**ORIGINAL RESEARCH**

# Bony hard swelling in mandible – osteoma – A systematic review

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**ABSTRACT**

**Introduction:** Rarity in region of oral and maxillofacial perspective accounts to osteomas. Bone of well differentiated nature in conjunction with benign and asymptomatic condition target osteoma, a neoplasm. Varieties are that of extra skeletal, periphery and central. Oral and maxillofacial region exhibit central and peripheral osteoma. Main sites of predilection in case of peripheral types are maxillary, ethmoid and frontal. **Materials and methods:** A detailed literature search was done pertaining to mandibular osteoma patients. Inclusion criteria include various studies done on the above based topics. **Results:** Grand total of 200 cases were identified from 100 papers published in English language literature. Of these 100, 59 were filtered narrowing down to 41 fully downloaded studies pertaining to the topic. **Conclusion:** Perspective from an oral maxillofacial physician, oral maxillofacial radiologist, oral maxillofacial pathologist and oral maxillofacial surgeon should be done in an exceptional way in patients with mandibular osteoma in order to achieve long-term success.

**Keywords:** oral and maxillofacial, pathology, medicine, radiology, surgery, osteoma, mandible

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## INTRODUCTION

Benign lesions can emerge from a diverse array of tissues within both the maxilla and mandible. In the context of odontogenic lesions, their genesis may stem from the tooth-forming epithelium, mesenchymal tissue, or a combination of both.(1) The mandible, specifically, sees the origination of odontogenic lesions predominantly situated superior to the mandibular canal.Notably, neural and vascular lesions frequently find their origins within the confines of the mandibular canal. Conversely, lesions with their epicentre positioned below the inferior alveolar canal typically trace their roots to nonodontogenic sources. This nuanced spatial distinction aids in differentiating between the odontogenic and nonodontogenic origins of lesions within the intricate anatomy of the mandible.(2)Osteomas exhibit a distinctive slow-growing nature and are unequivocally classified as benign jaw lesions, stemming from nonodontogenic sources. These lesions manifest as benign, gradually evolving formations composed of mature, well-differentiated bone. The demographic affected by osteomas spans a broad age range, with a slight prevalence among males and a typical diagnostic age falling between 40 and 65 years. This information underscores the unique characteristics and demographic patterns associated with osteomas, contributing to a more comprehensive understanding of these nonodontogenic jaw lesions.(2,3)The bulk of them are located in the craniofacial bones, particularly in the paranasal sinuses, however they can be found in various parts of the skeleton. Known as exostoses, they are most frequently found growing on the outside layer of bones (peripheral), and include mandibular and maxillary tori. Since they seldom or very slowly produce enlargement, those that originate inside the medullary area are referred to as enostoses, or central osteomas, and are thought to be distinct from peripheral osteomas. They frequently have radiological similarities with regions of localized bone sclerosis or dense bone islands (DBIs).(3)



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## MATERIALS AND METHODS

A comprehensive research was done. Articles from beginning to till date are considered. The literature databases included were pubmed, web of science, google scholar, scopus, medline followed by cross references. Keywords included pathology, oral, dental, lesions. Multi journals involving oral and maxillofacial surgery, oral and maxillofacial pathology, oral maxillofacial radiology and oral and maxillofacial medicines were included. Literatures in English language which are fully available were included. The important points include publication date, author name, journal name, date of issue and keypoints.

