


## CASE REPORT

# Surgical resection of osteochondroma in the mandibular condyle associated with directional orthodontic treatment: review of literature and a case report

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## Abstract

Characterised by abnormal bone and/or cartilage formation on the surface of a normal bone, osteochondroma is a benign tumour manifested by poorly oriented bony protuberances, covered by hyaline cartilage and of normal histological appearance. Osteochondroma rarely occurs in the buccal and maxillofacial region, and the signs and symptoms vary according to the size and location of the tumour. In cases that involve the condyle of the mandible, the commonly recommended management is the radical resection of the tumour including the condyle, aiming to re-establish aesthetics and function. When total condilectomy is chosen, there is a discussion in the literature about whether to perform mandibular condyle reconstruction or not. In the vast majority of cases reported, immediate reconstruction or association with other surgical procedures, such as orthognathic surgery, was chosen. This article aims to report a successful management of a case of osteochondroma in the mandibular condyle, resected surgically without immediate reconstruction, associated with directional orthodontic treatment and correlated it with the literature.

## Introduction

According to the World Health Organization (WHO), osteochondromas are bony projections surrounded by a cartilage layer, which rise on the outer surface of the bone. Its composition is predominantly bony; however, the cartilaginous portion is responsible for its growth<sup>1</sup>. There is still controversy as to its etiopathogenesis. Although the WHO defines it as a true neoplasm, some authors indicate it as developmental or reparative in nature<sup>2-6</sup>. Factors such as trauma and inflammation have been described in the literature as contributing to its development<sup>2</sup>. Osteochondroma is one of the most prevalent bone neoplasms, accounting for 35–50% of benign bone tumours and 8–15% of all bone tumours (benign and malignant)<sup>7</sup>. The most affected anatomical sites are the metaphyseal regions of the long bones of the

lower limbs (50% of cases), where the distal femur appears as the most prevalent<sup>8</sup>.

Although they are frequent in the axial skeleton, osteochondromas rarely affect the buccal and maxillofacial region, found approximately in 1% of the cases<sup>9</sup>. When present, the coronoid process and the mandibular condyle are the most affected areas<sup>10</sup>. Other sites such as the zygomatic bone, maxillary sinus and the base of the skull have also been reported in the literature<sup>11</sup>. The signs and symptoms vary according to the size and location of the tumour. Condylar osteochondromas present slow growth and cause an progressively increase in volume on the affected side, as well as pain, facial asymmetry, deformity of the chin, mandibular prognathism, limitation of the buccal opening, malocclusion with crossbite on the contralateral side of the condylar tumour, dental midline deviation, cracking

in the joint with mouth opening and enlarged lobular mandibular condyle head<sup>2-3,12-14</sup>. Although diagnosis can be suggested through clinical imaging and complementary findings (such as the use of bone scintigraphy<sup>15</sup>), diagnostic confirmation can only be executed by histopathological examination<sup>8,16</sup>.

The therapeutic choice for osteochondromas of the mandibular condyle usually involves surgical treatments, which may be immediate or delayed<sup>17</sup>. The recommended treatment should re-establish aesthetics and function, achieving acceptable facial harmony with buccal opening and occlusion<sup>2,3,5</sup> and depending on the aesthetic flaw, the treatment may vary between partial or total condilectomy, with or without adjunctive orthognathic surgery. Condylar reconstruction is also an option after the tumour resection and may be performed by ascending reposition of the sagittal ramus or vertically divided, autogenous bone graft or using joint prosthesis<sup>16,18</sup>. The present study aims to report a successful management of a case of osteochondroma in the mandibular condyle treated surgically without reconstruction, correlating it with the literature.

