



# An Unusual Giant Osteoma of the Maxillary Sinus: Case Report

E. Kalfarentzos<sup>1</sup> · I. Arapi<sup>2</sup> · V. Triantafyllou<sup>3</sup> · K. I. Tosios<sup>4</sup> · P. Christopoulos<sup>5</sup>

Accepted: 6 April 2020  
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## Abstract

Paranasal sinus osteoma is the most common benign tumour of the paranasal sinuses. Osteomas are described as giant when they measure more than 3 cm in diameter or weigh over 110 g. Giant osteomas of the maxillary sinus are extremely rare. Aetiology is unknown, but there are three theories about the causes, ‘The Developmental theory’, ‘The Trauma Theory’ and ‘The Infection Theory’. Osteomas are often noticed incidentally in radiological examinations, unless they are symptomatic. Symptomatic osteomas or/and osteomas occupying more than 50% of the sinus volume are treated surgically. An intraoral approach is often preferred for paranasal sinus osteomas. These tumours have also been associated with Gardner’s syndrome. No malignancy transformation has ever been reported and they have an overall excellent prognosis. In this paper, we describe a case of a 37-year-old woman, with a giant osteoma in the left maxillary sinus. The presence of osseous septums attaching the tumour to the walls of the maxillary sinus, creating a star-shaped configuration, has been previously described in only one case.

**Keywords** Osteoma · Giant osteoma · Maxillary sinus · Surgery

This article is part of the Topical Collection on *Surgery*

✉ I. Arapi  
ioulia.arapi@hotmail.com

E. Kalfarentzos  
evagelosk@gmail.com

V. Triantafyllou  
vastrian@hotmail.com

K. I. Tosios  
ktosios@dent.uoa.gr

P. Christopoulos  
panos@dentomfs.gr

- <sup>1</sup> Department of Oral and Maxillofacial Surgery, Dental School, University of Athens, Mediterraneo Hospital 8-12 Ilias Str, 16675, Glyfada, Athens, Greece
- <sup>2</sup> Dental School, University of Athens, 2 Thivon Str, 115 27, Goudi, Athens, Greece
- <sup>3</sup> Athens, Greece
- <sup>4</sup> Department of Oral Pathology, Dental School, University of Athens, 2 Thivon Str., 115 27, Goudi, Athens, Greece
- <sup>5</sup> Department of Oral and Maxillofacial Surgery, Dental School University of Athens, Mediterraneo Hospital 8-12 Ilias Str, 166 75, Glyfada, Athens, Greece

## Introduction

Osteomas are described as slowly developing, excessive growths of mature bone, located almost exclusively in bones with intramembranous ossification, such as the skull bones. Although the aetiology is unknown, there are three theories about the causes. ‘The Developmental Theory’ refers to the growth of osteomas between two different embryogenic originated tissues, ‘The Trauma Theory’ correlates osteomas with a head injury in puberty, when bone generation rhythm is higher and ‘The Infection Theory’ describes primary infectious stimulus prior to an osteoma formation [1].

As they usually originate from the wall of a sinus, paranasal sinus osteomas have an estimated incidence of 3%, as detected on radiological examination in patients with a range of sinonasal symptoms [1]. Paranasal sinus osteomas are the most common benign tumour of the paranasal sinuses, commonly located in the frontal and ethmoid and rarely in maxillary and sphenoid sinuses [2, 3]. They show a predilection for middle-aged males and measure from 0.2 to 3 cm [1, 4, 5]. Osteomas are described as giant when they measure more than 3 cm in diameter or weigh over 110 g [5]. Giant osteomas may cause symptoms [2]. An unusual case of a giant, star-shaped osteoma incorporating cholesterol clefts and associated with the root of an endodontically treated tooth, along with the therapeutical management of the patient and the follow-up, is described.

