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An Evaluation of Bone Mineral Density Using Cone Beam Computed Tomography in Patients with Ectodermal Dysplasia: A Retrospective Study at a Single Center in Turkey

Authors' Contribution:
 Study Design A
 Data Collection B
 Statistical Analysis C
 Data Interpretation D
 Manuscript Preparation E
 Literature Search F
 Funds Collection G

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Background: The aim of this retrospective study was to compare the measurements of bone mineral density (BMD) combined with volumetric dental tomography measurements taken from three main regions selected on the lower mandible, the right mandibular, medial mandibular, and left mandibular regions, in patients with ectodermal dysplasia and age-matched and gender-matched controls. Measurement of BMD in Hounsfield units (HUs) were evaluated using three-dimensional (3D) cone beam computed tomography (CBCT) imaging.





Material/Methods: Mandibular bone tomography images were evaluated from 9 women and 5 men diagnosed with ectodermal dysplasia and a control group of 9 women and 5 men. The HU values obtained according to age and gender of the total 28 study participants were measured. Statistical analysis of the data used Student's t-test.

Results: BMD in the ectodermal dysplasia group was significantly lower compared with the BMD in the control group. Comparison of the left and right mandibular angulus regions showed that the BMD of patients with ectodermal dysplasia was significantly lower when compared with the control group in both regions, but no significant difference was found between the two groups in the BMD of the central mandibular region. CBCT was found to be an effective method for the measurement of BMD.

Conclusions: In patients with ectodermal dysplasia, reduced BMD should be taken into consideration when planning surgical interventions involving bone tissue and when planning implant surgery. The results of this study may be of value in dentistry and other fields of medicine.

MeSH Keywords: **Bone Density • Cone-Beam Computed Tomography • Ectodermal Dysplasia**

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Background

During embryonal development, the skin and structures in the skin originate from the ectoderm. Ectodermal dysplasia is a heterogeneous group of inherited disorders in which at least two tissues are affected, with abnormalities in the ectodermal structure [1–5]. The classic triad of ectodermal dysplasia is formed of anomalies of the hair (hypotrichosis), the teeth (anodontia or hypodontia), and the sweat glands (anhidrosis or hypohidrosis) [6–10]. Currently, more than one gene has been identified in association with ectodermal dysplasia. However, abnormal expression of these genes is not limited to ectodermal tissues, but also to structures of mesenchymal origin [11–15]. Therefore, ectodermal dysplasia is associated with malformations in several other organs and systems [11–15]. The incidence of ectodermal dysplasia has been reported to be approximately 7/10,000 live births [6].

Abnormalities of dental development can be associated with other growth and maturation abnormalities in children. Generally, primary teeth start to emerge at between 5–8 months, beginning with the lower incisors [4,9]. Delayed dental development is defined as the non-emergence of primary teeth in infants at by 13 months. The most common reasons for delayed dental development include hypothyroidism, hypoparathyroidism, and genetic factors. Mechanical obstruction from impacted teeth as a result of gingival fibrosis and malocclusion may lead to the non-emergence of the teeth. However, most cases of non-eruption of the teeth are idiopathic with no identified cause. The most common dental developmental abnormality of the primary and permanent dentition is hypodontia, which is the developmental absence of one or more teeth (excluding the third molars). Hypodontia may arise as a non-syndromic familial form or as a component of a syndrome that may be sporadic. The typical radiographic finding in hypodontia is deformation in the shape of the existing teeth.

Bone is a tissue that continuously renews itself, with new tissue made to replace bone that is destroyed. However, by the fourth decade, bone destruction slightly overtakes bone renewal, and by the sixth decade, bone destruction increases, resulting in thinning of the bone and loss of bone resistance [7,9]. Periapical and panoramic X-ray imaging are not adequate to show bone density because the lateral cortical layer prevents the visualization of the density of trabecular bone. Bone density is determined more accurately with the use of computed tomography (CT), which provides an axial image, perpendicular to the long axis of the body.

