Anadolu Araştırmaları
Anatolian Research

İSTANBUL UNIVERSITY PRESS

AnAr, 30, 1–17
DOI: 10.26650/anar.2024.30.1306705 Research Article

Paleopathological Analysis of Craniosynostosis (CS): Two Cases of Plagiocephaly from the Archaeological Site of Kayalıpınar, Sivas, Turkey

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Submitted: 30.05.2023
Revision Requested: 03.01.2024
Last Revision Received: 22.04.2024
Accepted: 23.04.2024

Citation: Sarı, İ., Acar, S., & Açıkkol, A. (2024). Paleopathological analysis of craniosynostosis (CS): Two cases of plagiocephaly from the archaeological site of Kayalıpınar, Sivas, Turkey. *Anadolu Araştırmaları-Anatolian Research, 30*, 1–17.
https://doi.org/10.26650/anar.2024.30.1306705

ABSTRACT
This article describes two individuals diagnosed with craniosynostosis. The archaeological human remains were uncovered from Kayalıpınar excavations in Sivas's province in Turkey. The skeletons described here belong to the Byzantine period. Anterior plagiocephaly (unicoronal synostosis) observed in a female aged 30-34, and posterior plagiocephaly (unilambdoid synostosis) observed in a male aged 20-30. Both skulls show deformities due to synostosis. It is assumed that premature suture closure has no influence on the death of individuals. Craniosynostosis is a known case in clinical studies; however, its frequency in archaeological records/paleopathological literature is unclear. No example of craniosynostosis (especially plagiocephaly) has been found in ancient Anatolian populations, except for scaphocephaly. In conclusion, in addition to dental anomalies, bulging on the frontal bone and lambdoid suture fusion were found in this study, which provides a specific example and useful results for future research.
Keywords: Plagiocephaly, Craniosynostosis, Unicoronal synostosis, Unilambdoid synostosis, Paleopathology

Introduction

A congenital anomaly is defined as a physical condition that begins before birth and negatively affects the development of the foetus (Barnes, 2008). This anomaly may be hereditary or acquired at birth (Aufderheide and Rodriguez-Martin, 1998). It may also not reveal its presence after birth (Barnes, 2008). Genetic factors are among the most known causes of congenital anomalies and are one of the main causes of infant mortality. Congenital anomalies can result in physical and/or mental disturbances. Indeed, it is a crucial factor in the disturbance and mortality of children (Oliveira and Fett-Conte, 2013). As a result, the quality of life of individuals is most likely affected by these side effects (Aufderheide and Rodriguez-Martin, 1998).

Suture is a form of joint in which adjacent bones in the craniofacial system articulate with a thin fibrous tissue layer (Kumar et al. 2020). During birth, the bones surrounding the skull stand apart with structures called sutures and fontanelles. This situation causes the skull to take on a particular shape (makes for an easier birth) (Barnes, 2012).

Some problems such as the lack of suture formation or premature fusion of the sutures can be observed during the development of cranial sutures. Suture deficiency is often described as sutural agenesis and is derived from a genetic origin (Roberts and Manchester, 2010). The sutures on the skull fuse following a certain order, that is, a fusion begins in the bregma region and continues along the sagittal, coronal, and lambdoid sutures, respectively (Aufderheide and Rodriguez-Martin, 1998).

Craniosynostosis is a congenital disorder of the skull (Buchanan et al., 2017). In this study, we report a rare case of plagiocephaly (unicoronal and unilambdoid craniosynostosis of skull) in two calvaria from Kayalıpınar excavations, Sivas province, Turkey.

Craniosynostosis (CS)

A disturbance in the development and function of sutures may result in craniosynostosis (Rice, 2008). Roberts and Manchester (2010) stated that CS is defined as premature suture closure, a congenital disorder (Buchanan et al., 2017), and closure of one or more of the six most important sutures of the skull (Waldron, 2009). Single suture fusion is more common than multiple suture fusion (Buchanan et al., 2017).

