VIA MEDICA

CASE STUDY

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A paleopathological case of pituitary tumor, Eagle's syndrome and ossifying fibroma

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ABSTRACT

The salvage excavation in Mafraq region in Jordan uncovered a Byzantine female skull that died at an age of 18–25 years old. The visual and x-ray examination revealed diagnoses of pituitary adenoma, Eagle's syndrome, and maxillary ossifying fibroma. In addition, the case suffered a very poor oral health; dental caries, abscesses, and periodontal disease.

Key words: Pituitary tumor, Eagle's Syndrome, Ossifying fibroma, Byzantine, Jordan

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Introduction

Numerous diseases in the Byzantine period of Jordan were prevalent and had a high morbidity, such as osteoarthritis [1], periodontal diseases [2–4], anemia and rickets [5–7], infectious diseases [8–9], trauma [10], Paget's disease [11–12], and the Justinianic Plague from 541–750 AD [13], which caused massive loss of life on recurrent occasions [14]. The lack of reported tumorous lesions in the Byzantine skeletal materials is undoubtedly owing to the reliance on anthroposophical investigation by bioarchaeologists rather than a systematic radiological examination, and probably to the infrequent occurrence of such lesions on bones. This study reports a number of paleopathological lesions of a case from the Mafraq region in northeastern Jordan dating to the Late Byzantine period. These lesions were rare as being not reported in the bioarchaeological literature of Jordan.

Materials and methods

The skull was found after salvage archaeology at Mafraq in northern Jordan without the post cranial skeleton, it has an excellent state of preservation except for a postmortem fracture in the right parietal bone. Based on the associated pottery sherds, a Late Byzantine date was concluded (450–640 AD). The skull was examined visually and then using x-ray in the lateral projection.

Results

The age at death is estimated to be 17–25 years based on tooth attrition [15] and 18–44 years based on the vault sutures [16] yielding an approximate age of about 18–25 years. The size of the mastoid process, the surface of the nauchal area, the thickness of the supra-orbital margins, the profile of the supra-orbital ridge and the mental eminence indicate a female sex [17].

The styloid process was elongated; it was measured from its base to the apex, the point where the styloid process extended from the temporal bone [18], yielding a length of 27mm without the broken tip. The normal length is from 20–25mm [19]. In addition, the process is extremely slanted anteriorly towards the ascending ramus of the mandible (Fig. 1). The elongation and slanting could have caused head, neck and throat pain described as Eagle's syndrome [20].

The inferior portion of the maxilla exhibits four irregular bump-like bony growths superior to the greater palatine groove and adjacent to the alveolar bone of the molars (about 5 × 5 mm), and with central cavities (Fig. 2). This growth indicates a diagnosis of ossifying fibroma, which is a benign and painless tumor [21]. The cause of this disease is unknown and originates in the desmodontal cells of the alveolodental ligament [22]. It is common between the third and fourth decade of life and more frequent in women than in men [23].

Figure 1. A lateral view of the skull showing the elongated styloid process, photographed by H. Deebajah

Figure 3. Anterior view of the skull showing dental abscesses and periodontal disease, photographed by H. Deebajah