This imaging scale used for CT is named after the British physicist, Godfrey Hounsfield, who developed the imaging device in 1972, and the numerical data determined in the Hounsfield scale are known as Hounsfield units (HU). One of the criteria

for the objective evaluation of bone density is scoring with the HU [16,17]. Each axial CT image is formed from 260,000 pixels and each pixel has a Hounsfield unit (HU) CT number related to the tissue density. A high HU value indicates high bone density. Structures such as calcification and bone, which absorb X-rays are seen as white and give a high HU value (80–100 HU). The scale range includes water reflected at a moderate level (0 HU), fat at below zero (–80 HU) and air at the lowest values (–1000 HU) [17]. Compared with trabecular bone, cortical bone is denser but takes longer to heal. As bone density decreases, bone resistance also decreases. The most modern CT imaging devices include imaging slices of <0.5 mm. Each volumetric item of data includes three-dimensional (3D) voxels and the number of these 3D voxels determines the quality of the image, and a single shot image provides images from various angles [16].

Bone mineral density (BMD) is the most important objective parameter in the diagnosis and follow-up evaluation of systemic bone disease such as osteoporosis. BMD can also provide information about the health of the bone tissue around an orthopedic prosthesis, providing surgeons with the opportunity to review the treatment options. Fragility and resistance of the bone are directly proportional to the amount and organization of mineral substance, calcium, and phosphate in bone. As a result of local pathology or systemic disease, the reduction in mineral substance in the bone unit area shows high compatibility with volumetric BMD and the risk of fracture is increased.

CT is an imaging method in which the images obtained as slices from the body by collimation of X-rays, are transferred to a computer [18]. The CT device consists of the X-ray source, detectors, a gantry, and a computer unit. Obtaining a full image on CT occurs through separate radiation in a fan-shape for each slice taken in the axial plane. Then, the imaging slices are placed on top of each other. The detectors measure the value of the absorption of the X-rays passing through the tissue [19,20]. Signals taken from the detectors undergo analytical algorithms and are then transferred to the computer unit. The images are recorded in the computer unit in the Digital Imaging and Communications in Medicine (DICOM) format. DICOM was developed for the communication of medical images and the data related to these images [19].

Cone beam CT (CBCT), also known as dental volumetric tomography, or cone beam ray CT, and is an extra-oral image scanner that can create a 3D image. CBCT was developed in 1998 as an alternative to conventional CT for the head and neck region. The first CBCT unit used was the Tom 9000 (Quantitative Radiology, Verona, Italy). CBCT examination is performed using an X-ray light source surrounding the patient and two-dimensional surface detectors with gantry equipment. Fan-shaped X-rays are used in spiral CT, and cone-shaped X-rays are used in CBCT. From the X-ray source, a single beam moves 360 degrees

Table 1. The Hounsfield unit (HU) values of 14 patients in the group with ectodermal dysplasia and the 14 normal controls.

	Age	Gender	Ectodermal dysplasia group			Control group		
			Right mandibular	Medial mandibular	Left mandibular	Right mandibular	Medial mandibular	Left mandibular
1	10	Male	172	474	279	188	472	263
2	10	Male	420	369	358	339	336	324
3	11	Male	196	252	289	235	281	274
4	13	Male	219	394	294	344	344	410
5	16	Female	269	451	275	467	575	484
6	17	Female	272	338	322	553	455	459
7	17	Female	325	404	241	398	300	429
8	18	Male	380	465	328	317	505	302
9	18	Male	283	751	272	302	684	406
10	18	Female	269	484	339	378	714	481
11	20	Female	340	523	427	290	536	478
12	24	Male	315	389	338	384	754	433
13	30	Female	336	416	378	391	411	450
14	33	Female	266	823	337	423	779	511
	Mean		290.14	466.64	319.78	357.79	510.43	407.43

around the patient, and the detectors form many images for every each degree of movement (>150–600). Different tissue densities are identified in the tissues exposed to X-rays by the different absorption rates of the X-rays. As a result of the CBCT scan, images are formed as raw images, the scout image, and the guide image, from the flat panel detectors or a charged coupled device (CCD) camera for the X-rays passing over the patient. With the computer software, these images are then transformed to the 3D axial, coronal, and sagittal plane volumetric data. This process is known as reconstruction, and the number of images obtained depend on the dimensions of the imaging area [21,22].