CS is divided into syndromic and non-syndromic CS (Pospíšilová and Procházková, 2006; Çeltikçi et al., 2013). The second is stated to have a strong genetic component, possibly through gene-gene or gene-environment interactions (Boyadjiev, 2007). Epidemiological studies have shown that non-syndromic craniosynostosis (NCS) is more common than syndromic craniosynostosis and constitutes 90% of patients with craniosynostosis (Collmann

et al., 2011). The most common NCSs are sagittal (scaphocephaly), unilateral coronal/lambdoid (anterior/posterior plagiocephaly), bilateral coronal/lambdoid (brachycephalic), oxycephalic, and trigonocephalic (metopic) craniosynostosis (Barnes, 2012; Collmann et al., 2011).

The incidence of CS in living humans is variable and occurs in 3-14 individuals out of 10,000 births (Waldron, 2009). Among the genders, women are more affected than men (Roberts and Manchester, 2010). This appears to be a rare case in skeleton populations (Mann and Hunt, 2012).

Birth trauma, intrauterine infection (Roberts and Manchester, 2010), congenital, hereditary, and metabolic diseases (Mann and Hunt, 2012), and hematological disorders (such as thalassemia and sickle cell anemia) (Rice, 2008) can lead to craniosynostosis. It is commonly mentioned that mutations in a gene known as fibroblast growth factor receptor (FGFR) are related to craniosynostosis (especially syndromic ones). The most common FGFRs associated with craniofacial syndromes include Apert, Crouzon, Pfeiffer, and Muenke (Giuffra et al., 2011). Other syndromes such as Saethre-Chotzen and Carpenter are independent of the FGFR mutation (Buchanan et al., 2017).

Premature closure of the sutures gives rise to different skull deformities (Ortner, 2003; Collmann et al., 2011; Buchanan et al., 2017). Deformities differ according to the location of the sutures (Barnes, 2008; Waldron, 2009) and the age at the beginning of closure (Ortner, 2003). The size of the deformity depends on the time of suture closure, and it becomes clearer when it occurs during embryological development (Ortner, 2003).

Plagiocephaly

Plagiocephaly is a term used to describe an abnormal asymmetric skull shape (Pospíšilová and Procházková, 2006). David and his colleagues (1982) divided plagiocephaly into three types: frontal, occipital, and hemicranial. Plagiocephaly is described as unilateral premature fusion of the coronal or lambdoid suture (Cohen, 1995; Buchanan et al., 2017). The least common is lambdoid craniosynostosis (Boyadjiev, 2007; Buchanan et al., 2017). Unilateral lambdoid synostosis constitutes approximately 1-3% of all craniosynostosis (Collmann et al., 2011).

Paleopathologically, in skull samples with plagiocephaly, it is possible to see changes/deformities in the skull and face according to the place of fusion (Çeltikçi et al., 2013). Deformities may occur when they result in plagiocephaly due to intrauterine pressure (Cohen, 1995). The anterior and posterior sides of the skull are bilaterally flattened (Pospíšilová and Procházková, 2006). Kreiborg and Björk (1981) emphasize the marked asymmetry of the orbit, cranial floor, and calvaria. In anterior plagiocephaly, flattening is observed in the frontal region,

where fusion is observed, elevation (rim) is observed in the supraorbital margin, and bulging is noticed on frontal (the opposite side). Posterior plagiocephaly causes flattening and prominent mastoid protrusion in the affected area (David et al., 1982; Buchanan et al., 2017).

This study focuses on examples of craniosynostosis that are well known in clinical cases, but are quite rare in paleopathological studies of ancient Anatolian populations. Plagiocephaly caused by unilateral fusion of the coronal and lambdoid sutures. Studies of plagiocephaly have not been encountered in the literature (in paleopathological studies of ancient Anatolian populations), whereas scaphocephaly has been observed in a limited number of studies. Therefore, this paper includes discussing examples of craniosynostosis in the context of clinical cases and archaeological populations based on paleopathological findings.

Methods

This paper presents two cases of premature suture fusion. The skeleton collection is in the laboratory of Sivas Cumhuriyet University, Department of Anthropology. Skeletons were obtained from the Kayalıpınar cemetery area in Sivas (Yıldızeli), Turkey (Fig. 1). The archaeological site was detected as a result of surface surveys carried out in different years (Ökse, 1994; 1999). Archaeological excavations were started for the first time in 2005, and as a result of these studies, traces of different periods (Hellenistic-Early Byzantine, Hittite Great Imperial Period, Middle Hittite Period, Erhitite/Karum Period) were discovered (Fig. 2) (Müller Karpe and Müller Karpe, 2006; 2012).