## CASE REPORT

A 24-year-old female patient with leucoderma visited the Oral and Maxillofacial Surgery services of the State University of Maringá claiming to have 'an earache, a headache and facial asymmetry'. During the anamnesis, the patient reported having noticed mandibular asymmetry for roughly 3 years, and having been referred by her orthodontist for an evaluation for maxillary orthognathic surgery. Extraoral physical examination revealed an increase in volume in the region of the left temporomandibular joint, mandibular laterognathism to the right, facial asymmetry, mild mandibular prognathism (Fig. 1) and mouth opening limited to 30 mm. During the intraoral examination, the patient had unilateral posterior crossbite (right side) and dental midline deviation (Fig. 2). A cone beam computed tomography showed a well-defined hyperdense lesion involving the left mandibular condyle, with irregular and globular altered morphology, and approximately 30 mm in length (Fig. 3).

From the clinical and tomographic findings, diagnostic hypotheses of mandibular condyle tumour (benign or malignant) and unilateral condylar hyperplasia were raised. The patient underwent excisional biopsy under general anaesthesia and nasotracheal intubation. Endaural access was recommended to promote direct access to the lesion, causing a less

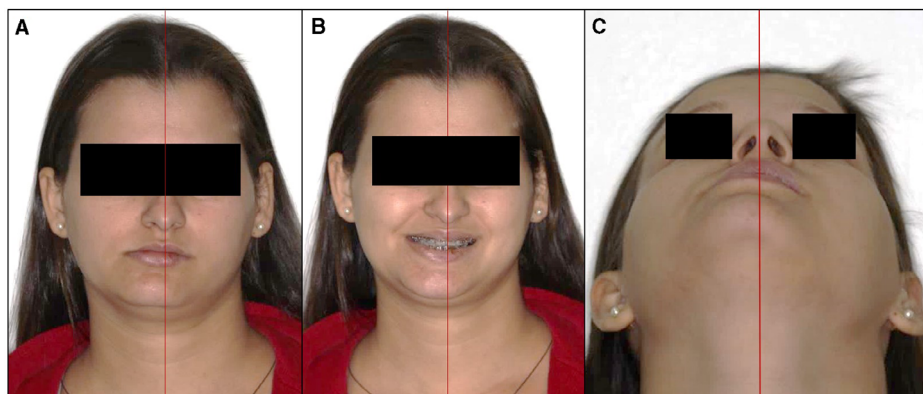
visible scar and offering less risk to noble structures. After incision and tumour exposure, osteotomies were performed with a piezoelectric saw, removing the entire lesion in small fragments (Fig. 4).

The surgical wound was sutured and the biopsy material was sent for histopathological examination. The histological sections revealed a bone lesion composed of mature bone without atypia with a chondral coating area also without atypia. The correlation with the clinical and radiological findings confirmed the hypothesis of osteochondroma of the mandibular condyle. A computerised tomography was performed in the immediate post-operative period (Fig. 5), evidencing the total excision of the lesion. Thereafter, the patient started the orthodontic treatment with the use of elastic therapy, in order to guide the occlusion correctly. This therapy lasted for 3 months, and after 8 weeks of stability without the use of elastics, orthodontic treatment was completed. The patient is undergoing a 1-year follow-up, presenting satisfactory function and aesthetics, with improvement in midline correction (Fig. 6), stable occlusion (Fig. 7), and re-establishment of an adequate oral opening of 48 mm. The panoramic and posteroanterior X-rays of the face (Fig. 8) emphasise the success of the proposed treatment and the patient does not show signs of relapse.

## Discussion

The occurrence of condylar osteochondromas can be explained by the presence of aberrant foci of epiphyseal cartilage on the bone surface<sup>19</sup>. It is believed that stress in the region of tendinous insertion, where there is accumulation of cells with cartilaginous potential, leads to the formation of these tumours<sup>20,21</sup>. Although osteochondromas account for most of the benign bone tumours, occurring mainly in the metaphyseal region of the long bones, they are rarely reported in the buccal and maxillofacial regions. Most of the osteochondromas of this region affect the coronoid process of the mandible<sup>22</sup>; however, its incidence in the mandibular condyle is extremely uncommon<sup>7</sup>.

