

# Prevalence of Multiple Miliary Type of Osteoma Cutis in the Maxillofacial Region as an Incidental Finding: A Retrospective Cone Beam Computed Tomography Study

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## ABSTRACT

**Objective:** Miliary osteoma cutis (MOC) is a rare variant of osteoma cutis in which multiple fragments of bone formations are embedded in the skin. In general, they are asymptomatic, benign and detected incidentally on radiographic examination.

**Methods:** This prevalence study was made by using cone beam computed tomography (CBCT) scans. A total of 893 CBCT scans were evaluated. Two hundred and two of them were excluded because of poor diagnostic quality. The occurrence frequency of incidentally found multiple miliary type of osteoma cutis in head and neck area was noted. Median and range were used to describe the age of the patients.

**Results:** A total of 691 CBCT images were screened. Of these, 268 (38.8%) were from female patients and 423 (61.2%) were from male patients. The median age of patients referred for CBCT was 45.0 (IQR = 30.0) years and within the age range of 5 to 84 years. A total of 22 (3.2%) multiple MOC cases in maxillofacial region were discovered on 691 patients' CBCT scans.

**Conclusion:** Calcified lesions in head and neck region were commonly seen in CBCT images. Although the most of the calcifications are asymptomatic and require no treatment but diagnosis should be carefully made to avoid unnecessary further diagnostic assessments. It will also provide the ability comprehensively evaluation of underlying diseases.

**Keywords:** Multiple miliary osteomas, osteoma cutis, prevalence

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## INTRODUCTION

Multiple miliary osteoma cutis (MOC) which is one of four subtypes of osteoma cutis, a rare skin disorder characterized by the bone formation within the dermis or subcutaneous tissue. Osteoma cutis is benign with limited and non-invasive growth as well as its subtypes (1). It may be primary or secondary but secondary form in association with a pre-existing inflammatory skin condition is mostly seen. In the majority of cases it is clinically asymptomatic and may be detected incidentally on radiographic examination (2, 3).

Recently, the use of cone beam computed tomography (CBCT) for various diagnostic purposes is more common in dentistry. There are two major advantages of CBCT compared to 2D radiographic modalities. One of them is elimination of geometric distortion and the second one is reduction of structural superimpositions (4). Additionally, in comparison with computed tomography, CBCT has a lower radiation dose in between 29 and 577  $\mu$ SV. Computed tomography has radiation exposures about 2000  $\mu$ SV (5). It can provide precious diagnostic information not only from the interested area but also the out of the region. The out of interested region findings which is described as incidental findings may sometimes have greater importance of determining an appropriate treatment plan. The purpose of this study was to investigate the characteristics and frequency of multiple MOC which is incidentally detected.

## SUBJECTS AND METHODS

This prevalence study was made by using CBCT scans. Cone beam computed tomography images used in the study were acquired on a 3D Accuitomo 170 (3D Accuitomo; J Morita Mfg. Corp., Kyoto, Japan) which were obtained between 2011-2016. A total of 893 CBCT scans were evaluated and 202 of them were excluded because of poor diagnostic quality of small FOV size. The study sample (n = 691) consisted of CBCT scans of patients who were referred for CBCT

evaluation to the Department of Dentomaxillofacial Radiology, Gulhane Training and Research Hospital, Ankara, Turkey. All CBCT images were evaluated by a dentomaxillofacial radiologist who has 12 years of experience in the basis of multiple miliary type of osteoma cutis which were detected incidentally (Figs. 1–3).

