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An Ottoman-period male skeleton with cleft palate and lip

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Abstract

Cleft palate and lip develop as a result of any disruption in the formation of palate and lip during the first 12 weeks of embryonic development. Besides the heredity, there also are cases suggesting that this deformation was caused by the problems, which the mother has experienced during the pregnancy period. In addition to the nutritional problems, this defect causes many other problems such as dental problems and infectious and psychological disorders. This defect is not a very commonly observed palaeopathological finding in studies carried out on archeological human skeletal remains. Cleft palate and lip were detected on the skull of a male constituting the study material, which was labeled with the number 1178 and dated to the Ottoman period. In literature, the first cleft palate and lip found in Anatolian archeological human skeletal remains were found in this individual. Computed Tomography (CT) images were taken for the skull, on which cleft palate and lip were found, and the analyses were conducted by obtaining the 3D image by making use of a 3D laser scanner. For the *control* group consisting of 20 individuals from the same community, 22 metric measurements were performed on the skulls by making use of the Materialise MiniMagics software. In this individual, nasal septum deviation has developed because of the effects of cleft palate and lip defects.

Introduction

Cleft palate and lip defects, which are found in archeological human skeletal remains, arise from the incomplete formation of the two halves of the palate during the fetal development process (Roberts and Manchester, 2010). Cleft palate and lip in the craniofacial area are congenital defects observed in approximately in 1/700 live births (Tur et al., 2016). Given the previous studies, the incidence of cleft palate and lip varies by geographical region, ethnical status, and gender. In their study, Yiğit Kızılelma et al. (2015) found cleft palate and lip in 19 of 17990 live births. Among these cases, cleft palate was found in 6 individuals, cleft lip in 1 individual, and both of them in 12 individuals. Its incidence in Turkey was found to be 1/947 live births. This ratio is 1.72-2.12/1000 for Asians and 0.2-0.72/1000 for blacks (Yiğit Kızılelma et al., 2015). Examining the distribution of this defect between genders, it was found that its incidence was higher among male individuals (Karaman, 2009; Tırank et al., 2022). The main reason for this distribution by gender is the effect of the X chromosome. Male babies have a disadvantage since they have one X chromosome but female babies have two (Wallace et al., 2011).

Considering the embryological development and epidemiological differences of individuals, some individuals are seen to have cleft lip and/or palate, whereas some individuals seem to have both cleft palate and cleft lip at the same time. The simplest classification was provided by Veau (1931) as follows;

- Unilateral cleft
- Bilateral cleft
- Incomplete cleft (incomplete cleft affecting the lip and not involving the cleft or nose)
- Complete cleft (complete cleft influencing lip, frontal maxilla, nose, and hard and soft palate)

Cleft lip and/or cleft palate, which are congenital defects developing as a result of genetic and environmental factors, develop due to the fusion defect on the face of an individual during the embryologic process. The palate, which consists of the primary palate and secondary palate structures, forms the upper lip as a result of the fusion of lateral nasal prominence and medial nasal prominence since the 6th week of embryologic development. The primary palate is formed by the left and right maxillary projections. After these two formations, the palatal protrusions located in the inner portion of the right and left maxillae fusing with the nasal septum form the secondary palate. Then, the soft palate forms in the following 11th week and the uvula forms in the 12th week (Sadler, 2011; Alpağan Özdemir, 2018). At the end of the 12th week, the palate and lip structures of the fetus are completed. Clefts develop as a result of any disruption in this process (Stainer and Moore, 2004; Karaman, 2009).