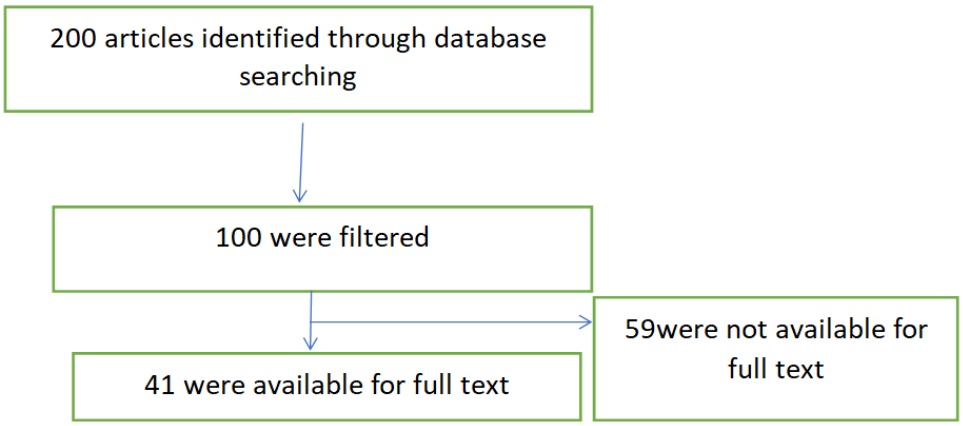


**Figure 1 - Flowchart of literature search**

## RESULTS

The vast literature search was ended up in 41 published articles which are fully downloaded in English from various databases. The universal language of science is English. In order to avoid biasing and erroneous decisions, other languages were excluded. Duplicate articles were removed. Articles which were not able to fully download were removed. Key areas included oral and maxillofacial pathology, oral and maxillofacial surgery, oral and maxillofacial radiology,oral and maxillofacial medicine, etc.

| S.No | Literature | Author | Year | Inference |
|---|---|---|---|---|
| 1 | American Journal of Roentgenology. | Cakir et al | 2011 | Differential diagnosis |
| 2 | Dental Clinics of North America | Gohel et al | 2016 | Imaging |
| 3 | Head and neck pathol | Saha et al | 2019 | Sino orbital region |
| 4 | J Clin Med | Tarsitano et al | 2021 | Reconstructive surgery |
| 5 | Journal of Craniofacial Surgery | Bulut et al | 2010 | Central osteoma |
| 6 | Journal of Clinical Imaging Science. | Panjwani et al | 2011 | Gardner syndrome |
| 7 | J Oral Pathology Medicine | Oyarbide et al | 2008 | Craniofacial osteoma |
| 8 | Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. | Kaplan et al | 2008 | Solitary central osteoma |
| 9 | J Craniomaxillofac Surg. | Kerckhaert et al | 2005 | Giant osteoma |
| 10 | Am J Hum Genet. | Gardner EJ | 1951 | Gardners syndrome |
| 11 | Am J Med Genet. | Gorlin et al | 1992 | Syndromes |
| 12 | Journal of Oral and Maxillofacial Surgery. | Lew et al | 1999 | Condylar osteoma |
| 13 | Dentomaxillofacial Radiology. | Fonseca et al | 2007 | Gardner syndrome |
| 14 | Journal of Cranio-Maxillofacial Surgery. | Kamel et al | 2009 | Gardner syndrome |
| 15 | J Oral Maxillofac Surg. | Sayan et al | 2002 | Peripheral osteoma |
| 16 | J Maxillofac Oral Surg. | Raghupathy et al | 2015 | Peripheral osteoma |
| 17 | J Maxillofac Oral Surg. 2015 | Halawi et al | 2013 | Craniofacial osteoma |
| 18 | Oral Surgery, Oral Medicine, Oral Pathology. | Cutilli BJ, Quinn PD. | 1992 | Peripheral osteoma |
| 19 | J Oral Maxillofac Surg. 1998 | Bodner et al | 1998 | Peripheral osteoma |
| 20 | American Journal of Rhinology. | Chiu et al | 2005 | Frontal sinus osteomas |
| 21 | British Journal of Plastic Surgery. | Gibson T, Walker FM | 1951 | Frontal sinus osteomas |
| 22 | J Oral MaxillofacPathol. | Yadalam et al | 2020 | Compact osteoma |



