Glenoid Fossa Osteoma Resulting in a Progressive Malocclusion: A Case Report

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An osteoma is a benign tumor essentially restricted to the craniofacial skeleton. Osteomas occur most frequently in the frontal, ethmoid, and maxillary sinuses, and rarely affect the glenoid fossa. Osteomas are usually pain-free and remain silent, ie, symptom-free, for many years but may lead to occlusal changes. The report describes the case of an adult man with an osteoma who presented with a chief complaint of malocclusion and who was misdiagnosed. The case points to the diagnostic reasoning necessary to arrive at a correct diagnosis, especially when signs and symptoms, as well as pathology, are rare, eg, not familiar to the clinician.
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According to the classification of the American Academy of Orofacial Pain, the clinical conditions involving the temporomandibular joints (TMJs) include disc derangement disorders, degenerative and inflammatory disorders, congenital or developmental disorders, condyle dislocation, ankylosis, and fracture.[1] The less frequent clinical conditions may present with the same signs and symptoms caused by the more frequently found conditions of disc disorders and/or degenerative and inflammatory disorders. This increases the risk of an incorrect diagnosis if the clinician is not attentive in interpreting the history and the clinical examination findings. This is also the case for the rarely occurring tumors of the TMJ. These originate from the articulating bones (glenoid fossa and mandibular condyle), the disc, the synovial membrane, and the capsule, the majority arising from the condyle.[2]

An osteoma is a benign osteogenic lesion resulting from the continuous formation of both cortical and cancellous bone.[3,4] This tumor is essentially restricted to the craniofacial skeleton and is rarely diagnosed in other bones.[5] Osteomas occur most frequently in the frontal, ethmoid, and maxillary sinuses,[6] and rarely affect the temporal bone.[7,8] Osteomas are usually pain-free and may remain silent, ie, symptom-free, for many years. Treatment is surgical and, once removed, recurrence does not occur.[9]

Fig 1a Frontal view: The mandibular midline is deviated by 4 mm to the left in relation to the upper midline.

Fig 1b Crossbite on the left side.

Fig 2 Dental cast. The dental arches close without a crossbite.

This article reports the case of a patient with an osteoma of the glenoid fossa of the temporal bone that caused a malocclusion. The case is interesting because it is, to the authors' knowledge, the second case of osteoma of the glenoid fossa reported in the literature[10] and because it points to the risk of misdiagnosis if the history and the findings of the clinical examination are not carefully interpreted.

Case Report

History

A 43-year-old man presented with the chief complaint of malocclusion. He had noticed a change in his occlusion, which resulted in deviation of the mandible to the left. The patient had been previously assessed by two orthodontists who proposed to correct the malocclusion orthodontically and by a maxillofacial surgeon who suggested orthognathic surgery. The patient arrived at the Research Centre for the Study of Craniomandibular Disorders and Orofacial Pain for a further opinion.

The patient was in good health and without a history of previous facial trauma or contributory medical factors. He had never had any facial pain or jaw movement limitation.

Inspection, Clinical Examination, and Imaging

The intraoral examination revealed that the mandibular central incisors were deviated by 4 mm to the left with respect to the maxillary central incisors (Fig 1a). A cross-bite was present on the left. The upper and lower teeth on the left side had wear facets that could not be explained by the present occlusion (Fig 1b). Any effort to manipulate the mandible to correct the malocclusion was in vain.

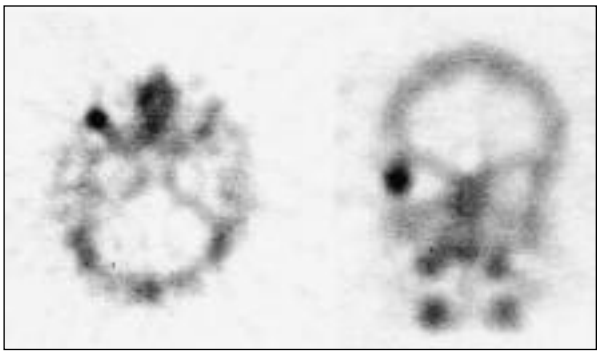
Palpation of the masticatory muscle and the TMJs was negative. There was no clicking or crepitation. Active mouth opening was 48 mm, without pain or any deviation. Lateral excursions were 9 mm to the right and 6 mm to the left, without pain. Protrusion reached 6 mm, with neither pain nor deviation.