The main environmental factors playing role in the development of the cleft palate and lip include the medications used by the expectant mother during the pregnancy period (vasoactive medications -pseudo-epinephrin, aspirin, ibuprofen, amphetamine, anticonvulsant medications - phenobarbital, dilantin, naproxen, glucocorticosteroids, corticosteroids), alcohol use, smoking, stressful pregnancy, trauma, diseases, X-ray exposure to the baby while in the womb, folic acid deficiency, and vitamin deficiency (A, B6, B12) (Riley and Murray, 2007). If the first child in the family has this defect, then the incidence of this defect in the second child was 2-4% and that for the third child was 10%, whereas the incident for the third child after two children with this anomaly was 20, the incident for the first child when either mother or father has this defect was 5-6%, the incident for the first child when both mother and father have the defect was 25% and the incident was reported to be 40-60% for identical twins, 5% for fraternal twins, and 0.5-1% for the cousins (Yıldırım and Seymen, 2015).

In studies examining the gene factor influencing the cleft palate and lip, the factors were analyzed rather than the relationships between the factors. The genes examined by the researchers the most are transforming growth factor alpha (TGF-α), Drosophila msh homeo box homolog-1 (MSX1), transforming growth factor beta (TGF-β), B-cell CLL/lymphoma 3 (BCL3), interferon regulatory factor-6 (IRF6), poliovirus receptor (PVR), and paired box 9 (PAX9) (Vieira et al., 2005). Cleft palate and lip defects develop not only due to genes but due to both gene and environmental effects.

The aim of the study is to reveal the formation of cleft palate and cleft lip and how this formation affects the morphological appearance in skull number of 1178 among Ottoman period skeletons.

Materials and method

The skulls taken from Istanbul Karacaahmet Cemetery between 1925 and 1932 constitute the Ottoman Period population. Skull 1178, identified among 500 individuals, constitutes the material of our study (Fig. 1). The biological age estimation of the Ottoman period skulls was made according to the closure of the cranial sutures. Sex estimation was based on eyebrow arches, mastoid process, structure of the orbits, zygomatic process, morphology of the mandible, muscle attachment marks, and general structure of the skull (Buikstra and Ubelaker, 1994; Workshop of European Anthropologist, 1980; Ortner and Putschar, 1981).

Following Roberts and Manchester (2005), the paleopathological phenomena obtained from the skull of an adult male individual numbered 1178 were determined by making use of the national and international paleopathology resources. Moreover, the anomaly was analyzed in detail by using a 3D laser scanner and computed tomography (CT). The analyses were conducted with metric measurements from the skull by using Materialise MiniMagics utilized in imaging and measuring the mesh-based data (Fig. 2).

Results

There was no mandible on the skull of the adult male individual Nr. 1178, which constitutes the material of this study. Then, the examinations were performed considering only the maxilla.

Given the first analysis, the cleft palate and lip detected on the skull of the male individual Nr. 1178 was observed mainly on the left side of the skull. Accordingly, there was a gap starting from the half of 2nd right upper incisive tooth to the left upper canine tooth. Half of the 2nd right incisive tooth, 1st left incisive tooth, 2nd left incisive tooth, and a part of the left canine tooth constituted the oral cleft. In this formation, the distance between the 2nd right incisive tooth and the left canine tooth was found to be 15.46 mm (Fig. 3).

The left-oriented oral cleft also affected the nasal formation of the individual. Nasal septum of the individual was observed to deviate to the right side and there was no excessive growth in the pneumatized turbinates. The main reason for the medial nasal wall's deviation from the center to the right side was because the oral cleft has formed mainly on the left maxilla of the individual (Fig. 4).