They may appear as a solitary lesion (75%) or as multiple lesions (25%) and occur more frequently in women in the second decade of life<sup>8,16</sup>. The case presented corroborates these studies, since it was a single lesion in a 24-year-old female patient. The diagnostic hypotheses in the case reported were osteochondroma of the mandibular condyle and unilateral condylar hyperplasia; however, unilateral condylar hyperplasia manifests as an increased



**Figure 1** Photographs showing facial asymmetry with shift of the chin to the right side; (A) front with lip sealing; (B) front smiling; (C) axial view. [Colour figure can be viewed at [wileyonlinelibrary.com](http://wileyonlinelibrary.com)]



**Figure 2** Intraoral photograph showing malocclusion with unilateral posterior crossbite (right side) and midline deviation. [Colour figure can be viewed at [wileyonlinelibrary.com](http://wileyonlinelibrary.com)]

condylar process with unchanged morphology, whereas osteochondroma exhibits abnormal morphology with irregular and globular appearance<sup>16</sup>; thus, the tomographic findings were inclined to a diagnosis of osteochondroma.

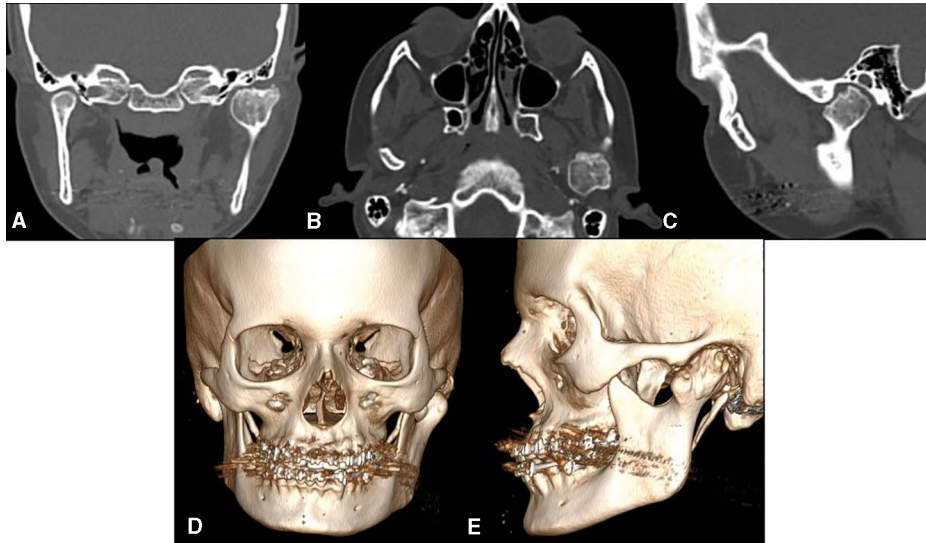
Although clinical and imagery findings may suggest the existence of an osteochondroma, it is the histopathological examination that confirms the diagnosis, revealing aberrant cartilaginous proliferation and calcification, as well as a cartilaginous layer that is characteristic of the tumour<sup>8,16</sup>. The patient in the present case had most of the signs and symptoms described in the literature; nevertheless, facial asymmetry was the main reason for seeking care. Mild degrees of facial asymmetry are common in society; however, at higher levels, they may result in aesthetic and functional discomfort, making surgical interventions as treatment options<sup>23-24</sup>. In the case

reported, the patient sought care with interest in performing an orthognathic surgery, unaware of the presence of the pathology. Due to the clinical and historical aspects of the disease, the suspicion of condylar hyperplasia or osteosarcoma was raised, but after the analysis of the tomographic images, the main diagnostic hypothesis was osteochondroma.