CBCT is used in the examination of the paranasal sinuses, in orthognathic surgery, in trauma cases, temporomandibular joint diseases, to evaluate inflammation of the head and neck region, for the evaluation of cysts and tumors, for functional endoscopic sinus surgery, and the examination of the osteomeatal complex and sinuses [23–25]. CBCT is used in the surgical areas of endodontics and orthodontics, in the treatment planning of complex cases that require a multi-disciplinary approach, and in cases requiring evaluation with 3D software [20]. Also, CBCT is useful for the measurement of bone volume, bone area or morphometry, and bone density [26–32]. The HU scale used in CBCT includes numbers between –1000 and +1000, and while fat and air have negative HU values, bone, blood, and soft tissues have positive HU values [19]. HU values corresponding to different tissues are shown in Table 1.

The aim of this retrospective study was to investigate whether or not there was any difference in bone density evaluated by CBCT, measured in HU, in the three regions of the lower mandible, the right mandibular, medial mandibular, and left mandibular regions, between a patient group with ectodermal dysplasia and an age-matched and gender-matched control group.

Material and Methods

Patients and study design

A retrospective study included patients and normal individuals who had undergone three-dimensional (3D) cone beam computed tomography (CBCT) imaging and measurements of bone mineral density (BMD) using Hounsfield units (HUs). Three regions of the mandible were evaluated, the right mandibular, medial mandibular, and left mandibular regions from 9 women and 5 men diagnosed with ectodermal dysplasia and a control group of 9 women and 5 men. The HU values obtained according to age and gender of the total 28 patients were measured

Cone beam computed tomography (CBCT) imaging and measurements of bone mineral density (BMD)

Of the radiological methods to measure BMD, quantitative computed tomography (QCT) was applied using X-rays and CBCT. The BMD result of an area measured by QCT was given

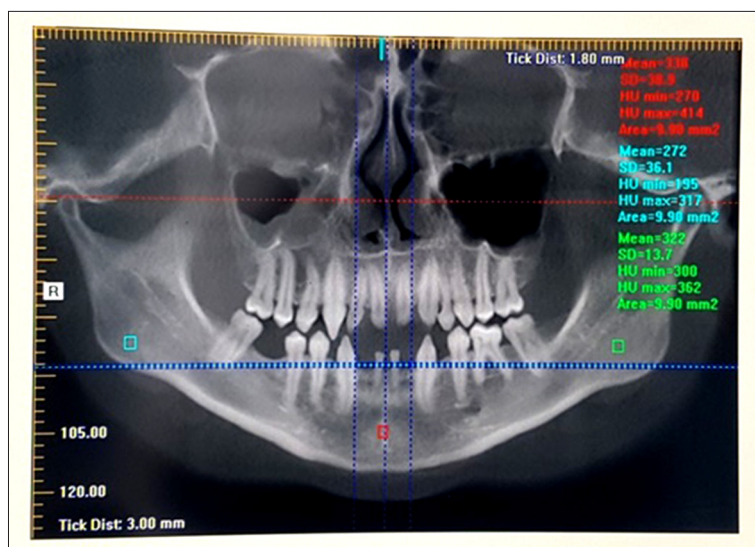


Figure 1. The measurement of the Hounsfield unit (HU) values on a panoramic radiograph in a case of ectodermal dysplasia.