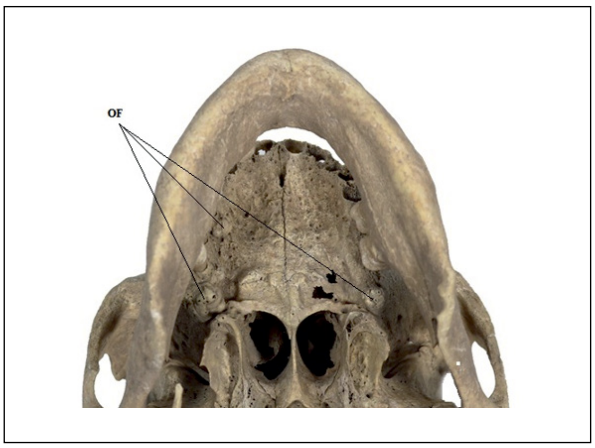

Figure 2. Ossifying fibromas (OF) of the maxilla, photographed by H. Deebajah

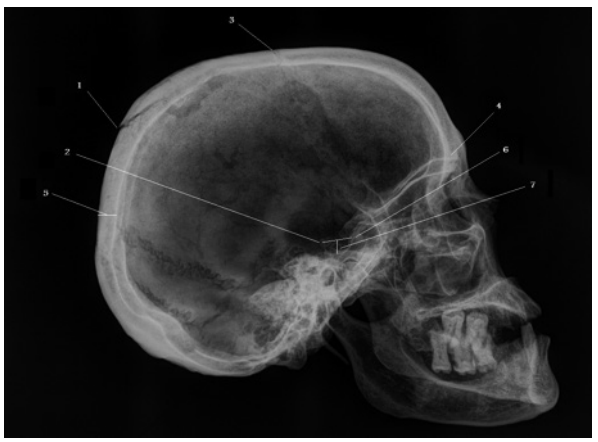

Figure 4. Skull lateral x-ray, 1 and 3 are postmortem fractures, 4 – closed frontal sinuses, 6 – length of the sella turcica, 7 – depth of the sella turcica, 2 – dorsum exostosis, and 5 – bone thickness

Premortem tooth loss, dental caries (buccal and interproximal), alveolar abscesses, and periodontal disease were also noted (Fig. 3). The oral health of the individual indicates a poor oral hygiene and a diet rich in carbohydrates [6].

The x-ray examination showed a cup-shaped enlargement of the sella turcica, marked thinning of the left posterior clinoid process with exostosis, and marked thinning of the sellar floor (Fig. 4). A normal pituitary gland measures about 8mm in its length (greater anteroposterior diameter) and 12mm in its traverse [24]. The average radiographic measurements of the normal adult sella have been given as length 10.6mm and depth 8mm [25]. The modern population of Jordan has an average length of 8.72mm and width 7.68mm [26]. The incidence rate of pituitary adenoma among Jordanians is 9.3% [27]. The length and depth measurements were exceeded in this case as the length is 17mm and the depth is 9mm. In addition, the skull bones are thick (about 1mm) compared to normal. The morphological changes in the sella turcica suggest a pituitary tumor with evidence of a marked intracranial pressure as shown by thick skull bones [28–29]. Frontal sinuses aplasia is also noted in the radiograph, which could be a normal variant among humans and occurs in 5% of normal adults [30], or a sign of acromegaly in pituitary adenoma [31].

Discussion

Approximately 4% of the living population have an elongated styloid process but not necessarily develop symptoms of Eagle's syndrome [32]. The symptoms may include dysphagia, neck pain, and a sensation of foreign object in the back of throat [33]. Few cases

of Eagle's syndrome were reported in the paleopathological literature [34–37], where none of them was reported from Jordan. The first case of ossifying fibroma in paleopathological literature was reported by Colrad et al. [38] from Pas-de-Calais region in France, dated to the 6th–7th century AD. Their investigation revealed a large lesion (30 × 20 mm) that occupied the right maxilla; they claimed that the lesion could have caused ophthalmological signs, such as, lachrymation, ptosis and exophthalmia. The fibroma of current case is relatively small suggesting a death before the progression of disease.

The tumor of the presented case would have caused a considerable increase in the volume of the pituitary and probably attributed to the bone exostosis in the posterior clinoid process. The generated pressure of the pituitary usually impairs the optic chiasm and, thus, visual impairment [39]. The thickness of the cranial bones associated with frontal sinus aplasia refers to a tumor that was acromegalic. Accordingly, the most probable diagnosis is a secreting pituitary adenoma. However, there have been few archaeological cases that illustrate the pituitary tumor in skeletal material [29, 31, 40–41], which signifies the rarity of the disease in antiquity.

Skeletal remains are the second most abundant material in Jordan's archaeological sites, where careful examination aided by physical and chemical analytical techniques (e.g.: radiographic imaging) may uncover a wide array of paleopathological lesions that were thought to be absent, tumors are an example but one. Many diseases in the Byzantine period were widespread and had a high morbidity [42]. Contrary to the typical believe that the rural life of the Byzantine people in Jordan was of poor quality, the people had better health and more wealth [43–44]. However, this does not negate the presence of rare and serious diseases such as pituitary tumor.

Conclusions

Tumors were not uncommon in antiquity and probably contributed to the morbidity and mortality as they do today. The prevalence of tumors in ancient skeletal material was underestimated; however, many of them are not associated with skeletal alteration but the use of modern diagnostic techniques may prove very rewarding.

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