Figure 1: Location of Kayalıpınar, Yıldızeli/Sivas, Turkey

Figure 2: Kayalıpınar archaeological settlement area (google.com) (04.05.2023)

Kayalıpınar individuals were recovered from the uppermost layer (Layer 1) of the archaeological site (Müller Karpe and Müller Karpe, 2012) and from tomb types such as sarcophagi, stone, pottery, and simple earthen (Müller Karpe, 2006; Müller Karpe and Müller Karpe, 2009). Many tombs have been damaged due to erosion and agricultural activities (Müller Karpe and Müller Karpe, 2006). The uncovered tombs date to the Hellenistic-Early Byzantine period (Müller Karpe and Müller Karpe, 2012). The skeletons, which analysed in this study date to the Byzantine period. Both individuals were found in west-east direction, and the heads were found looking to the west. The tomb, numbered 207, was recovered from a simple earthen tomb. The tomb, numbered 217, was surrounded by mudbrick. With the help of the wooden pieces on the tomb of this individual, it is assumed that the tomb was covered with wooden. There were no tomb gifts in the tombs.

Sex and age determination criteria (WEA, 1980; Lovejoy et al., 1985; Szilvàssy and Kritscher, 1990; Kaur and Jit, 1990) were used for both individuals in the study. The regression equation as stated by Goodman and Rose (1990) was used to determine the estimated age of linear enamel hypoplasia. Identification and classification of craniosynostosis samples were performed according to Barnes (2012). Paleopathological analyses of human remains were performed macroscopically according to Ortner and Putschar (1981).

Results

Tomb number 207 was a female individual and, between 30 and 34 years old at the time of death (Fig. 3). The individual had plagiocephaly (anterior) in the skull. The left side of the coronal suture was fused from the bregma to the pterion (Fig. 4). This region was also closed endocranially. Bulging on the frontal was obvious (Fig. 4). Traces of fusion were

observed in the sagittal suture, but the other sutures on her skull were opened. While the right sphenofrontal suture was fully open, the fusion process started on the left side (Fig. 5). There were no wormian ossicles on the sutures. There was no significant difference between the two orbits in terms of width and height. Both orbital depths and orbital distance appeared normal (Fig. 6). Midface and mandible prognathism were not observed. The palate was generally narrow (not suitable for measurement). In the maxilla, the dental arches were parallel to each other. The greatest differences in the measurement values were observed in skull height (porion-bregma), mastoid length, maximum ramus height, and processus mastoideus (medial-lateral width) (Table.1). The individual exhibited a dolichocephalic skull (cranial endis; 73,33). The right foramen rotundum was slightly larger than the left (Fig. 6).

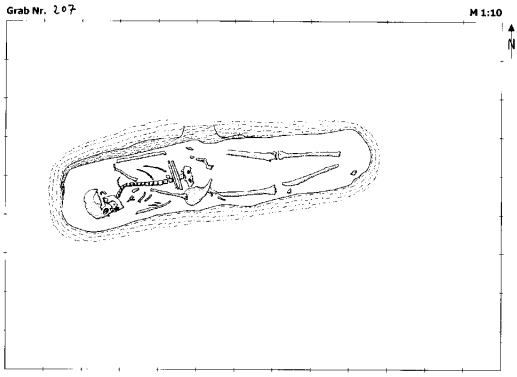

Figure 3: Architectural drawing of Tomb 207

Figure 4: Vertical view of the skull and CT of the skull (Tomb 207)

Figure 5: Right and left lateral views of the skull (Tomb 207)

The individual was examined paleopathologically. Accordingly, there were no cribra orbitalia in either orbital ceiling. There were slight traces of porotic hyperostosis in the occipital bone. Periostitis has been detected on the tibias. In addition, enthesopathy was seen in its initial stage on both patella bones. Mild osteoarthritis on the articular surfaces of many bones has occurred. Moderate osteoarthritis was also observed on the lumbar and thoracic vertebrae bodies and mild osteoarthritis formations on the upper and lower joint surfaces of the cervical, thoracic, and lumbar vertebrae. The processus spinalis of the existing vertebrae were not discrete. At the anterior right edge of the promontorium, there was a prominent bone formation extending towards the lumbar vertebrae. Caries, abscess, antemortem tooth loss (AMTL), different accumulations of calculus, linear enamel hypoplasia, moderate and severe tooth wear, and periodontal disease were detected in the jaw and teeth of the individual. Some teeth had hypoplasic (linear) defects (showing distribution a range of the age of 4 to 6 years).