Upper and lower dental impressions were taken to fabricate dental casts to check if it could be possible to make the dental arches fit together in maximum intercuspation without the left cross-bite, which was the case (Fig 2). This indicated that the malocclusion most likely had to be secondary to a joint pathology. Thus, computed tomography (CT) images of the TMJs were taken, and revealed a mass filling the right glenoid fossa, with the right condyle pushed forward out of the fossa (Figs 3a and 3b). Magnetic resonance imaging (MRI) showed that this mass was characterized by isointensity on T_1-weighted images and by high intensity on T_2-weighted images. To find out if the process was still active, bone scintigraphy was performed using a ^{99m}Tc-labeled complex. A distinct localized area of increased uptake was seen in the right TMJ (Fig 4). This uptake was consistent with the position of the lesion on CT and indicated bone growth.

Fig 3a Computed tomogram of the left TMJ, which appears normal.

Fig 3b Computed tomogram of the right TMJ. A bone mass is visible dorsally to the condyle that is located under the eminence.

Fig 4 The SPECT (single photon emission computed tomography) of the skull (transverse and coronal view) shows an abnormally increased isotope uptake in the area of the right TMJs.



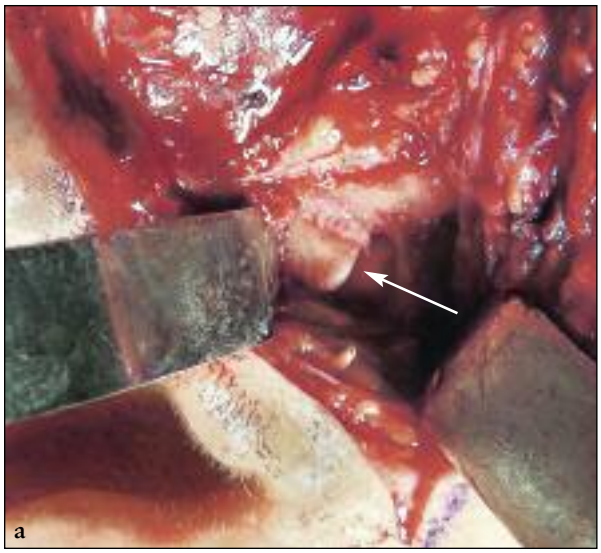


Fig 5a Intraoperative view of the glenoid fossa osteoma (indicated by the arrow).

Fig 5b After osteoma removal, the condyle reenters the fossa (the arrow shows the empty fossa).

Therapy

The patient was referred to the Department of Maxillofacial Surgery at Spedali Civili Hospital (Brescia, Italy) for surgery. Under general anesthesia, a preauricular incision was made through the skin and the temporal fascia to expose the deep temporal fascia. An inverted "L" incision was then made in the deep temporal fascia, and the temporal part of the arch was reached, minimizing the risk of injury to the frontal branch of the facial nerve. The glenoid fossa was therefore exposed and revealed a bone formation occupying the entire articular space and forcing the head of the condyle outside of the glenoid fossa (Fig 5a). The bone formation was removed, allowing the condyle to move back into the fossa (Fig 5b) and the mandible to close into the previous intercuspal position as visible from the

Fig 6 Frontal view after surgery. The dental arches close without crossbite and the midlines are almost perfectly aligned.

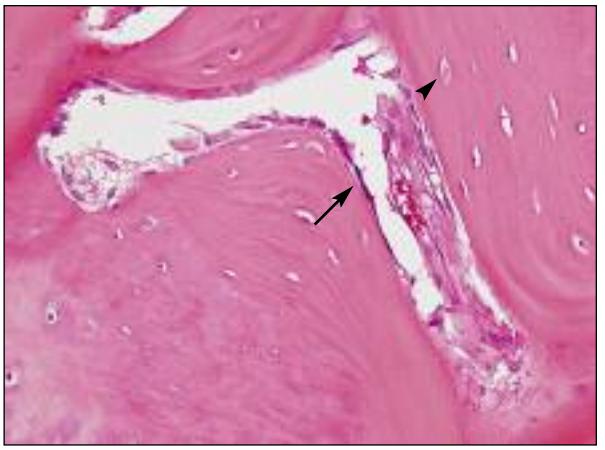


Fig 7 Histologic section of the osteoma (magnification ×200). Areas of compact bone proliferation are evident: osteoblastic line *(arrow)* and osteoclastic gaps *(arrowhead)*. Differentiation, necrosis, or atypical cells are absent. The picture is consistent with benign bone neoplasia.