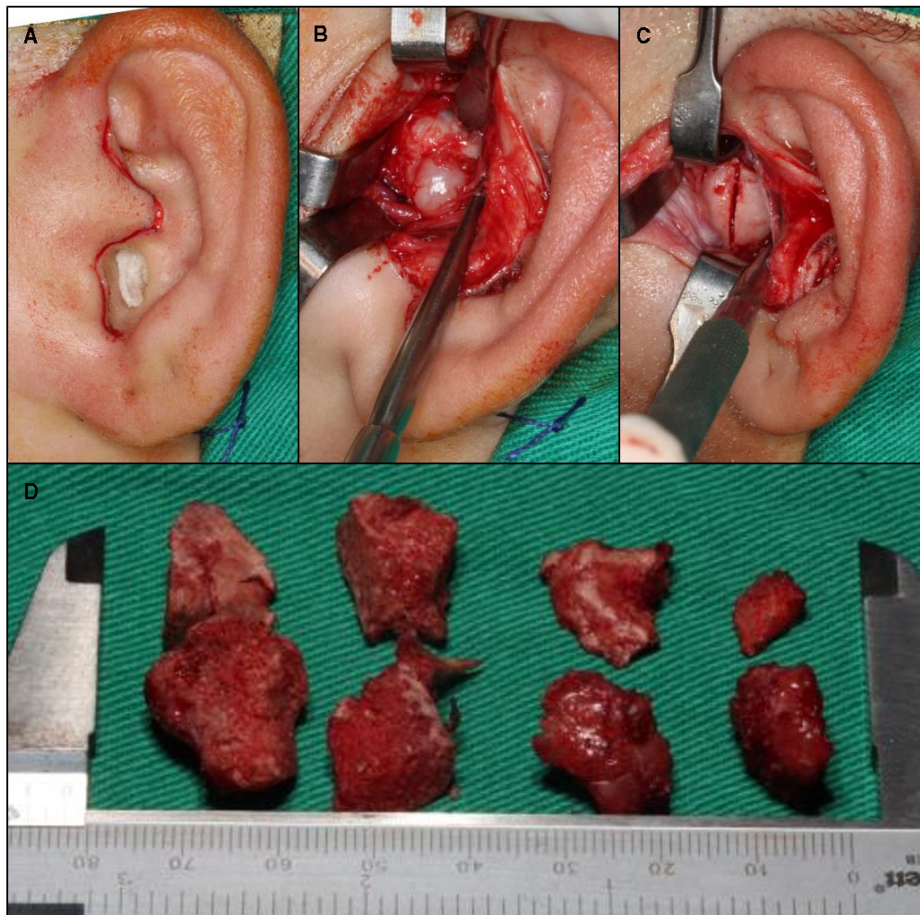
The treatment of this pathology is surgical, and its removal is fundamental for the resolution of clinical changes promoted by the tumour, but may vary from partial to total condilectomy, and in some cases requires adjuvant orthognathic surgery<sup>16,18,25-26</sup>. Conservative procedures are employed in restricted cases and present a higher rate of recurrence of the lesion, and more rarely to a malignant transformation; and may require correction of additional occlusal and facial asymmetry<sup>18,27-29</sup>. When total condilectomy is opted, there is a discussion in the literature about whether or not to perform mandibular condyle reconstruction. Authors mention that after resection, there may be an open bite on the contralateral side, facial asymmetry, changes in mandibular movement and opening deviation, thus, condylar reconstruction would be fundamental to re-establish the height of the mandibular ramus and, consequently, maintenance of the occlusion, mandibular movements and mandibular symmetry<sup>16,18,29</sup>.