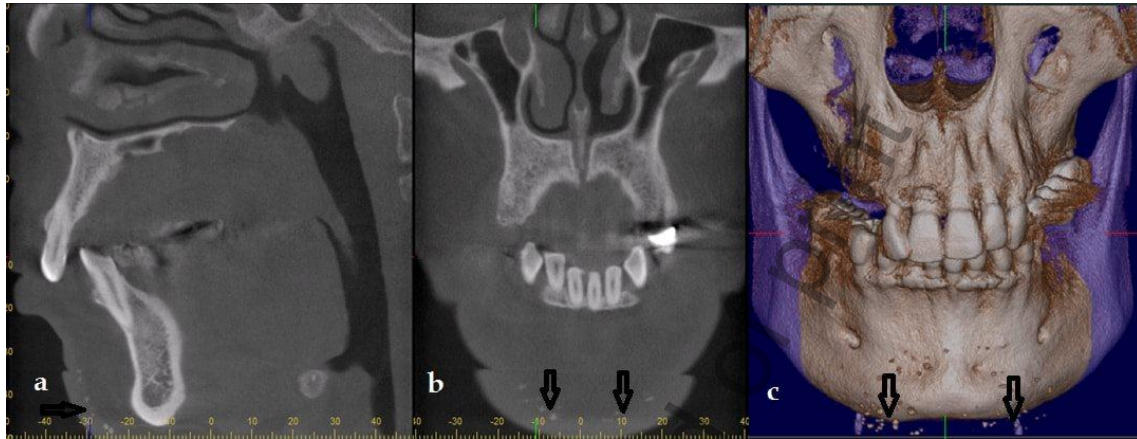


Fig. 1: A 45-year-old female referred for implant rehabilitation. On CBCT examination it was detected multiple dot like radiopaque masses around chin (arrow). (a) Sagittal view. (b) Coronal view. (c) 3D reconstruction.

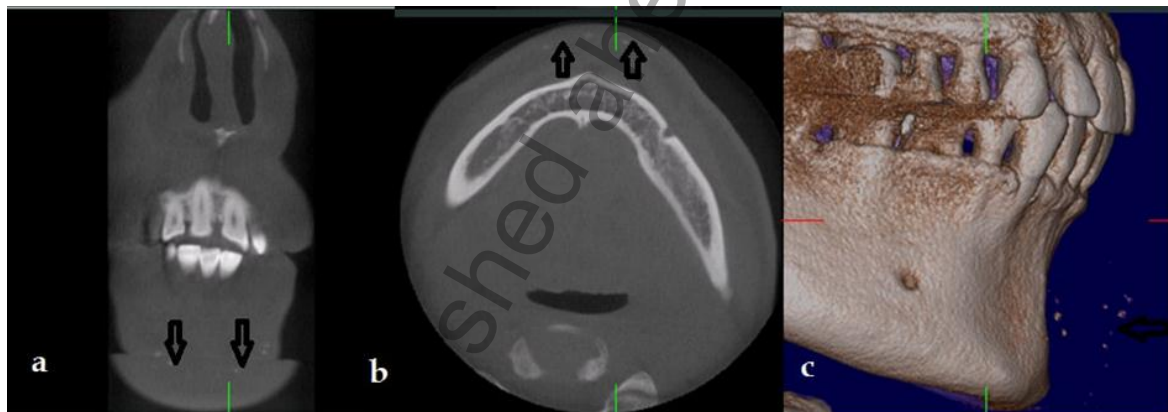


Fig. 2: A CBCT view of a 35-year-old male patient showing multipl radiopaque masses around chin (arrow). (a) Coronal view. (b) Axial view. (c) 3D reconstruction.