Table 1. Measurement values of female individual (mm)

Measurements	Left	Right
Maximum Skull Width	132	
Maximum Skull Length	180	
Bizygomatic Width	119	
Orbital Width	36,03	35,32
Orbital Height	35,56	34,87
Frontal Axis	110,70	
Sagittal Axis	112,48	
Occipital Axis	92,76	
Frontal Arc	12,9	
Sagittal Arc	12,4	
Occipital Arc	11,1	
Foramen Mentale Width	44,49	
Porion-Bregma Height	124	120
Mastoid Length	30,59	28,13

Mandible Body Thickness	12,58	12
Bicondylar Width	113,78	
Minimum Ramus Width	32,43	32,38
Maximum Ramus Width	41,97	40,43
Mandible Body Height (between M2-M3)	30,07	28,42
Maximum Ramus Height	66	70
Processus Mastoideus Internal-External Width	12,85	10,43
Sphenoid Wingspan	27,21	28,08

Tomb number 217 was a male individual and, at the age of 20-30 years old (Fig. 7). The skull of this individual had plagiocephaly (posterior). The left side of the lambdoid suture was fused from the lambda to the asterion (Fig. 8-9). The right lambdoid suture was nearly fused endocranially, whereas the left lambdoid suture was completely fused endocranially. Other existing sutures were opened. There were no wormian ossicles on the sutures. The cranial index could not be calculated because of the preservation status of the skull. The distance between the orbits and both orbital depths were normal. Midface and mandible prognathism were not observed. In the maxilla, the dental arches slightly protruded outward. Measurement values of male individual are shown in Table 2.

Figure 6: Anterior and basal views of the skull (Tomb 207)

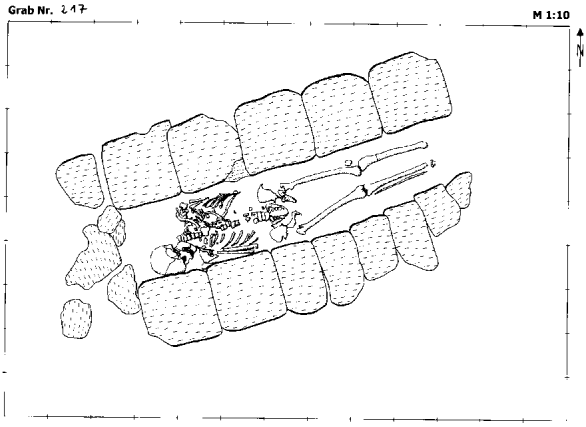

Figure 7: Architectural drawing of Tomb 217

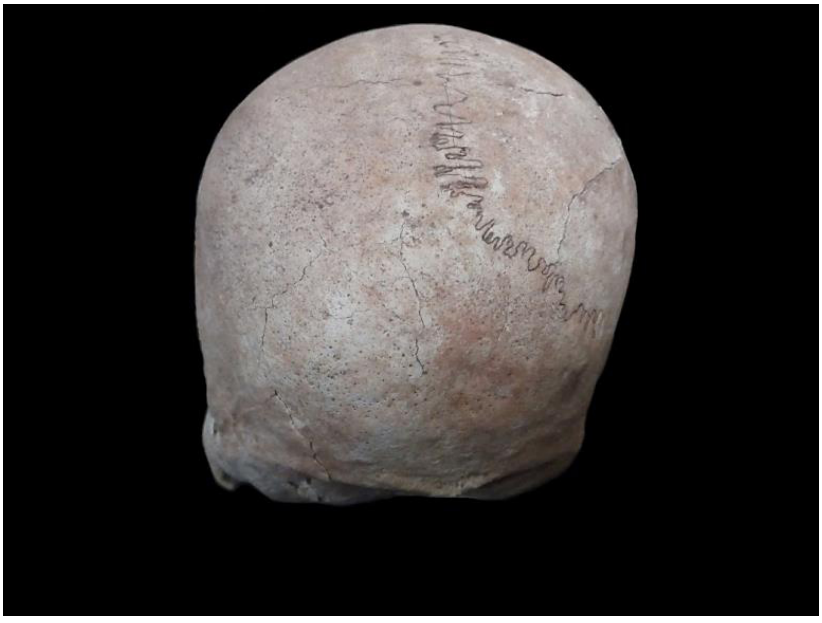

Figure 8: Posterior view of the skull (Tomb 217)