In this regard, there are different forms of reconstruction, such as the use of autogenous grafts (costochondral graft, fibula, clavicle, iliac crest), vertical osteotomies of the mandibular ramus and alloplastic grafts (prosthetic materials)<sup>16,18,29-31</sup>. However, despite their advantages, these techniques also present failure rates. Non-vascular autogenous grafts require long-term immobility, donor site morbidity,

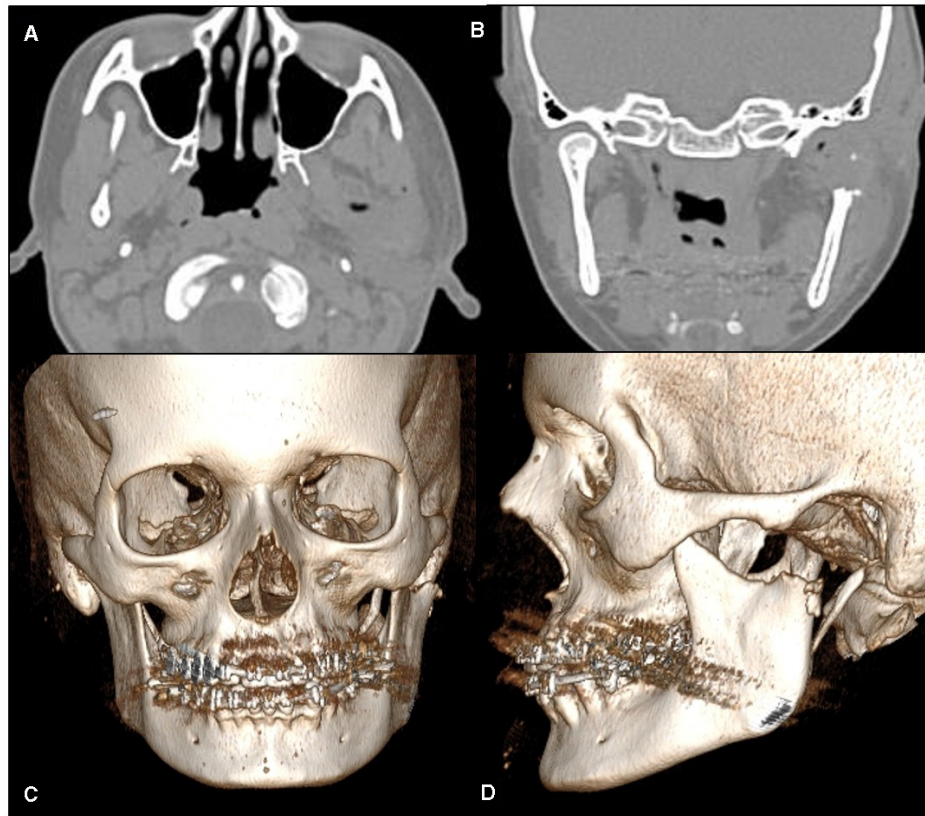




**Figure 3** CT scans showing a well-delimited hyperdense lesion involving the left mandible, with its altered morphology. (A) Coronal cut; (B) axial cut; (C) sagittal cut (D) 3D reconstruction frontal view; (E) 3D reconstruction left lateral view. [Colour figure can be viewed at [wileyonlinelibrary.com](http://wileyonlinelibrary.com)]



**Figure 4** Access and removal of the tumour. (A) Incision for endaural access; (B) tumour exposure; (C) osteotomy with piezoelectric motor; (D) fragments of the lesion after excision. [Colour figure can be viewed at [wileyonlinelibrary.com](http://wileyonlinelibrary.com)]



**Figure 5** Cone-beam computed tomography of immediate post-operative showing the absence of the left condyle. (A) Axial cutting; (B) coronal reconstruction; (C) front 3D reconstruction; (D) left lateral 3D reconstruction. [Colour figure can be viewed at [wileyonlinelibrary.com](http://wileyonlinelibrary.com)]



**Figure 6** Post-operative photographs of 1 year. (A) Front view with lip sealing; (B) front view smiling; (C) axial view. [Colour figure can be viewed at [wileyonlinelibrary.com](http://wileyonlinelibrary.com)]

high rates of reabsorption and the possibility of autograft ankylosis in the TMJ<sup>18,32</sup>. Microvascular bone grafts require high-cost planning, and as well as vertical osteotomies of the mandibular ramus, they can cause nerve and vascular lesions, promote

inadequate mandibular condyle height and consequently secondary orthognathic and masticatory disorders<sup>18,33</sup>. Furthermore, reconstructions with alloplastic material require cutting-edge technology and high-cost material available<sup>18,32</sup>.