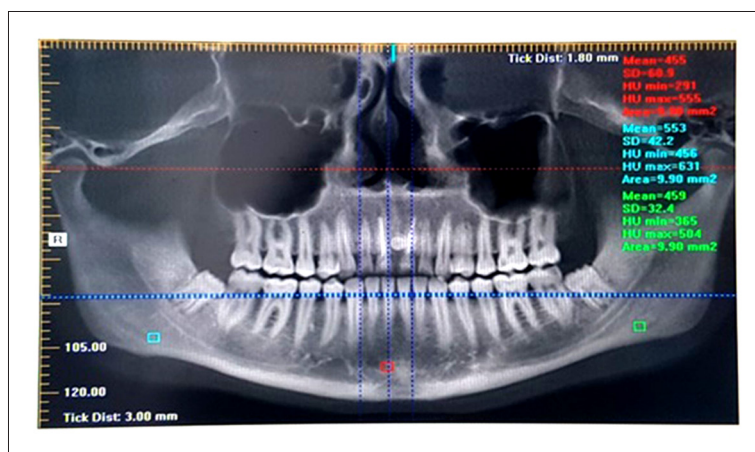


Figure 2. The measurement of the Hounsfield unit (HU) values on a panoramic radiograph in a normal control without ectodermal dysplasia.

as gr/cm^2 . The method of measurement of the HU values on a panoramic radiograph is shown in Figure 1 as an example from the ectodermal dysplasia group, and in Figure 2 as an example of the control group. [19]. Measurement of the HU values of the BMD of the panoramic films was obtained from the standard I-CAT Vision™ program of the CBCT device (Imaging Science International, Hatfield, USA).

3D CBCT images were taken of all patients as gr/cm^2 of a mean 10 mm^2 area with an I-CAT Vision™ unit (Imaging Science International, Hatfield, USA). An image area of 10 mm^2 was selected from the three regions of the lower mandible, the right mandibular, medial mandibular, and left mandibular regions on the panoramic radiographs, with shot parameters of 12 KvP, 5 mA, and 8.9 secs. Each image using the device was made with a single 360-degree rotation around the patient. All the images obtained were recorded using the Digital Imaging and Communications in Medicine (DICOM) format.

Panoramic images of the mandible obtained on tomography for both study groups were included in the analysis. The measurements were made of 10 mm^2 areas consisting of the densest areas of bone from the three separate zones of the mandible, with the densest point including the inferior and lateral edges of the angulus regions and the central anterior region. All measurements were performed in triplicate and the mean measurement was used.

Statistical analysis

The data obtained were analyzed using SPSS version 22.0 software. The normal distribution of data was assessed using the Shapiro-Wilk test. Comparison of the data between the two independent groups was performed for data with a normal distribution using Student's t-test. Descriptive data were presented as the mean \pm standard deviation (SD). A P-value <0.05 was considered to be statistically significant.

Table 2. Comparison of the mean Hounsfield units (HU) values between the 14 patients in the group with ectodermal dysplasia and the 14 normal controls.

	Groups	N	Mean	Standard deviation (SD)
Right mandibular	Ectodermal dysplasia	14	290.1	68.55
	Control	14	357.8	92.78
Medial mandibular	Ectodermal dysplasia	14	466.6	152.3
	Control	14	510.4	170.3
Left mandibular	Ectodermal dysplasia	14	319.79	48.98
	Control	14	407.43	82.891

Table 3. Analysis of the comparison of the Hounsfield unit values between the 14 patients in the group with ectodermal dysplasia and the 14 normal controls, analyzed with the Student's t-test.

Comparison of results between the two groups	Student's t-test	P-value
Right mandibular	-2.194	P<0.05 (significant)
Medial mandibular	-0.717	P>0.05 (non-significant)
Left mandibular	-3.406	P<0.05 (significant)

Results

The results of the bone mineral density (BMD) measurements for both groups, the ectodermal dysplasia, and age-matched and gender-matched controls are shown in Table 1. A retrospective evaluation was made of the tomography images of the mandible of 14 patients with ectodermal dysplasia (9 women and 5 men) and a control group of 14 healthy subjects (9 women and 5 men). The Hounsfield unit (HU) values of the 28 study participants, according to age and gender, are shown in Table 1. The comparisons of the mean HU values of both groups are shown in Table 2. The results of the data analysis with the Student's t-test are shown in Table 3.