The individual was examined paleopathologically. There were porotic hyperostosis in the occipital and parietal bones. There was a blunt trauma just above the right arcus superciliaris on the forehead. The cribra orbitalia lesion could not be examined because of the fracture of the relevant region. There was periostitis in both tibiae. Mild osteoarthritis, which was in the initial stage, was observed in the bodies of the cervical, thoracic, and lumbar vertebrae and on the lower and upper joint surfaces. Processus spinalis was not distinct in the existing vertebrae. There were caries, abscess, calculus, AMTL, mild and moderate tooth wear, and

mild periodontal disease in the jaw and on the teeth of the individual; however, hypoplasia was not observed. Sarı and Açıkkol Yıldırım (2021) stated that the deciduous canine teeth in the maxilla of the individual did not fall out. In addition, the upper left permanent canine tooth (as connected to the deciduous canine tooth) performed the eruption in the lingual direction. Moreover, the upper right permanent canine completed development was impacted in the jaw (Fig. 10).

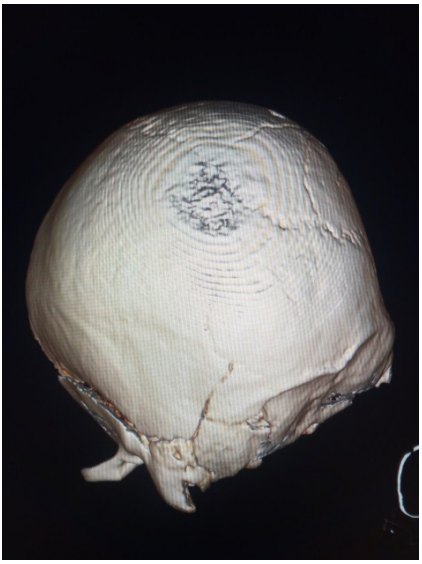

Figure 9: Posterior (CT) view of the skull (Tomb 217)

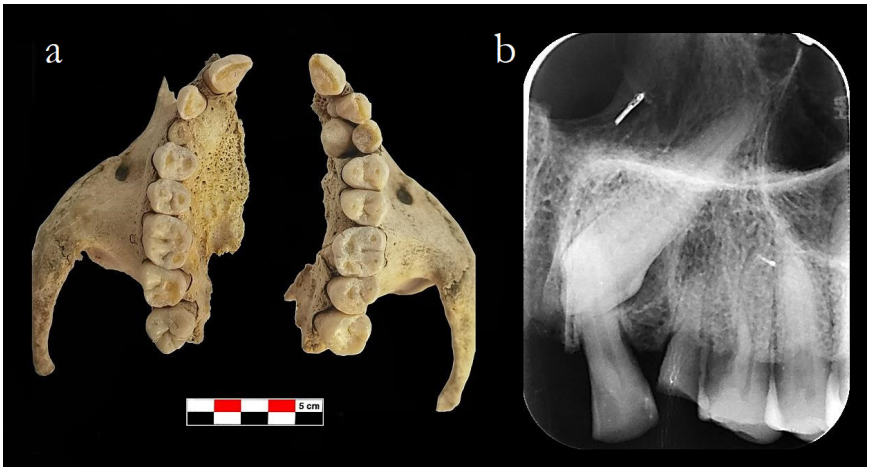

Figure 10: Impacted right upper canine and radiological view of tooth-Tomb 217 (Sarı and Açıkkol Yıldırım, 2021)

Table 2. Measurement values of male individual (mm)

Measurements	Left	Right
Maximum Skull Width	126	
Sagittal Arc	12,3	
Occipital Arc	11,4	
Mastoid Length	29,92	fractured
Mandible Body Thickness	11,66	12,12
Bicondylar Width	108,46?	
Minimum Ramus Width	31,39	30,11
Maximum Ramus Width	-	42,04
Maximum Ramus Height	67	67
Processus Mastoideus Internal-External Width	11,73	11,60
Bigonial Width	78,61	

Discussion

The diagnosis of unilateral coronal synostosis on a dry skull is made when there is no coronal suture on one side (Tulasne, 1987). It is possible to say that a similar situation applies to the lambdoid suture. Existence of craniofacial asymmetry does not mean that there is a fusion anywhere in the coronal suture system. The human face, cranial base, and calvaria may be also symmetrical despite unilateral fusion of the coronal suture (Tulasne, 1987). Furthermore, an infection or injury in an individual may appear as a congenital disorder or be confused with some complications (Barnes, 2008).