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|---|---|---|---|---|
| 23 | Archives of Otolaryngology - Head and Neck Surgery | Yamasoba et al | 1990 | Middle ear osteoma |
| 24 | Revue de Stomatologie et de Chirurgie Maxillo-faciale | Caufourier et al | 2009 | Craniofacial osteoma |
| 25 | OJMI | Nnah et al | 2019 | Comparision |
| 26 | Journal of Surgical Case Reports | Tan et al | 2020 | Retromastoid osteoma |
| 27 | BMJ Case Rep. | Nilesh et al | 2020 | Condylar osteoma |
| 28 | BMJ Case Rep. | Ortega et al | 2021 | Mandible |
| 29 | Case Reports in Dentistry. | Demircan et al | 2020 | Mandible |
| 30 | Journal of Stomatology, Oral and Maxillofacial Surgery. | Ghita et al | 2021 | Central compact osteoma |
| 31 | BMJ Case Rep. | Nayak et al | 2020 | Peripheral osteoma |
| 32 | Journal of Oral and Maxillofacial Surgery. | Lazar A, Brookes CCD. | 2021 | Giant osteoma |
| 33 | Eur J Radiol. | Cerase A, Priolo F. | 1998 | Skeletal |
| 34 | European Radiology. | Woertler K. | 2003 | Benign |
| 35 | Journal of Oral and Maxillofacial Surgery. | Angelopoulos C | 2008 | Mandible |
| 36 | Journal of Cranio-Maxillofacial Surgery. | Tarsitano et al | 2018 | Mandible |
| 37 | RadioGraphics | Cure et al | 2012 | Differential diagnosis |
| 38 | Int J Oral MaxillofacSurg | Furlenato et al | 2004 | Osteoma of zygomatic arch osteoma |
| 39 | Journal of Oral and Maxillofacial Surgery. | Longo et al | 2001 | Solitary osteoma |
| 40 | Journal of Cranio-Maxillofacial Surgery. | Ciocca et al | 2015 | Mandible |
| 41 | Journal of Cranio-Maxillofacial Surgery. | Tarsitano et al | 2016 | Mandible |