Figure 1: Skull of the male individual Nr.1178

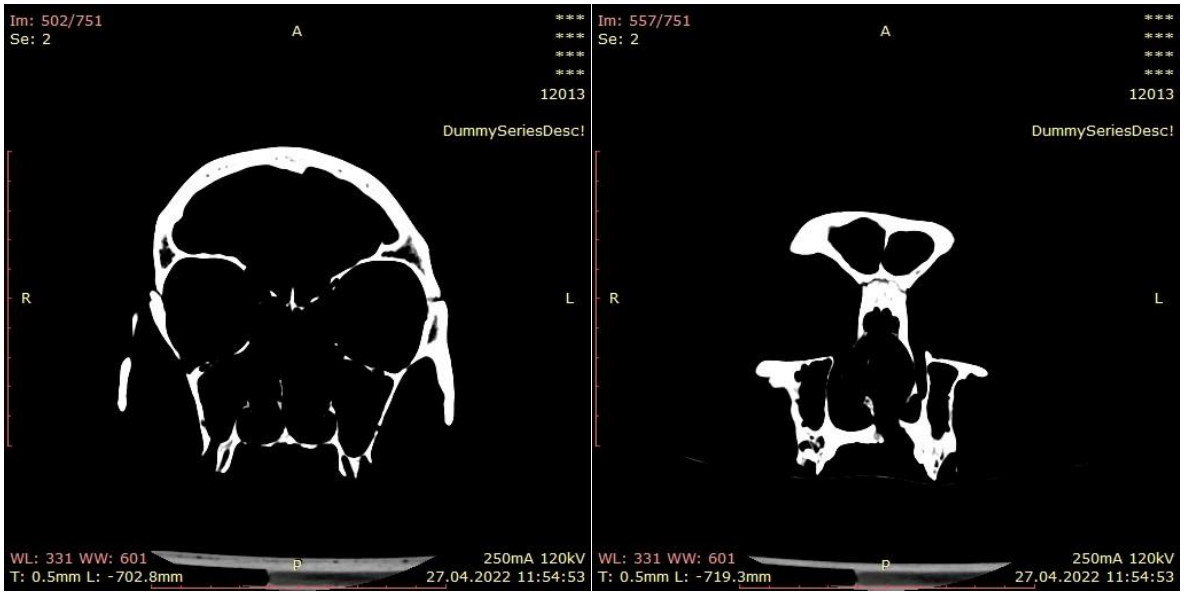

Figure 2: CT images of the male individual Nr.1178

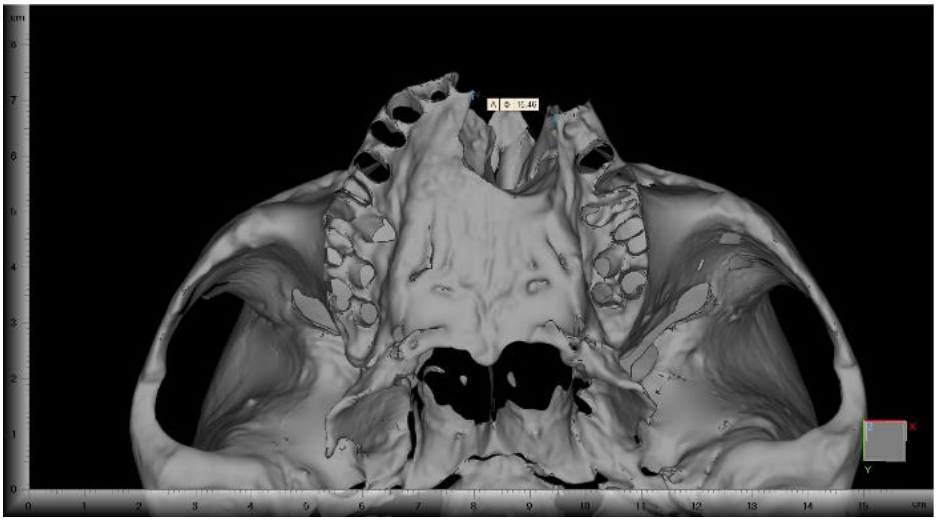

Figure 3: Measurement of cleft palate and lip gap

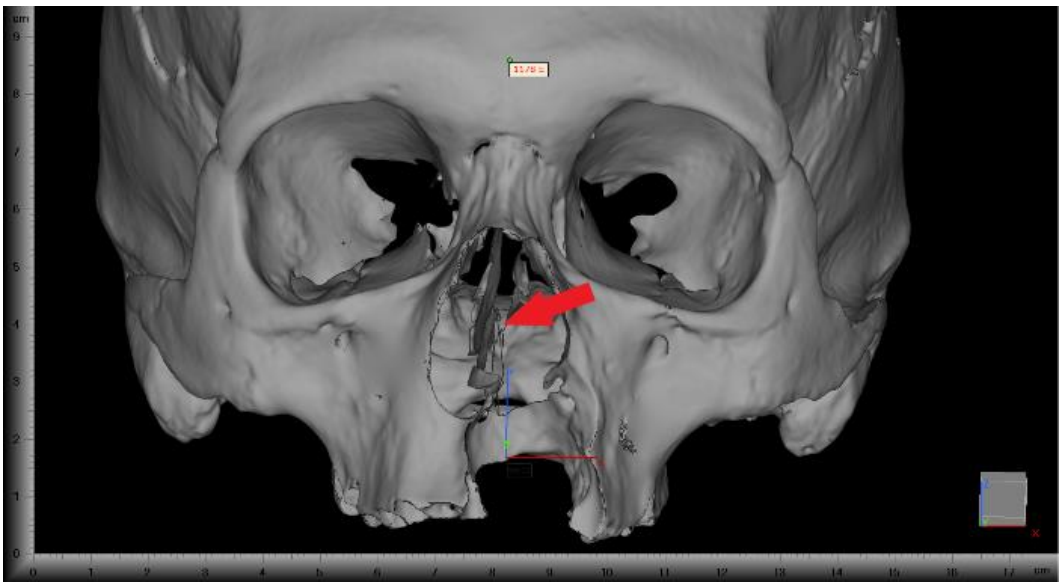

Figure 4: Deviation of nasal septum to the right side

In order to determine the effect of cleft mouth on general morphology, 20 male individuals were randomly selected as a control group among 500 individuals from the population of the Ottoman period. The skulls were in harmony with each other in terms of age and gender. Twenty-one skulls examined were scanned using a KScan Magic 3D laser scanner and then transferred to the digital environment. Anatomic points to perform craniometric measurements were determined on the skulls. Then, by using the Minimagics, a 3D imaging software, 24 measurements were performed at the predetermined points (Table 1).

Discussion and conclusion

Cleft palate and lip defect was observed on the skull of a male individual labeled with number 1178 and dated to the Ottoman period. Individuals having congenital cleft palate and lip defects face many difficulties throughout their lives. Nutrition, which is the most important phenomenon of the babyhood period, is the most important problem for babies born with cleft palate and lip. The babies having only cleft palate have a slight advantage. The babies having cleft palate and lip cannot produce sufficient negative pressure during breastfeeding. Longer breastfeeding time in comparison to the other babies is a problem for the baby (Gottschlich et al., 2018). In babyhood, which is the first period of an individual's life, deaths occur because of insufficient nutrition due to this defect. Regarding the nutrition, these individuals experience growth and developmental delay in comparison to their peers. Since the lower jaw and body skeleton of the individual with cleft palate and lip were not available, no clear information could be given about his social status. However, although it is difficult for individuals with cleft palate and cleft lip to continue their lives, the skull numbered 1178 probably continued his life until adulthood with careful care. This formation, which is called complete cleft, develops as a result of genetic and environmental factors until the 12th developmental week of an embryo in the womb. This formation can be clearly detected as of the 12th week of embryonic development.

As a result of the gap in the mouth and nose of individuals having a cleft palate and lip defect, respiratory problems and then otitis media develops in these individuals. This deformation, which directly influences speaking, also causes learning difficulties. It is thought that the oral cleft of individual Nr. 1178 having the length of 15.46 mm from the 2nd right incisive tooth to the left canine tooth caused speech impediment. Cleft palate and lip defect prevents

the tongue from being positioned appropriately while speaking (Akarsu Güven and Kocadereli, 2015). Nr. 1178 individual's cleft palate and cleft lip, it is observed that this opening in the mouth causes both speech problems and facial asymmetry in terms of appearance, since the normal positioning of the tongue does not occur due to this opening.