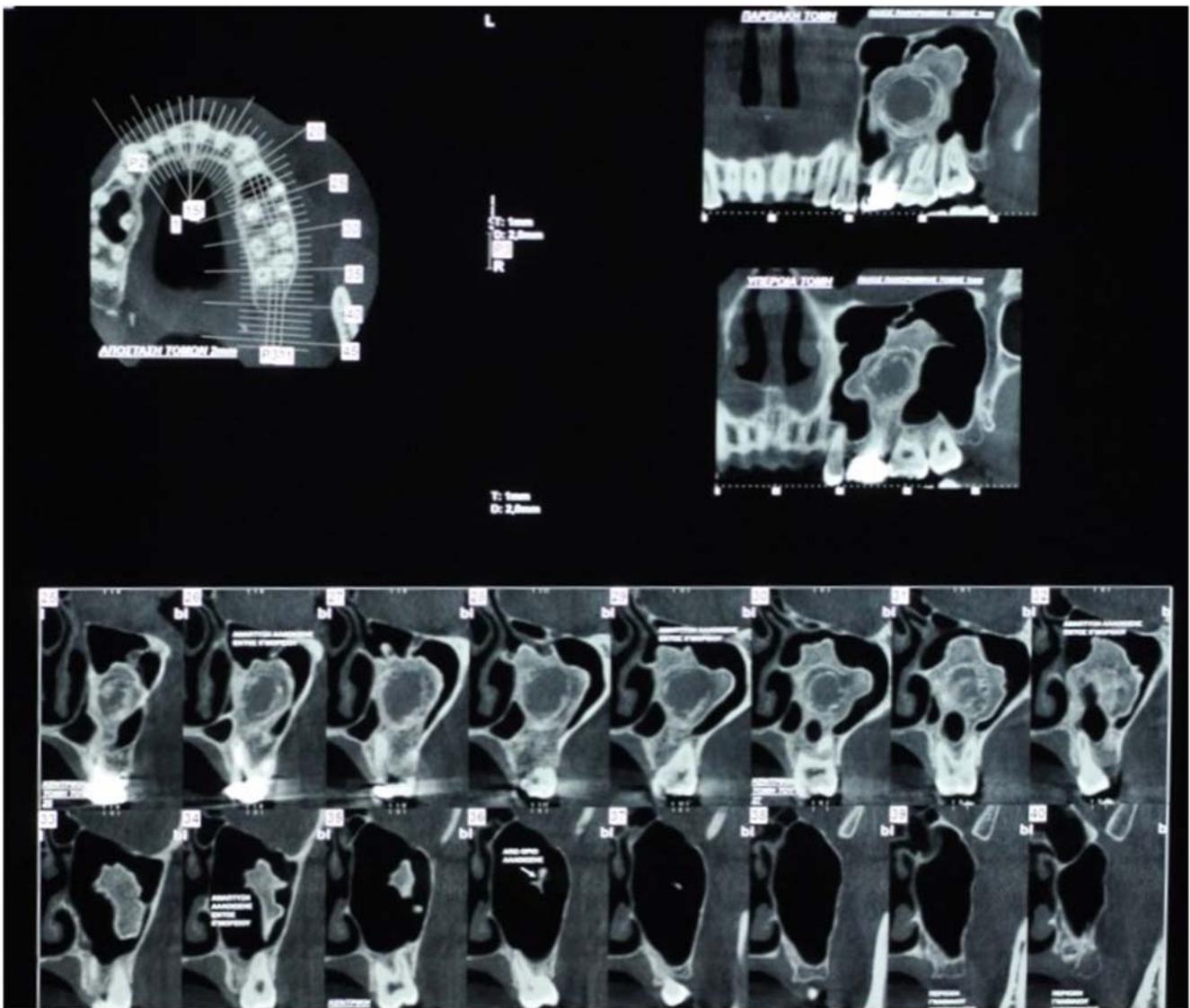


Fig. 1 Preoperative CBCT in order to define dimensions and extension of the lesion. Frontal and vertical planes



Fig. 2 Intraoperative picture showing the centre of the lesion, containing soft tissue

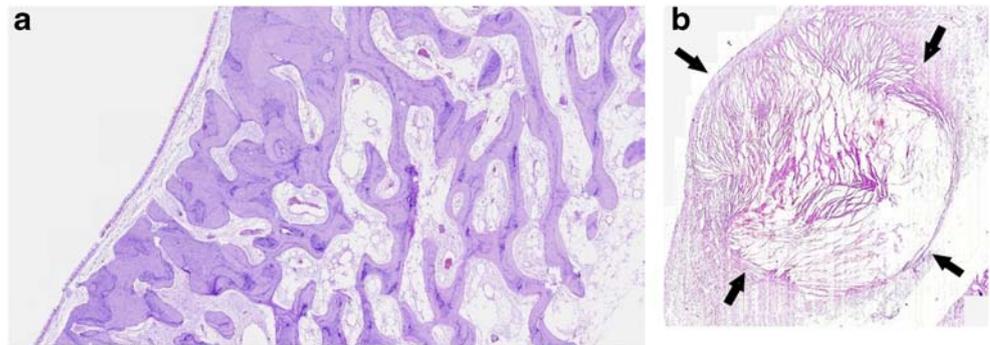
### Case Report

A 37-year-old woman was referred to our outpatient clinic because of a tumour in the left maxillary sinus, incidentally found in a panoramic radiograph. The patient did not



Fig. 3 Postoperative orthopantomogram at the 1-year follow-up

**Fig. 4** **a** Histological section of the peripheral lamellar bone (haematoxylin and eosin stain, original magnification  $\times 25$ ). **b** The central tissue is composed of fibrous connective tissue with numerous cholesterol clefts, delineated by arrows (haematoxylin and eosin stain, original magnification  $\times 25$ )



report any symptoms and her medical history was not remarkable.