**Table 1 – An overview**

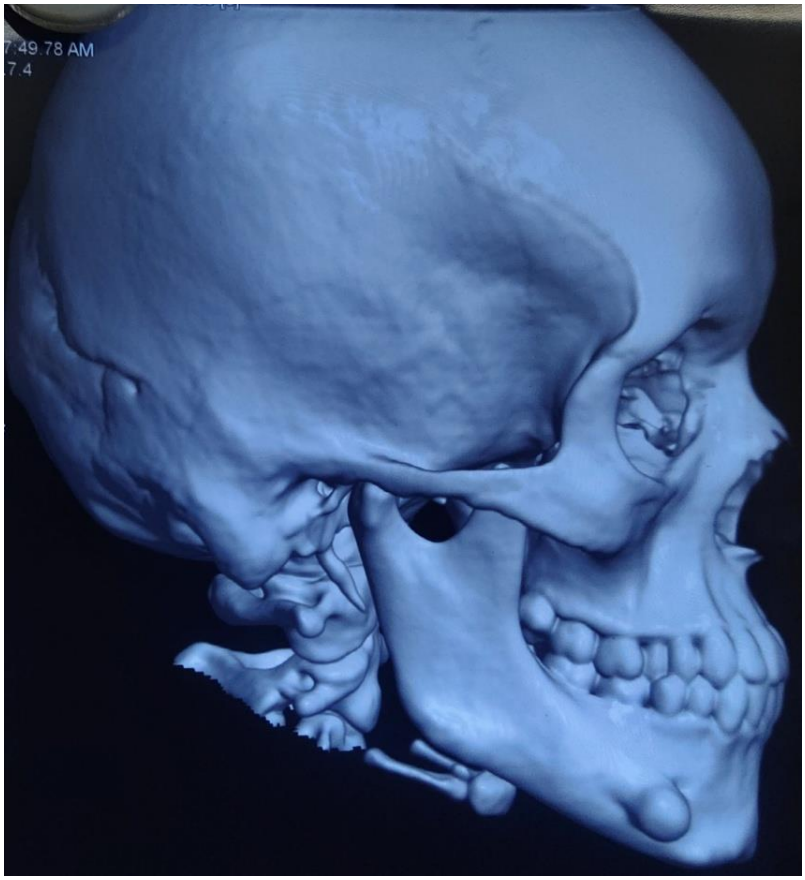


**Figure 2 – Mandibular osteoma (Picture Courtesy – Dr Karthik Shunmugavelu)**



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## CLASSIFICATION

Osteomas, a distinct class of lesions, are categorized based on their location and structural attributes. Depending on the location, three distinct subtypes of osteomas are identified:Central osteomas are characterized by a progressive endosseous development, ultimately leading to the complete replacement of the affected bone segment, Peripheral osteomas are defined by periosteal development, presenting as a pedunculated mass and Extraosseous osteomas refer to those that develop within soft tissues, particularly in the muscles.(4) Characterized by well-defined uniform radiopacities, osteomas can exhibit an exophytic growth pattern. Their presence in the sinuses adds another dimension to their diagnostic characteristics. These rare osteogenic lesions, considered genuine neoplasms, are distinguished by the growth of cancellous and/or compact bone and can be extra skeletal, peripheral, or central in nature (5).Notably, the identification of multiple osteomas raises concern for Gardner's syndrome, a condition distinguished by the concurrent development of multiple osteomas, epidermoid cysts, and intestinal polyps, which exhibit a propensity for malignancy. It is noteworthy that osteomas may precede the emergence of asymptomatic intestinal polyps, underlining the importance of their early recognition in clinical assessments.(2,6)In contrast, vascular lesions such as central hemangiomas and arteriovenous malformations predominantly localize in the posterior mandible, presenting as either unilocular or multilocular lesions. Characterized by a considerable marrow space and coarse trabeculation, these lesions, when centered within the canal, can induce its enlargement, along with a potential enlargement of the mental foramen. The erosive impact on the surrounding bone further complicates the clinical presentation.(2)Furthermore, it is crucial to note that calcifications, identified as phleboliths, are frequently observed in the context of venous malformations within the adjacent soft tissue. This nuanced understanding of radiographic features provides valuable insights for healthcare professionals, facilitating accurate diagnosis and timely intervention. Clinicians should maintain a high index of suspicion when encountering such radiological findings, especially when considering the diverse array of pathologies that can manifest in the maxillofacial region. (2)

## CENTRAL-OSTEOMA

In contrast to their peripheral counterparts originating from the endosteum, solitary central osteomas are notably less prevalent and pose substantial challenges in terms of accurate diagnosis. The scarcity of well-documented, non-syndromic instances of central osteomas, totalling only 11 cases, complicates the differentiation of their characteristics and behaviour in comparison to peripheral osteomas (5,7,8). Despite frequently presenting with asymptomatic features, central osteomas can exert local pressure effects, especially when involving the paranasal sinuses, leading to discomfort, headaches, and deformities. Additionally, in rare instances, bone islands may induce root resorption, further complicating the diagnostic process as they could be mistakenly identified as osteomas.(3)Given that central osteomas essentially represent a phenomenon of "bone within bone," we propose that the diagnosis of a central osteoma requires indications of expansion, displacement, or at the very least, discernible signs of ongoing growth. It is crucial to differentiate central osteomas from other lesions, like idiopathic osteosclerosis or condensing osteitis, which may also manifest as central radiopaque masses. These alternative lesions share microscopic characteristics with osteomas, being composed of cancellous or dense bone, thus rendering them indistinguishable from osteomas under microscopic examination. (5)

## PERIPHERAL OSTEOMA

In most cases, peripheral osteomas appear as hard, radiopaque, mushroom-shaped masses. They are frequently edunculated, but they can also have a wide base that connects them to the cortical plates. Although they often have a limited capacity for development, if left untreated, they will continue to grow slowly. This is not always the case; a small number of documented cases of peripheral osteomas have grown to remarkably great dimensions, being referred to as gigantic, gigantiform, and other comparable terms. (5,9)