picture taken 7-days postoperatively (Fig 6). There were no postoperative complications and the patient was discharged on the second postoperative day.

Macroscopically, the specimen consisted of compact dense bone measuring 1-cm wide and 1-cm long. The histological section revealed the presence of compact bone leading to the diagnosis of osteoma (Fig 7). At the 1-year postoperative follow-up, there was a stable occlusion with no sign of osteoma recurrence.

Discussion

Clinical conditions affecting the TMJ are characterized by common signs and symptoms that have to be investigated by means of proper examination.[11] Temporomandibular disorders (TMD) encompass a series of different conditions that may present with the same signs and symptoms, therefore the final diagnosis must derive from careful comparison of the history data and the clinical examination findings.[1] This concept is even more important when facing some unusual conditions that are not included in the most common diagnostic subgroups.

This case report points to the diagnostic pitfalls when a patient presents with a rare pathology, eg, a pathology that is not familiar to the clinician, for instance in the presence of tumors or other developmental disorders. The prevalence of such conditions is low in comparison to the prevalence of TMJ disc disorders and/or degenerative and inflammatory disorders. In these cases, the risk of overlooking the correct diagnosis is elevated. For this reason, a careful history and clinical examination are fundamental in order to be able to rule in and/or rule out all possible causes.

The main complaint of this adult patient was the change in occlusion. History revealed that the occlusal change was not secondary to a trauma and was not accompanied by facial pain or masticatory impairment. The clinical examination was negative for signs of TMD leading to the exclusion of TMD as a possible cause. In patients with occlusal changes that occur after growth, it is important to determine whether these are due to alterations of the dental arches, eg, to tooth migration, or to a mandibular position change caused by an intra- or extracapsular disorder and/or pathology (Fig 8).

The evaluation of dental casts is of great help for this differential diagnosis, as they allow the clinician to find out if the occlusal arches and the wear facets still fit together. If this is the case, the occlusal alteration is due to a mandibular position change that can be of intra- or extracapsular origin. Intra-articular causes encompass intra-articular swelling due to TMJ inflammation,[12] condylar resorption due to degenerative and/or inflammatory changes,[12] idiopathic condylar resorption,[13] condylar exostosis,[14] condylar hyperplasia, posterior disc displacement,[15] and benign or malignant neoplasms such as chondromablastoma, osteochondroma, osteoma, osteoid osteoma, osteoblastoma, non-ossifyng fibroma, central giant cell granuloma,

