Janias 2005; Jones 2006). The tumour may be identified accidentally during an X-ray examination. Osteomas often coexist with Gardner's syndrome, which is a genetic disorder characterized by the presence of multiple polyps in the colon and tumours outside of the colon, such as osteomas of the skull, thyroid cancer, epidermoid cysts, fibromas and sebaceous cysts (Gardner 1951; Plenk and Gardner 1954). Because of the valuable nature of bone material and radiological image indicative of osteoid osteoma, no histological specimens were prepared to avoid damage to the skull structure. Inflammatory diseases, osteomyelitis and other benign tumours were considered during differential diagnosis. The final diagnosis was made based on the macroscopic appearance and the characteristic radiological image.

Conclusion

The study of the human skull alone provides a significant amount of information regarding health and diseases in historical populations. Every case report of a tumour found in excavated remains helps to reconstruct the history of the spread of tumours throughout a population.

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Authors' Contributions

AGS: responsible for radiological examinations and analysis, made conceptual contributions and wrote the manuscript, performed the review work; BK: responsible for project, made conceptual contributions, made measurements and wrote the manuscript, performed the review work; JSz: conducted statistical analysis, made measurements and wrote the manuscript, performed the review work; SG: collected material, made conceptual contributions, examined skeletal material; PD: made conceptual contributions, wrote the manuscript, performed the review work.

Conflict of interest

The authors declare that there are no conflicts of interest.

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