Table 1: Comparison between individual Nr.1178 and control group

Measurements (mm)[1]	1178 Individual[2]	Control Group	
		Mean (N=20)[3]	SD
Maximum head length (G-Op)	175.17	173.85	7.49
Maximum head width (Eu-Eu)	126.23	131.73	6.03
Head length (Ba-B)	131.40	135.89	5.86
Cranial base length (Ba-N)	96.20	97.82	5.48
Minimum frontal width (Ft-Ft)	91.76	94.63	4.63
Facial base length (Ba-Incision)	69.13	92.86	6.19
Upper face width (Fmt-Fmt)	99.52	100.84	3.96
Bizygomatic width (Zy-Zy)	130.34	129.92	5.30
Maxilla-alveolar width (Ecm-Ecm)	57.38	57.39	5.10
Maxilla-alveolar length (Incision-Alv)	32.97	53.38	3.75
Palatal length (Incision-Sta)	29.88	48.69	3.52
Palatal width between canine teeth	27.84	26.15	6.60
Palatal width between 2nd molar teeth (Enm-Enm)	33.26	39.11	2.71
Min. nasal bone width-simotic chord	7.41	9.26	1.58
Interorbital width-dacrial chord (D-D)	17.03	19.87	1.91
Intraorbital width by skull base length	17.70	20.81	2.78
IntraOrbital width by bizygomatic width	13.06	15.30	1.40
Maxilla frontal width (Mf-Mf)	16.38	15.77	2.19
Orbital width (Ect-D) (Right)	40.36	39.98	1.59
Orbital width (Ect-D) (Left)	39.71	40.17	1.35
Orbital height (orbitalhHeight) (right)	35.98	36.24	2.20
Orbital height (orbital height) (left)	35.19	35.82	2.55
Max. nasal width (Al-Al)	26.83	23.71	2.05
Nasal bone length (N-Rhi)	18.77	17.42	3.26

1. Measurements according to Martin (Martin and Saller, 1957). Craniometric Points: alv (alveolon), ba (basion), b (bregma), d (dacrion), ecm (ectomolare), enm (endomolare), ekt (ectoconchion), eu (euryon), g (glabella), gn (gnathion), fmt (frontomalaretemporale), ft (frontotemporal), ecm (ectomolare), mf (maxilloofrontal), n (nasion), op (opisthocranion), sta (stafilyon), zy (zygion).
2. Individual having an oral cleft.
3. 20 individuals constitute the Control Group.

For individuals having a cleft palate and lip defect, it is possible to experience problems related with the nasomaxillary, pterygomaxillary, and pharyngeal anatomy deviating from the standard anatomical order, problems in maxillary sinus formation, and sinusitis problems. The deviation of the nasal cavity to the right side because of the defect in individual Nr. 1178 is thought to prevent the individual from healthy respiration and he had experienced upper airway disorders throughout his life.

Individuals having cleft palate and lip have dental problems (especially those related with maxilla), differences in saliva structure, oral and nasal halitosis, oral candida colonization, oral hygiene, and periodontal health problems.

Comparing 24 craniometric measurements from the control group and the individual Nr.1178, the highest level of difference was found in the location of the oral cleft. These measurements were facial base length, maxilla-alveolar length, and palatal length (Diagram 1). Apart from these measurements, no anomalies were observed in any measurements in the skull of the individual.