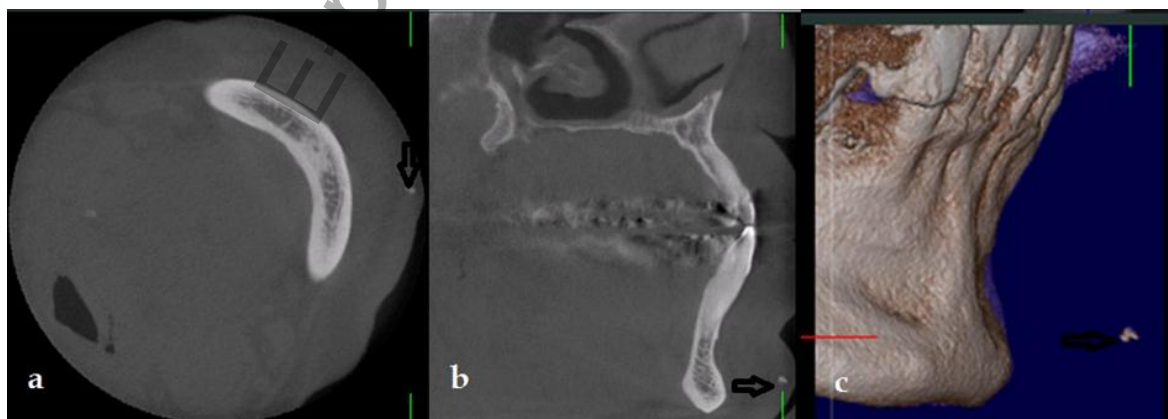


Fig. 3: A CBCT view of a 41-year-old male patient. The arrows point to radiopaque lesion around chin. (a) Axial view. (b) Sagittal view. (c) 3D reconstruction.

For CBCT evaluations, proprietary manufacturer software (i-Dixel 2.0/One Data Viewer/One Volume Viewer; J Morita Mfg. Corp.) was used. Images were viewed in a dimly lit room on a 30 inch Dell™ 3008WFP Flat Panel Monitor (Dell Inc., Round Rock, TX, USA) at a screen resolution of 1920 x 1200 pixels and 32-bit colour depth.

### Statistical analyses

Data were analysed by descriptive statistics. The occurrence frequency of incidentally found multiple miliary type of osteoma cutis in head and neck area was noted. Median and range were used to describe the age of the patients. Statistical analyses were performed using the SPSS software (version 15.0; SPSS Inc., Chicago, IL, USA) and MS Excel 2003.

### RESULTS

A total of 691 CBCT images were screened. Of these, 268 (38.8%) were from female patients, and 423 (61.2%) were from male patients. The median age of patients referred for CBCT was 45.0 (IQR = 30.0) years and within the age range of 5 to 84 years. A total of 22 (3.2%) MMOC cases in maxillofacial region were discovered on 691 patients' CBCT scans. Of 22 patients, 14 (63.6%) were males and 8 (36.4%) were females. A predominance of males was found with a ratio of 14:8 over females. The median age of patients were as 49.6 years and within the age range of 21 to 81.

The age-gender distribution and frequency distribution of the sample with multipl MOC are shown in Table 1. According to Table 1, multipl MOC was seen most frequently at 41–50 (n = 7, 31.8%) and 51–60 age groups (n = 7, 31.8%). The least frequent age group was found as 71–81 (n = 1, 4.5%).

Table 1: The age-gender distribution and frequency distribution of the patients with multiple miliary osteoma cutis

Age Groups	21–30		31–40		41–50		51–60		61–70		71–81	
	M	F	M	F	M	F	M	F	M	F	M	F
Gender	2	-	1	1	4	3	4	3	2	1	1	-
<b>TOTAL</b>	<b>2</b>		<b>2</b>		<b>7</b>		<b>7</b>		<b>3</b>		<b>1</b>	

## DISCUSSION

Osteoma cutis is true bone formation of the skin and is classified as primary and secondary (metaplastic) ossification (6). Secondary osteomas constitute 85% of cutaneous ossifications and develop in preexistent neoplastic or inflammatory skin lesions (7). Primary osteoma cutis accounts for approximately 15% of cutaneous ossifications and develops in itself (8). Classification of osteoma cutis was shown in Table 2.