## GARDNER'S SYNDROME

Eldon J. Gardner (1909–1989), distinguished as a genetics professor, first delineated Gardner's syndrome in 1951. This syndrome is a rare autosomal, dominant, and highly penetrant inherited disorder, exhibiting a distinctive triad of clinical features encompassing multiple osteomas, colonic polyposis, and mesenchymal tumors affecting the skin and soft tissues. It is a variation of familial adenomatous polyposis syndrome, which is known to be brought on by a mutation in the chromosome 5q21 (band q21 on chromosome 5) Adenomatous Polyposis Coli (APC) gene. (6,10). The diagnosis of Gardner's syndrome requires the presence of osteomas. Although the mandible is the most common site, osteomas can also develop in the long bones, paranasal sinuses, and skull.(11) The mandibular angle and inferior surface are the traditional sites of osteomas in the mandible. Osteomas may be useful indicators of the condition since they occur before the clinical and radiographic signs of colonic polyposis or Gardner's syndrome. About 90% of people with Gardner's syndrome have skeletal abnormalities, with osteoma thought to be the most prevalent (12). Thirty percent of the patients have impacted teeth, complex odontomes, and/or supernumerary teeth (6).Gardner's syndrome can be identified early with the use of panoramic radiography



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by the dentist, since it allows for the diagnosis of the entity's constituent parts, including impacted teeth, osteomas, odontomas, and supernumerary teeth. However, when taking into account the superimposition of the two-dimensional picture and the bone structures, panoramic radiography is not very useful in assessing, localizing, and extending the tumour mass. (6,13). The syndrome's maxillofacial characteristics may manifest years before the intestinal polyposis (6,13). As a result, dental practitioners need to understand the importance of the syndrome as a risk factor for cancer. Dental care comprises resection of osteomas for aesthetic or functional purposes, as well as extraction of impacted teeth and cysts of the jaw or face. Due to the total lack of periodontal space brought on by hypercementosis and the widespread increase in alveolar bone density, tooth extraction might be challenging (6,14).

**AETIOLOGY**

The aetiology of paediatric mandibular osteomas is enigmatic, commonly demonstrating a sporadic nature rather than a hereditary predisposition. While specific cases may show associations with genetic conditions, the predominant consensus attributes the majority to spontaneous emergence. Inflammatory processes or traumatic injuries constitute the primary causes, with prevailing theories suggesting a response to stress or infection. (15,16). Occlusal trauma has been reported as a causative factor for the development of osteoma in a patient.(5) In certain instances where the cause remains unclear, there is a potential link to disorders such as Gardner's syndrome. (5)The mandible exhibits a higher frequency of involvement, with the predominant sites being the lingual aspect of the body, the angle, and the inferior border of the mandible. (5)

**PATHOGENESIS**

The pathogenesis of osteomas remains a subject of ongoing debate, with various onset sites documented in the literature, including the frontoethmoidal junction or the temporal bone, where osteomas may be linked to congenital cholesteatoma (17,18). Consequently, some authors propose a congenital origin for osteomas, positing their development from an embryonal cartilaginous rest or a persistent embryological periosteum (7). The observed association between osteomas and colonic diseases, such as Gardner’s syndrome, raises the possibility of a hereditary nature . Conversely, some of the more common sites for osteoma onset are prone to trauma (e.g., the frontal bone or the angle and lower border of the mandible), suggesting that prior trauma may contribute to the formation of these tumours. The intricate molecular and cellular processes orchestrating their development in paediatric patients necessitate further in-depth investigation.(4)

**HISTOLOGICAL FINDINGS**

Histologically, an osteoma is characterized as an accumulation of abnormal dense bone, with possible origins from the periosteum or bone marrow. This distinction leads to the classification of two types of osteomas: (1) compact or "ivory," and (2) cancellous, trabecular, or spongy, delineating their structural characteristics.(4,5,19)Compact osteomas, commonly referred to as "ivory," are comprised of mature lamellar bone characterized by minimal marrow spaces and occasional haversian canals, devoid of any fibrous structure. Conversely, trabecular osteomas, often termed "mature," consist of cancellous trabecular bone with bone marrow enclosed by a cortical bone margin, encompassing osteoblasts and exhibiting an architectural resemblance to mature bone. (4,5,15). The concept of "zonation of histology" is referenced in certain studies, delineating two distinct regions within osteomas: a fibrous central area, abundant in osteoblasts and blood vessels, actively undergoing growth from the centre to the periphery, and a peripheral area that is less vascularized and metabolically active (20). This divergence justifies the potential consideration of a partial resection limited to the proliferative centre of the lesion to impede its growth. However, it is noteworthy that the literature does document cases of recurrence following partial treatment (4,21).

