ORIGINAL ARTICLE

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

K Gunduz[1], G Serindere[2], H Avsever[3], K Orhan[4]

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. A total of 202 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 5–84 years. A total of 22 (3.2%) multiple MOC cases in the maxillofacial region were discovered on 691 patients' CBCT scans.*

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

Keywords: Multiple miliary osteomas, osteoma cutis, prevalence.

INTRODUCTION

Multiple miliary osteoma cutis (MOC) 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 a 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 the elimination of geometric distortion and the second one is the reduction of structural superimpositions (4). Additionally, in comparison with computed tomography, CBCT has a lower radiation dose between 29 and 577 μSV. Computed tomography has radiation exposures of about 2000 μSV (5). It can provide precious diagnostic information not only from interested area but also out of the region. The out-of-interested region findings which are described as incidental findings may sometimes have greater importance in determining an appropriate treatment plan. The purpose of this study

From: [1]Department of Dentomaxillofacial Radiology, Faculty of Dentistry, Ondokuz Mayıs University, Samsun, Turkey, [2]Department of Dentomaxillofacial Radiology, Faculty of Dentistry, Hatay Mustafa Kemal University, Hatay, Turkey, [3]Department of Dentomaxillofacial Radiology, Gulhane Faculty of Dentistry, Health Sciences University, Ankara, Turkey and [4]Department of Dentomaxillofacial Radiology, Ankara University, Ankara, Turkey.

Correspondence: Dr G Serindere, Department of Dentomaxillofacial Radiology, Faculty of Dentistry, Hatay Mustafa Kemal University, 31060 Hatay, Turkey. Email: gozdeserindere@mku.edu.tr

 DOI: 10.7727/wimj.2018.030

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. CBCT 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 and 2016. A total of 893 CBCT scans were evaluated and 202 of them were excluded because of the poor diagnostic quality of small field of view 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 Faculty of Dentistry, Health Sciences University, Ankara, Turkey. All CBCT images were evaluated by a dentomaxillofacial radiologist who has 12 years of experience on the basis of multiple miliary type of osteoma cutis, which were detected incidentally (Figs 1–3).

For CBCT evaluations, proprietary manufacturer software (i-Dixel 2.0/One Data Viewer/One Volume Viewer; J Morita Mfg. Corp., Kyoto, Japan) 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 × 1200 pixels and 32-bit colour depth.

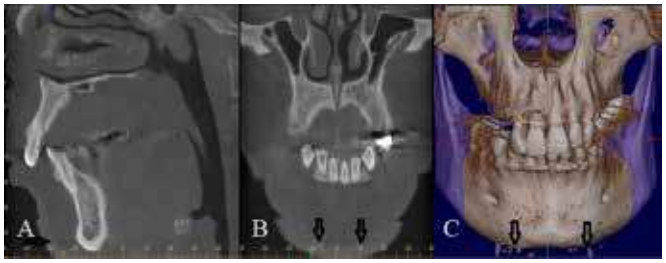

Fig. 1: A 45-year-old female referred for implant rehabilitation. On CBCT examination, multiple dot-like radiopaque masses were detected around the chin (arrow). (A) Sagittal view. (B) Coronal view. (C) Three-dimensional reconstruction.

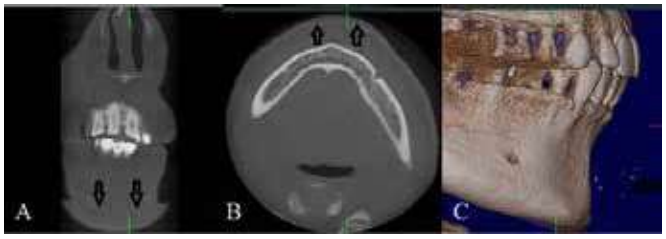

Fig. 2: A CBCT view of a 35-year-old male patient showing multiple radiopaque masses around the chin (arrow). (A) Coronal view. (B) Axial view. (C) Three-dimensional reconstruction.

Statistical analyses

Data were analysed by descriptive statistics. The occurrence frequency of incidentally found multiple MOC 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 5–84 years. A total of 22 (3.2%) multiple MOC cases in the 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 was 49.6 years, within the age range of 21–81.

The age–gender distribution and frequency distribution of the sample with multiple MOC are shown in Table 1. According to Table 1, multiple 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**	
Gender	M	F	M	F	M	F	M	F	M	F	M	F
	2	–	1	1	4	3	4	3	2	1	1	–
Total	2		2		7		7		3		1	

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 pre-existent neoplastic or inflammatory skin lesions (7). Primary

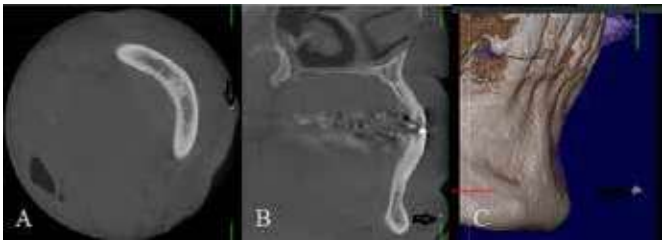

Fig. 3: A CBCT view of a 41-year-old male patient. The arrows point to radiopaque lesion around chin. (A) Axial view. (B) Sagittal view. (C) Three-dimensional reconstruction.

osteoma cutis accounts for approximately 15% of cutaneous ossifications and develops in itself (8). Classification of osteoma cutis is shown in Table 2. 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 MOC (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). However men were more frequently affected than women in our study.