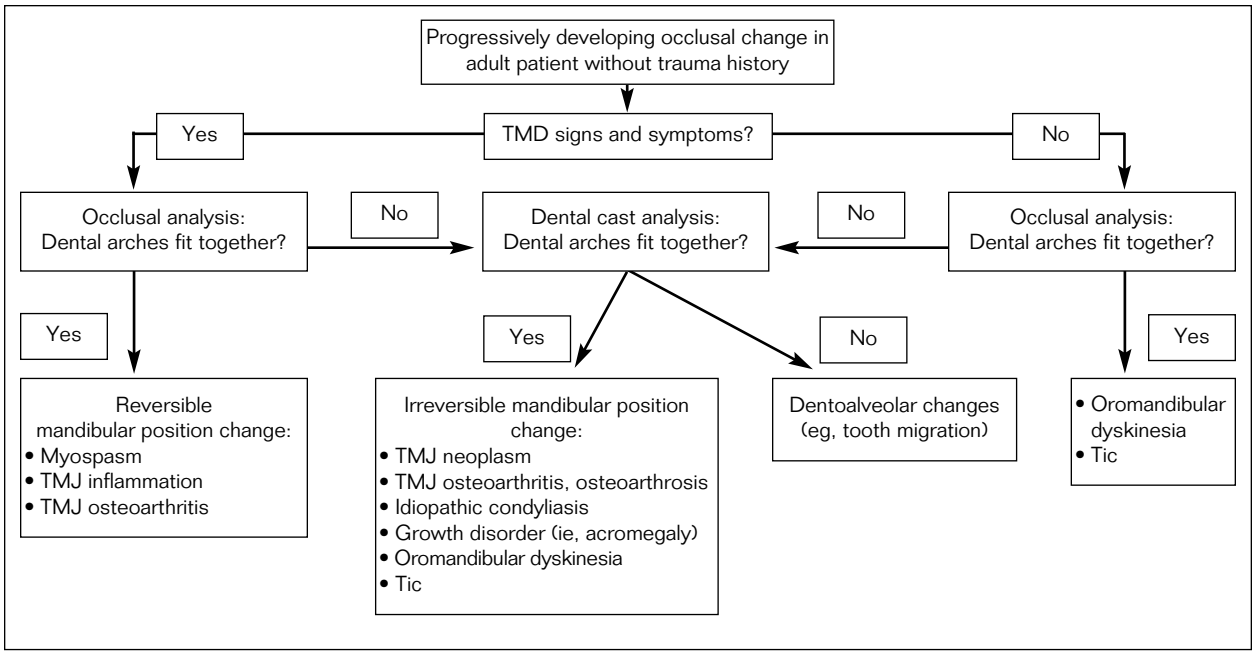


Fig 8 Diagnostic flowchart.

Table 1 Causes of Secondary Malocclusion

Acute		Chronic	
Conditions	Additional features	Conditions	Additional features
Trauma	Pain	TMJ inflammation	Pain, swelling
Posterior disc displacement	Posterior open bite with or without pain	Degenerative changes of articular surfaces	Pain, TMJ sounds like crepitus
Myospasm	Pain	TMJ tumors	

osteosarcoma, or chondrosarcoma.[2,16,17] Among the extra-articular causes, the most common is likely a sustained contraction of the inferior lateral pterygoid muscle. [18]

Occlusal alterations may appear suddenly or progressively (Table 1). Among causes of acutely induced malocclusion, trauma, posterior disc displacement, and myospasm have to be considered. The patient in this case noted neither previous trauma nor pain in the orofacial region, so a traumatic origin of the malocclusion was excluded. In rare cases, disc displacement can occur in the posterior direction; in such cases posterior open bite may result,[15] and this was not apparent in the present case. Myospasm is a tonic muscle contraction. When a muscle is contracted, jaw positional changes result, depending on the muscle in spasm. If the involved muscle is the inferior lateral pterygoid muscle, a contralateral cross-bite results.[18] Generally, myospasm presents with pain.

Possible causes of slower, progressively induced malocclusion also then need to be taken into account. First of all, TMJ inflammation can occur with intra-articular swelling that can cause an ipsilateral malocclusion.[12] Inflammation presents with pain. Degenerative changes in the osseous surfaces can occur as a consequence of inflammatory arthritis involving the articular surfaces of the TMJ. In such a case, an anterior open bite may result, and sounds such as crepitus may be recorded on TMJ palpation during opening and closing of the jaw.[12] An anterior open bite may also be due to idiopathic condylar resorption.[13] Exostosis of the condyle may slowly produce a malocclusion without any other symptoms.[14] Finally, TMJ tumors may cause dislocation of the condyle and lead to malocclusion.

In the present case, the casts met in the previous maximum intercuspation position. The lack of history of trauma, the inability to correct the malocclusion by jaw manipulation, and the lack of facial pain led to the diagnostic hypothesis of an intra-articular pathology, in particular of a neoplastic process within the TMJ that caused a progressive mandibular asymmetry and therefore an occlusal change without any other accompanying clinical sign, in particular pain.[2,16] TMJ imaging confirmed the presence of a neoplastic mass and the histological evaluation revealed the presence of an osteoma.