Although it is impossible to determine the frequency of congenital diseases in archaeological collections, it has been determined in some studies. In the study of Menard and David (1998), the true unilateral lambdoid synostosis rate was calculated as 0.98 (2/204). Indeed, samples of occipital plagiocephaly caused deformation existed in their research. The existence of these studies also provides an opportunity to understand the social behaviours towards disabled or physically disabled individuals in ancient populations (Giuffra et al., 2011).

Craniosynostosis refers to premature fusion of skull sutures (Ortner, 2003), and the most common type of CS in archaeological context is sagittal synostosis (scaphocephaly) (Duncan and Stojanowski, 2008; Évinger et al., 2016). In this study, we focussed more on samples related to our cases (plagiocephaly) and unearthed in archaeological contexts.

Gracia et al. (2009) reported that the left lambdoid suture was fused prematurely in a child at the Sima de los Huesos (SH) site (Atapuerca, Spain) in the Middle Pleistocene. Fusion may be of prenatal or traumatic origin.

Évinger et al. (2016) pointed out that the right side of the coronal suture was completely fused in a woman aged 30-35, who was dated from 9th century in Zalavár, Hungary. More

samples of plagiocephaly were found in the obtained skeletons from the Broumov Cemetery, which was dated to the 13-18th century in the Czech Republic. Anterior plagiocephaly was detected in two individuals (1 adult and 1 child) and posterior plagiocephaly in one individual (adult) (Pospíšilová and Procházková, 2006). Another example of plagiocephaly was found in the obtained skeleton during excavations at the 16th century Fort King George archaeological site in Georgia (US). It was a male between aged 23-45. The left coronal suture fused prematurely (Duncan and Stojanowski, 2008).

In a young adult (East Indian), estimated to be 20-25 years old and unknown gender, premature suture fusion on the right side of the coronal suture was discovered (Kreiborg and Björk, 1981). Suzuki and Jkeda (1981) reported early fusion of the right side of the coronal and lambdoid sutures in a 5 or 6 year old child from Rorei on Sakhalin Island. It was also fused early in the sagittal suture in this child. In another study, at the Vrolik Museum in Amsterdam, Netherlands, 160 skulls were examined, and unilateral coronal synostosis was determined in four individuals (two of them were adults and two children (Oostra et al., 2005).

Researchers have pointed out that there are deformities in the skull and facial bones of the individuals aforementioned and diagnosed with plagiocephaly (Kreiborg and Björk, 1981; Suzuki and Jkeda, 1981; Pospíšilová and Procházková, 2006; Duncan and Stojanowski, 2008; Évinger et al., 2016). It is possible to observe a similar outcome in a clinical study (Sakurai et al., 1998).

The number of individuals observed premature suture fusion in paleopathological examinations of ancient Anatolian populations is rare. Since plagiocephaly is the aim of this study, other craniosynostosis samples are briefly mentioned in the context of the period and population. Sevim Erol and Pehlivan (2014) noticed a sample of the premature fusion in the sagittal suture in a girl child of age 14-15 dated to 19th. The coronal suture of a young adult woman, aged of 17-25 years, from the Karagündüz (Early Iron Age) population fused early (Sevim et al., 2002). A sample of scaphocephaly was encountered in a 3-4-year-old child (probably a girl) from the Heracleia Perinthos (Byzantine) population (Demirel and Özkanlı, 2014). The presence of scaphocephaly was determined in an adult woman from the Beybağ (Ottoman) population (Karaöz Arıhan, 2021). The presence of scaphocephaly was detected in an adult male individual (Young Ottoman) unearthed from the Istanbul Karacaahmet Cemetery (Sağır et al., 2009). Şenyürek (1951)-Kaledoruğu-No 1 (Copper Age)- detected premature suture fusion in the sagittal and coronal sutures of a child (probably a girl) aged approximately 7 years. Şenyürek-Alacahöyük-No 9 (Copper Age) also reported a premature suture fusion in the sagittal suture with 17-18 years old female.