Miliary osteoma cutis is generally observed as skin coloured papules in the scalp, the face as well as the trunk, the breast, the extremities and the buttocks. In patients who have chronic acne, the differentiation between 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 in the 40–59 age group. Kim *et al* (16) evaluated 1315 consecutive sinus computed tomography scans. Among the total number of males 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 were so higher than our study rates. 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 an aesthetic problem because of multiple MOC. Until today, no curative treatment for multiple 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 the patients. However, the knowledge of calcified lesions improves the extensive evaluation of underlying diseases. It is very important that early diagnosis will save the life of the patients.

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

Primary		Secondary
Albright's hereditary osteodystrophy	Not associated with Albright's hereditary osteodystrophy • Multiple miliary osteomas of the face • Isolated osteoma • Widespread osteoma • Congenital plaque-like osteoma	(1) Inflammatory skin disease • Progressive systemic sclerosis and CREST syndrome Dermatomyositis Morphea (2) Tumours/Neoplasms Basal cell carcinoma, pilomatricoma, *etc* (3) Trauma and scars

AUTHORS' NOTE

KG conceived paper, oversaw data collection and revision of manuscript and approved final version. GS participated in study design, wrote manuscript and approved final version. HA participated in study design, data analysis and interpretation of data and approved the final version. KO participated in study design, data analysis and interpretation of data and approved final version. The authors declare that they have no conflicts of interest.

REFERENCES

1. Duarte IG. Multiple injuries of osteoma skin in the face: therapeutical least invasive in patients with acne sequel—case report. An Bras Dermatol 2010; **85:** 695–8.
2. Safi Y, Valizadeh S, Vasegh S, Aghdasi MM, Shamloo N, Azizi Z. Prevalence of osteoma cutis in the maxillofacial region and classification of its radiographic pattern in cone beam CT. Dermatol Online J 2016; **22:** 2.
3. White SC, Pharoah MJ. Oral radiology principles and interpretation. 7th ed. St. Louis: Mosby; 2014: 536–7.
4. Pazera P, Bornstein MM, Pazera A, Sendi P, Katsaros C. Incidental maxillary sinus findings in orthodontic patients: a radiographic analysis using cone-beam computed tomography (CBCT). Orthod Craniofac Res 2011; **14:** 17–24.
5. Cheung T, Oberoi S. Three dimensional assessment of the pharyngeal airway in individuals with non-syndromic cleft lip and palate. PLos One 2012; **7:** 1–5.
6. Freedberg IM, Eisen AZ, Wolff K et al, eds. Fitzpatrick's dermatology in general medicine, 5th ed. McGraw-Hill: New York; 1999.
7. Fazeli P, Harvell J, Jacobs MB. Osteoma cutis (cutaneous ossification). West J Med 1999; **171:** 243–5.
8. Cottoni F, Dell' Orbo C, Quacci D, Tedde G. Primary osteoma cutis: clinical, morphological, and ultrastructural study. Am J Dermatopathol 1993; **15:** 77–81.
9. Virchow R. Dia Krankhafton Geschwulate. Vol. 2. Hirchwald: Berlin, Germany; 1864: 103.
10. Hopkins JG. Multiple miliary osteomas of the skin. Arch Dermatol Syphilol 1928; **18:** 706.
11. Myllylä RM, Haapasaari KM, Palatsi R, Germain-Lee EL, Hägg PM, Ignatius J et al. Multiple miliary osteoma cutis is a distinct disease entity: four case reports and review of the literature. Br J Dermatol 2011; **164:** 544–52.
12. Thielen AM, Stucki L, Braun RP, Masouyé I, Germanier L, Harms M et al. Multiple cutaneous osteomas of the face associated with chronic inflammatory acne. J Eur Acad Dermatol Venereol 2006; **20:** 321–6.
13. Chabra IS, Obagi S. Evaluation and management of multiple miliary osteoma cutis: case series of 11 patients and literature review. Dermatol Surg 2014; **40:** 66–8.
14. Kishi K, Kawahara K, Moriya I, Komatsu H, Sato M, Aono K. Clinical and radiographic study of multiple miliary osteomas of the skin. Dentomaxillofac Radiol 1984; **13:** 105–8.
15. Shigehara H, Honda Y, Kishi K, Sugimoto T. Radiographic and morphologic studies of multiple miliary osteomas of cadaver skin. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1998; **86:** 121–5.
16. Kim D, Franco GA, Shigehara H, Asaumi J, Hildenbrand P. benign miliary osteoma cutis of the face: a common incidental CT finding. Am J Neuroradiol 2017; **38:** 789–94.
17. Wu M, Wang Y, Zhang D, Jia G, Bu W, Fang F et al. A case of giant primary osteoma cutis successfully treated with tissue expansion and surgical excision. Indian J Dermatol Venereol Leprol 2011; **77:** 79–81.
18. Baskan EB, Turan H, Tunali S, Toker SC, Adim SB, Bolca N. Miliary osteoma cutis of the face: treatment with the needle microincision-extirpation method. J Dermatolog Treat 2007; **18:** 252–4.