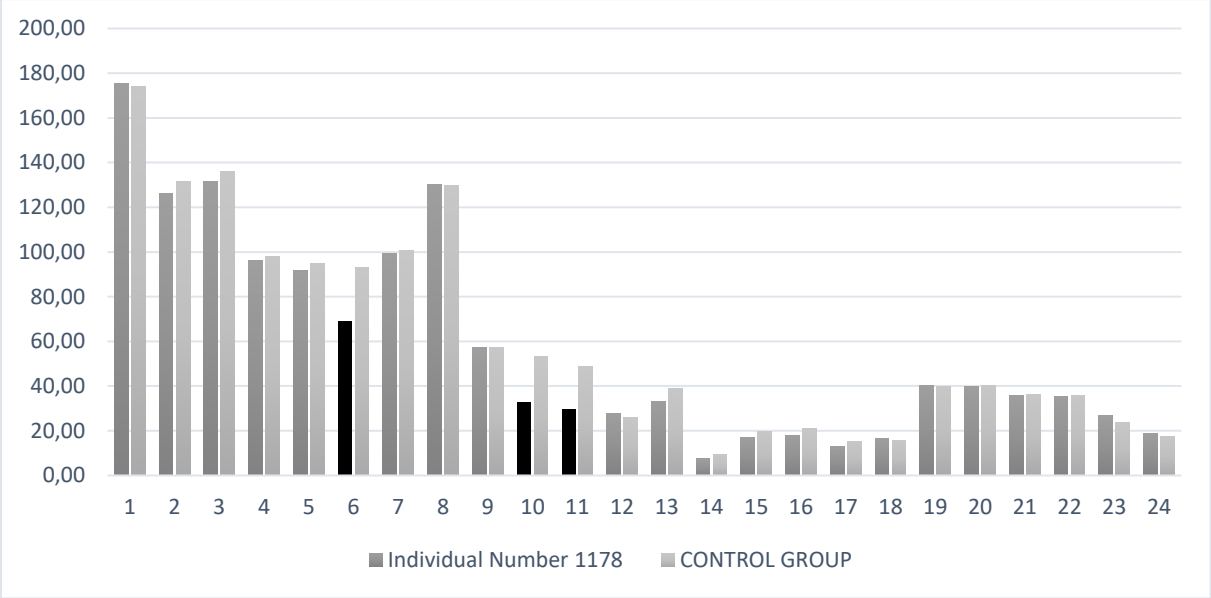

Diagram 1: Measurement results of Individual Nr.1178 and control group

Cleft palate and lip defect, which is rarely seen in archeological human remains, have been found in 52 individuals in total in Europe, America, Egypt, Russia, and Australia (MacCurdy, 1923; Derry, 1939; Berndorfer, 1962; Brooks and Hohenthal, 1963; Atherton, 1967; Alexandersen, 1967; Gladykowska Rzeczycka, 1980; Gregg et al., 1981; Brothwell, 1981; Miller and Merbs, 1993; Anderson, 1994; Webb, 1995; Malinowski et al., 1996; Hofman and Hudgins 2002; Ortner, 2003; Hegyi, 2004; Phillips and Sivilich 2006; Hawass et al., 2010; Barnes, 2012; Adams, 2016; Tur et al., 2016; Bodorikova et al., 2022). The individual Nr.1178 dated to the Ottoman period is the first case found in Anatolia and having lived with this defect.

In conclusion, it is thought that the cleft palate and lip defect, which was observed on the skull of male individual Nr.1178 dated to the Ottoman period, is thought to have caused inevitable problems in his life such as external appearance, nutrition and respiration disorders, and psychological and neurological problems. Despite the oral cleft affects the life of an individual, Individual Nr. 1178 constituting the study material reached adulthood and could maintain his life. Nowadays, alveolar bone grafting can be performed to restore the alveolar bone for children born with cleft palate and lip. The first successful alveolar bone restoration was performed in the year 1914 by Drachter using the tibia bone tissue. For bone grafting, attention is paid to performing the grafting before the complete shoot of the canine tooth and not preventing the growth of teeth. Besides the tibia, the other bones preferred for grafting include the iliac crest, calvarial bone, costa, and mandibular symphysis (Saatci and Ataç, 2021).

By making use of the alveolar bone grafting method, the problems in the lives of children born with cleft palate and lip can be minimized.

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