Table 2: Classification of osteoma cutis (based on 7, 8)

Primary	Secondary
Albright's hereditary osteodystrophy	Not associated with Albright's hereditary osteodystrophy
	1) Inflammatory skin disease
	-Progressive systemic sclerosis and CREST syndrome
	-Dermatomyositis
	-Morphea
	2) Tumors/neoplasms
	- Basal cell carcinoma, pilomatricoma, etc.
	3) Trauma and scars

Principal causes of this condition are progressive osseous heteroplasia, Albright's hereditary osteodystrophy and multiple MOC in the face (6).

First, Virchow in 1864 determined this condition as miliary osteoma cutis (9). In 1928, the role of acne was first suggested by Hopkins in the development of multiple MOC (10).

The pathogenesis of this condition is unknown. Some hypotheses were present about the origin of the cell forming osteoma. Fibroblast metaplasia is the most accepted theory (11, 12). According to *In situ* hybridization techniques, dermal fibroblasts have the talent to differentiate into osteoblasts and to produce collagen Type 1 and osteonectin (11).

There is another hypothesis. According to this hypothesis, embryonic mesenchymal cells, faultily migrated to the dermis, might differentiate into the osteogenic lineage. Gene mutations in syndromes of cutaneous ossification can be one of the reasons (11).

Multiple MOC is an uncommon situation that generally affects middle-aged women associated with a history of severe acne (13). But differently, men were more frequently affected than women in our study.

Miliary osteoma cutis is generally observed as skin coloured papules on the scalp, the face as well as the trunk, the breast, the extremities and the buttocks. The patients who have chronic acne, the differentiation from microcomedones and macrocomedones can be difficult (12).

Miliary osteoma cutis is an infrequent condition, with approximately 50 cases in the literature (11). After an extensive literature search, a few studies that have evaluated the prevalence rate were found about osteoma cutis in the maxillofacial region. According to the study of Safi *et al* (2), 6500 CBCT scans were evaluated and multiple MOC was found in 130 (2%) cases. Kishi *et al* (14) evaluated 2089 individuals and calculated the incidence of multiple miliary osteomas as 2.2%. Similar to our study, Kishi *et al* (14) reported that males were more frequently affected than females. In our study, multiple MOC was observed at the rate of 3.2%. This result is close to the results of Safi *et al* (2) and Kishi *et al* (14). Shigehara *et al* (15) assessed 33 cadavers and

158 living subjects. They detected multiple miliary osteoma in 27.8% (44/158) of the living subjects. Similar to our study, Shigehara *et al* (15) reported that incidence was higher in males than in females and multiple miliary osteomas were seen most frequently at 40–59 age group. Kim *et al* (16) evaluated 1315 consecutive sinus computed tomography scans. Among the total number of male and females, they found in 252 males and 301 females who had small facial calcified nodules (42.1% and 42.0%, respectively). In comparison with our study, these rates was so higher than our study rate. In the study of Shigehara *et al* (15), this higher rate may result from the preference technique. As for the study of Kim *et al* (16), it was hypothesized that this encountered facial calcification represents primary MOC. According to our opinion, this may explain the high result of the study of Kim *et al* (16).

The patient may have esthetic problem because of multipl MOC. Until today, no curative treatment for multipl MOC has been found. However, different treatments with different results have been mentioned in the literature such as oral isotretinoin or tetracycline, the method of needle microincision-extirpation, surgical treatment and carbon dioxide laser (17, 18).

## CONCLUSION

In conclusion, it was decided that CBCT provides more accurate detection of soft tissue calcifications such as osteoma cutis than conventional radiographic methods and additionally provides extensive information for accurate diagnosis. Thus, it assures of the patients. However, the knowledge of calcified lesions improves the extensive evaluation of underlying diseases. It is very important that early diagnosis will save life of the patients.

## AUTHORS' NOTE

K Gunduz conceived paper, oversaw data collection and revision of manuscript and approved final version. G Serindere participated in study design, wrote manuscript and approved final

version. H Avsever participated in study design, data analysis, and interpretation of data and approved final version. K Orhan participated in study design, data analysis, and interpretation of data and approved final version. The authors declare that they have no conflicts of interest.

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