When a patient complains about malocclusion without pain or functional impairment, it may be easy to focus just on dental treatment, that is, on correcting the malocclusion. But it is a fundamental question to consider why an adult has suddenly noted his malocclusion. What has changed? Keeping an open mind and considering such questions are crucial for the patient's well-being. Even if an osteoma is a benign tumor, an earlier diagnosis is crucial for a correct therapy. A comprehensive history and a careful interpretation of the findings of the clinical examination are mandatory for a correct diagnosis in cases of occlusal alterations in adult patients.

References

1. Okeson JP. Differential diagnosis and management considerations of temporomandibular disorders. In: Okeson JP. Orofacial Pain: Guidelines for Assessment, Classification and Management. Chicago: Quintessence, 1996: 113–184.
2. Stern D. Benign and malignant tumors. In: Laskin DM, Greene CS, Hylander WL. Temporomandibular Disorders: An Evidence-Based Approach to Diagnosis and Treatment. Chicago: Quintessence, 2006:319–333.
3. Dorfman HD, Czerniak B. Bone tumors. St. Louis: Mosby, 1998:85–114.
4. Lucas RB. Pathology of Tumours of the Oral Tissues, ed 4. Edinburgh: Churchill Livingstone, 1984:191.
5. Siar CH, Jalil AA, Ram S, Ng KH. Osteoma of the condyle as the cause of limited-mouth opening: A case report. J Oral Sci 2004;46:51–53.
6. Sayan NB, Üçok C, Karasu HA, Günhan Ö. Peripheral osteoma of the oral and maxillofacial region: A study of 35 new cases. J Oral Maxillofac Surg 2002;60:1299–1301.
7. Gungor A, Cincik H, Poyrazoglu E, Saglam O, Candan H. Mastoid osteomas: Report of two cases. Otol Neurotol 2004;25:95–97.
8. Ben-Yaakov A, Wohlgelernter J, Gross M. Osteoma of the lateral semicircular canal. Acta Otolaryngol 2006;126: 1005–1007.
9. Clayman L. Surgical management of benign and malignant neoplasms. In: Laskin DM, Greene CS, Hylander WL (eds). Temporomandibular Disorders: An Evidence-Based Approach to Diagnosis and Treatment. Chicago: Quintessence, 2006:509–532.
10. Attanasio R, Bavitz JB, Cohen D, Gelline S. Prosthodontic and surgical management of a patient with an osteoma of the temporomandibular joint. Compend Contin Educ Dent 1998;19:258–262.
11. Dworkin SF, LeResche L. Research diagnostic criteria for temporomandibular disorders: Review, criteria, examinations and specifications, critique. J Craniomandib Disord 1992;6:301–355.
12. Okeson JP. Temporomandibular joint pains. In: Okeson JP. Bell's Orofacial Pains: The Clinical Management of Orofacial Pain, ed 6. Chicago: Quintessence, 2005: 329–379.
13. Pogret MA, Chigurupati R. Management of idiopathic condylar resorption. In: Laskin DM, Greene CS, Hylander WL. Temporomandibular Disorders: An Evidence-Based Approach to Diagnosis and Treatment. Chicago: Quintessence, 2006:533–540.
14. Cimino R, Steenks MH, Michelotti A, Farella M, Nocini P. Mandibular condyle osteochondroma. Review of the literature and report of a misdiagnosed case. J Orofac Pain 2003;17:254–261.
15. Westesson PL, Larheim TA, Tanaka H. Posterior disc displacement in the temporomandibular joint. J Oral Maxillofac Surg 1998;56:1266–1273.
16. Hatcher DC, McEvoy SP, Mah RT. Maxillomandibular relationships: Mandibular asymmetry, occlusion, and imaging. In: McNeill C (ed). Science and Practice of Occlusion. Chicago: Quintessence, 1997:273–293.
17. Kroon HM, Schuurmans J. Osteoblastoma: Clinical and radiologic findings in 98 new cases. Radiology 1990;175: 783–790.
18. Okeson JP. Pains of muscle origin. In: Okeson JP. Bell's Orofacial Pains: The Clinical Management of Orofacial Pain, ed 6. Chicago: Quintessence, 2005:287–328.