Many features related to syndromic craniosynostosis are catalogued in clinical and paleopathological studies. When evaluated at the skeleton level, these can include early fusion of the skull sutures, skull deformities, midface hypoplasia, mandibular prognathism, cleft palate, shallow orbit, hypertelorism, fusion and enlargement of hand and foot bones, fusion of the elbow (ankylosis), and fusion of cervical vertebrae (Kreiborg and Björk, 1982; Rice, 2008; Giuffra et al., 2011; Oberoi et al., 2012; Kumar et al., 2013; Évinger et al., 2016; Kumar et al., 2020). Moreover, early fusion of spheno-occipital and petro-occipital synchondrosis may occur in individuals with Apert and Crouzon syndromes (Rice, 2008). Craniosynostosis, anomalies in the eye and midface, and abnormalities in the hands and feet may be similar in Crouzon, Pfeiffer, Apert, Jackson-Weiss, and Muenke syndromes (Giuffra et al., 2011).

Similarly, features such as impacted teeth, ectopic eruption, crowding, delayed tooth eruption, supernumerary teeth (hyperdontia), enamel hypoplasia, congenital tooth deficiency (hypodontia), and malocclusion, both deciduous and permanent teeth, can be seen in syndromic cases (Rice, 2008; Oberoi et al., 2012; Kumar et al., 2020).

The paleopathological results of the two individuals were given in this study. As a result, no correlation was found between the data obtained and the syndromic CS symptoms reflecting the human bone. However, Kreiborg and Björk (1982) stated that in an 18-year-old and possibly female individual with Crouzon syndrome, in addition to skull deformities, both deciduous canine teeth are preserved in the maxilla, whereas the permanent canines are located on the palate. Apart from the preservation of both deciduous canines in the upper jaw of the male individual (tomb no 217), the upper right permanent canine was also impacted. Although these results are in good agreement with the study of Kreiborg and Björk (1982), the current data are not sufficient to mention from any syndrome.

Besides, the mouth and dental health of both individuals were not good. Caries, abscess, dental calculus, AMTL, tooth wear, and periodontal diseases from jaw and dental pathologies were observed in both individuals. Although many factors are responsible for the formation of enamel defects (Mays, 1998; Schultz et al., 1998), systemic metabolic stress, localised trauma and hereditary anomaly was taken into consideration as the main components (Goodman and Rose, 1990). The female individual (tomb no 207) may have experienced a health problem between the ages of 4 and 6 that would affect her growth and development. Based on our findings, it is difficult to say whether this situation is associated with premature suture fusion.

Conclusions

This study is an important source of suture fusions in the archaeological record. As the number of examples exhibiting these pathologies is scarce, each new finding will be crucial

for the evaluation of ancient populations. The characteristics of the specimens in the study are not sufficient to support the presence of syndromic craniosynostosis (e.g. syndactyly, shallow orbit, fusion of elbow, fusion and enlargement of hand and foot bones). On the basis of the available data, it is not possible to say whether two individuals who have observed craniosynostosis have visual disturbances (visual discomfort), cognitive, or any health problems (such as headache) or their social status in the society in the course of their lives. Nevertheless, bulging on the frontal bone and linear enamel hypoplasia on the teeth were remarkable. Considering the reasons for their occurrence (linear enamel hypoplasia), it is likely that the female individual has some health problems at the age of 4–6 years. Another interesting finding was the preservation of deciduous canine teeth and impacted permanent canines. As a result, it is assumed that the lower frequency of lambdoid suture fusion in the archaeological record has increased the importance of this study.

Acknowledgement: We would like to thank Andreas Müller Karpe and Vuslat Müller Karpe with mercy and gratitude.
Ethics Committee Approval: N/A.
Peer-review: Externally peer-reviewed.
Author Contributions: Conception/Design of Study- İ.S., S.A., A.Ç.; Data Acquisition- İ.S., S.A., A.Ç.; Data Analysis/Interpretation- İ.S., S.A., A.Ç.; Drafting Manuscript- İ.S., S.A., A.Ç.; Critical Revision of Manuscript- İ.S., S.A., A.Ç.; Final Approval and Accountability- İ.S., S.A., A.Ç.
Conflict of Interest: The authors have no conflict of interest to declare.
Grant Support: There is no financial support.

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