Extraoral and intraoral examination was within normal limits. The panoramic radiograph showed an oval, mixed radiopaque/radiolucent lesion with well-defined borders, located apically to the upper left first molar that was endodontically treated. A cone beam computed tomogram (NewTom Giano, 90 kVp, 125  $\mu$  voxel size, 8x8cm field of view) revealed a low density, mixed osteoplastic and osteolytic osseous lesion, occupying approximately 3/4 of the left sinus volume (Fig. 1). In the centre of the lesion, a radiolucent content was evident, resembling soft tissue or cyst. As the lesion was attached with four thin osseous septums to the walls of the maxillary sinus, it had a star-shaped appearance.

With a clinical diagnosis of giant osteoma, the tumour was surgically removed through a Caldwell–Luc approach, under general anaesthesia. On partitioning, a cavity containing soft tissue was found in its centre (Fig. 2). A frozen section biopsy was negative for malignancy. The tumour specimens were fixed in 10% buffered formalin and submitted for microscopic examination. Panoramic radiographs at the 6-month and the 1-year follow-up (Fig. 3) examinations showed no recurrence of the tumour.

Macroscopically, after realigning the four parts of the osteoma, the tumour measured approximately 4  $\times$  3  $\times$  3 cm. They were white, bony hard, and their surface was smooth with four discrete, ray-like extensions. A cavity was seen in the centre of the structure and its soft shiny content was removed for separate examination. The hard tissue specimens were decalcified in an EDTA-based solution. Haematoxylin and eosin stain sections revealed that the tumour was composed of lamellar bone with fibrofatty marrow, covered by respiratory epithelium (Fig. 4a). The central tissue was composed of fibrous connective tissue with numerous cholesterol clefts and osteoid spicules (Fig. 4b). The final diagnosis was consistent with a ‘peripheral sinus osteoma, cancellous type’.

## Discussion

The osteoma described in this paper is unusual, as it developed in the maxillary sinus, was of giant size and had a star-like

configuration with cholesterol clefts in its centre, while it was associated with an endodontically treated first molar. According to Cheng et al., giant osteomas of the maxillary sinus are extremely rare, with only few cases reported in the literature [5]. The presence of osseous septums attaching the tumour to the walls of the maxillary sinus, creating the star-shaped configuration, has been previously described in only one case [6].

Osteoma presents as well-circumscribed radiopaque mass with broad base [7], corresponding to a sclerotic centre. However, in our case, the predominant feature found in the centre of the lesion was the presence of numerous cholesterol clefts, corresponding cholesterol crystals. Cholesterol clefts are commonly found in apical periodontitis, where they are thought to be derived from disintegrating cells or the circulating plasma lipid, and their accumulation causes a non-resolving chronic inflammation [8]. In the presented case, the lesion was associated with the root of an endodontically treated molar, where apical periodontitis could have been developed. Inflammation has been suggested in the pathogenesis of some osteomas, where apical periodontitis could have been the inflammatory stimulus [1, 3].

Symptomatic osteomas or/and osteomas occupying more than 50% of the sinus’ volume are treated surgically by open, endoscopic or combined process, depending on their dimensions, extension and localization [9]. An intraoral approach is preferred for paranasal sinus osteomas, mainly with Caldwell–Luc technique for better surgical visualization [9]. In our case, sectioning of the tumour allowed its removal through the window of the Caldwell–Luc approach, with minimal normal tissue sacrifice.

Osteomas have an excellent prognosis. However, it should be kept in mind that multiple osteomas may be a usual extra-intestinal manifestation of Gardner’s syndrome [10].

## Compliance with Ethical Standards

**Conflict of Interest** Dr. Kalfarentzos declares that he has no conflict of interest. Mrs. Arapi declares that she has no conflict of interest. Mrs. Triantafyllou declares that she has no conflict of interest. Prof. Tosios declares that he has no conflicts of interest. Prof. Christopoulos declares that he has no conflicts of interest.

**Ethical Approval** This article does not contain any studies with human participants or animals performed by any of the authors.

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