BMD measurements (HU) were significantly increased in the control group compared with the patient group with ectodermal dysplasia ($p<0.05$). The results of the comparison of the HU values between the ectodermal dysplasia patient group and the control group are shown in Table 4.

Comparison of the mean HU values of the right mandibular and left mandibular regions of the lower mandible in the ectodermal dysplasia group were significantly different from the control group. No statistically significant difference in the HU values was identified between the two groups for the medial mandibular region.

Discussion

Bone is the hardest structure in the body and is composed of 33% organic material and 67% inorganic minerals and mineral salts. The primary mineral is calcium [36], and mineral salts include calcium phosphate, calcium carbonate, magnesium phosphate and a small amount of sodium and iron. Vitamin D has a role in bone mineralization. The flexibility of bone is provided by the 33% of organic materials formed from cells and protein. The organic structure of bone is formed from collagen fibrils, osteocytes, osteoblasts and osteoclasts [18,33]. Calcium and vitamin D are the two nutritional elements that play a key role in bone health. It is important that a sufficient amount of calcium is obtained in during childhood and adolescence to achieve the highest levels that can be stored in the skeleton. In Turkey, current guidelines recommend that the use of bone mineral density (BMD) measurement is an important component of patient diagnosis and response to treatment [18].

Table 4. Comparison of the total average Hounsfield unit values between the 14 patients in the group with ectodermal dysplasia and the 14 normal controls.

	Groups	N	Mean	Standard deviation	Student's t-test	P-value
Total	Ectodermal dysplasia	14	358.86	125.3	2.334	P<0.05 (significant)
	Control	14	425.21	135.1		

The findings of the present study showed that BMD measurements in Hounsfield units (HUs) were significantly increased in the control group compared with the patient group with ectodermal dysplasia ($p < 0.05$). The most common finding in patients with ectodermal dysplasia is the absence of primary and permanent dentition and so dental prosthetic treatment is recommended during the early years of development for the majority of cases. In dentistry, consideration of the BMD of patients with ectodermal dysplasia can be useful in planning the treatment approach and can have a positive effect on patient prognosis. Cone beam computed tomography (CBCT) imaging is preferred to CT for the measurement of BMD as the radiation dose is lower, the acquisition time is shorter, and there are fewer imaging artifacts [34]. Also, CBCT is recommended for the diagnosis of odontogenic tumors of the oral cavity and in periodontal disease [35,36]. Although the ease of use of CBCT is an advantage, the high costs of the imaging equipment and its use are a disadvantage [34].

To the author's knowledge, this is the first study to evaluate BMD using CBCT imaging in patients with ectodermal dysplasia compared with age-matched and gender-matched controls. Therefore, the results of this preliminary study might be of value in contributing to current knowledge on the diagnosis of ectodermal dysplasia and the evaluation of BMD. However, although this study has demonstrated the value of CBCT in the evaluation of BMD in ectodermal dysplasia, this imaging method is not widely available and patients with ectodermal dysplasia should continue to be evaluated in specialist centers.

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Conclusions

A retrospective study was conducted at a single specialist center in Turkey to compare the measurements of bone mineral density (BMD) from three main regions selected of the lower mandible, the right mandibular, medial mandibular, and left mandibular regions, in patients with ectodermal dysplasia and age-matched and gender-matched controls. Measurement of BMD, in Hounsfield units (HUs), was evaluated using three-dimensional (3D) cone beam computed tomography (CBCT) imaging. The findings showed significantly lower BMD in patients with ectodermal dysplasia compared with the control group and showed that BMD could be measured with CBCT. Comparison of the left and right mandibular angulus regions showed that the BMD of patients with ectodermal dysplasia was significantly lower when compared with the control group in both regions, but no significant difference was found between the two groups in the BMD of the central mandibular region. Reduced BMD in patients with ectodermal dysplasia should be taken into consideration when planning surgical interventions involving bone tissue and when planning implant surgery.

Conflict of interest

None.

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