**PREVALENCE AND EPIDEMIOLOGY:**

The precise incidence of osteomas is challenging to ascertain due to their often small and asymptomatic nature. Estimated to range from 0.002 to 3%, these lesions exhibit a predilection for young males, particularly in the age group of 15 to 30 years (15). While osteomas are commonly noticed in individuals in their sixth decade of life, reports suggest a wider age distribution spanning from 16 to 74 years (5,9). Notably, males are affected at twice the frequency of females (5,15,22). Paediatric mandibular osteomas are deemed rare, and their prevalence within the broader population lacks comprehensive documentation. Incidence patterns may exhibit a predilection for specific age groups, while gender-specific tendencies could also be discernible.

**CLINICAL PRESENTATION**

In the majority of instances, osteomas exhibit an asymptomatic course, with diagnosis typically occurring incidentally during radiological investigations conducted for unrelated reasons. However, in rare cases, osteomas can attain considerable dimensions, leading to aesthetic and/or functional issues resulting from bone distortion and potential compression of nearby structures. The clinical manifestations of craniofacial osteomas display high variability depending on the sites of onset (4,20,23).The jaw and the paranasal sinuses, including the frontal, ethmoid, maxillary, and sphenoid sinuses, represent the most common sites of



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osteoma occurrence. Subsequently, the internal and external cranial planking, along with the maxillary bone, are also reported as sites, with comparatively lesser frequent onset. (4)

When the paranasal sinuses are affected by osteomas, these lesions can occupy the ostiomeatal complex, leading to the disruption of mucus drainage and airflow. Clinically, this can manifest as sinusitis, pain, headaches, and nasal obstruction (23). In instances where osteomas involve the midface, facial asymmetry may occur(4,8,24,25). Moreover, localization of an osteoma within the orbit has the potential to result in exophthalmos. (4).When osteomas affect the mandibular condyle, their growth can lead to a range of dysfunctions. This may include malocclusion, functional impairment of the temporomandibular joint (TMJ), restricted mouth opening due to ankylosis, and in rare instances, symptoms such as tinnitus and deafness (26,27).

Ortega et al. documented a case of mandibular osteoma leading to temporomandibular joint ankylosis (28). Demircan reported an instance of osteoma in the mandibular ramus, causing swelling and facial asymmetry in a 17-year-old male (29). Nilesh et al. reported a case of osteoma in the mandibular condyle, resulting in restricted mouth opening (27). Ghita et al. described a case of facial swelling attributed to an osteoma in the posterior mandibular region, with a similar presentation reported by Torres et al. in a 21-year-old male (30). Nayak et al. reported a patient presenting with swelling in the lower left back tooth region due to a posterior mandibular osteoma (31). Lazar et al. documented a case involving swelling and airway deviation (32).In the presence of multiple facial lesions, it is recommended to conduct a comprehensive assessment through a total body computed tomography (CT) scan and a colonoscopy. This approach aims to rule out Gardner's syndrome, an autosomal dominant autoimmune disorder characterized by intestinal polyposis, multiple osteomas, skin fibroids, epidermoid cysts, as well as the presence of permanent and supernumerary dental elements [53,54,55] (4,6,10)

## RADIOGRAPHIC PRESENTATION

Osteomas frequently evade detection, given their propensity to remain asymptomatic, typically surfacing only when incidentally discovered during routine radiographic surveys. Radiographically, mandibular osteomas manifest as well-defined, elliptical, radiopaque masses firmly adherent to the cortical surface of the host bone via a broad base or pedicle.(33) They are discernible through imaging modalities such as panoramic radiographs and computed tomography (CT) scans.(5)

The characterization of the tumour, encompassing aspects like size, location, and its interrelation with adjacent structures, is pivotal for accurate diagnosis and the formulation of an effective treatment plan. Single-beam computed tomography (CT) stands out as the optimal imaging modality for evaluating the relationship between osteomas and adjacent structures, as well as for facilitating precise surgical planning (4)The comprehensive assessment for differential diagnosis should encompass conditions such as osteochondroma, fibrous dysplasia, chondroma, ossifying fibroma, condensing osteitis, tori, and exostoses, idiopathic osteosclerosis, osteoblastoma, cementoblastoma, and complex odontoma.In a CT scan, an osteoma presents as an exceedingly radiodense lesion, resembling the normal bone cortex, and mature osteomas may exhibit central marrow. Typically, round or oval, osteomas feature well-defined, smooth margins, lacking a perilesional halo (34). CT imaging excels in delineating the epicentre of a bone lesion (medullary, cortical, periosteal, or periosteal) and discerning its behaviour concerning adjacent structures, indicating either a benign or aggressive growth pattern. Notably, osteomas can lead to bone expansion, a distinctive characteristic aiding in the differential diagnosis from idiopathic osteosclerosis. Various CT findings are described in the literature based on osteoma subtypes: the ivory type is distinguished by very dense bone with small, well-defined lucent areas, while the mature type exhibits uneven bone density mixed with less dense areas, resembling a fibrous matrix. CT surpasses conventional radiography, providing detailed insights into the relationship between the osteoma and adjacent structures (35). Additionally, CT studies, with 2D and 3D reconstructions, offer significant support for surgical planning, particularly in cases involving complex anatomical locations(36).The utility of MRI in assessing craniofacial osteomas is constrained by the nature of these lesions. Given that osteomas are dense bone lesions, their evaluation is expedited and more effective with CT imaging (37). However, MRI can serve as a complementary tool to CT, particularly for evaluating adjacent soft tissues and complications related to osteomas, such as inflammatory changes in mucosa when an osteoma arises in the paranasal sinuses. (4)

## MANAGEMENT

The management of paediatric mandibular osteomas requires a nuanced approach, considering factors such as tumour dimensions, location, and impact on neighbouring structures. Surgical excision stands as the cornerstone of treatment, aiming to eliminate the tumour while preserving both functional capacity and aesthetic integrity. Meticulous planning is essential to minimize potential complications and ensure optimal postoperative outcomes. The recommended course of action is surgical intervention, with recurrence being infrequent, and importantly, there are no reported instances of malignant transformation (5,15,38).In cases involving mandibular osteomas where only cosmetic alterations are desired, therapy typically entails the straightforward excision of the lesion, with



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extraoral techniques reserved for larger osteomas requiring more extensive exposure (36,39). However, in certain scenarios, especially when managing smaller, asymptomatic lesions, a conservative strategy involving vigilant monitoring may be considered. The comprehensive care of patients in such situations necessitates the collaborative efforts of a multidisciplinary team, comprising oral and maxillofacial surgeons, paediatric surgeons, and radiologists. This collaborative approach ensures a holistic and informed strategy to address the complexities associated with paediatric mandibular osteomas.Over the past decade, computer-assisted surgery for jaw lesions has demonstrated superior outcomes compared to traditional techniques (36). This clinical advancement is attributed to the ability to simulate demolitive and reconstructive surgery preoperatively. The utilization of image-based planning for surgical resection, coupled with intraoperative navigation, holds significant promise in the realm of bone surgery and has particularly become pivotal in oncological cranio-maxillofacial surgery. Pre-operative resection plans can be accurately replicated intraoperatively using surgical navigation systems, allowing for precise identification of crucial anatomical structures (e.g., nerves, vessels, and muscles). This facilitates less demolitive surgical interventions and contributes to improved outcomes. (4) Typically reserved for patients with significant central osteomas characterized by large lesions in the mandibular or maxillary regions, where reconstructive procedures may be deemed necessary. Current medical literature underscores the efficacy of computer-assisted design and manufacturing techniques in jaw reconstruction as the optimal approach for achieving improved aesthetic and functional outcomes (40,41).

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