**Figure 7** Post-operative intrabuccal photograph of 1 year; patient with crossbite correction and improvement in midline deviation. [Colour figure can be viewed at [wileyonlinelibrary.com](http://wileyonlinelibrary.com)]

In addition, the literature has shown few studies of osteochondromas treated by surgical resection without simultaneous reconstruction<sup>16,23,32,34</sup>, associated or not with directional orthodontic treatment<sup>34,35-38</sup>. In this situation, cases of orthodontics with elastic use for 6–12 weeks were cited which, due to their ability to resist muscular forces and the tendency of skeletal relapse<sup>35,39</sup>, are able to guide patient occlusion, gradually adjust the affected mandibular condyle within the glenoid fossa, and facilitate TMJ remodelling<sup>35</sup>. Thus, we performed a bibliographic review, summarised in Table 1, the main cases reported in the literature with their forms of treatment, positive results and complications obtained.

The surgical approach of choice in this case was resection of the tumour by condilectomy without immediate reconstruction and without orthognathic surgery. The decision was made considering the fact that it is a tumour that has not yet been definitively diagnosed, and therefore, partial removal could lead to relapses, possibility of malignant transformation, and maintenance or worsening of the clinical picture<sup>18,27-28</sup>. Besides that, the facial and occlusal

changes were not as discrepant, leading us to wait for the result of this procedure to re-evaluate the need for other complementary interventions. The recommended access was an endaural access taking into account the anatomical location of the tumour. Usually, the surgical window used to approach this type of tumour is a region with important innervation and abundant vascularisation. Due to the proximity of noble structures to the intervention site, a decision was made based on an approach for this access, reducing the surgical risks related to the nearby structures and resulting in a scar with minimal aesthetic impact for the patient<sup>25</sup>.

After the surgery, physiotherapy and directional orthodontic treatment with the use of elastics were started to direct the occlusion of the patient<sup>35</sup>. This procedure was performed for roughly 3 months, and after the end of this period, we observed an adequate occlusion with acceptable facial aesthetics and preserved mandibular movements. Based on these findings, we consider new surgical procedures unnecessary at the moment. In addition, factors such as autogenous graft morbidity and the high cost of a condylar prosthesis<sup>18,32</sup> also pointed to other procedures. The patient has been in the follow-up period for 1 year and shows no signs of relapse, which evidences the success of the proposed treatment. This may point out that, on some occasions, we can opt for less aggressive approaches with lower biological and financial costs, obtaining satisfactory results.

## CONCLUSION

Osteochondromas in the mandibular condyle are extremely rare, and when they occur, the main goal of treatment is to return function and improve facial aesthetics, and it is important to choose a technique that brings the least harm to the patient. In the case reported, the proposed treatment without immediate reconstruction, associated with directional orthodontic



**Figure 8** Follow-up 1-year radiographs. (A) Panoramic radiography; (B) posteroanterior face radiograph.

**Table 1** Summary of treatments proposed in the literature through case reports, including surgical access, its positive results and complications

Authors	Access	Treatment	Positive results	Complications
Chen et al. (2014)	Preauricular	Resection of the tumour	<ul style="list-style-type: none"> <li>• Facial aesthetics</li> <li>• No sign of recurrence after 1 year</li> <li>• Functional stability</li> </ul>	Not reported
Friedrich et al. (2012)	Preauricular	Resection of the tumour with the cutting line at the interface of the clumpy lesion to the base of the condyle	After 6 months, the malocclusion was recovered No sign of recurrence	<ul style="list-style-type: none"> <li>• Malocclusion persisted for several months</li> <li>• Mouth opening is restricted 35 mm</li> </ul>
Iwata et al. (2016)	Temporal flap	Tumour and articular disc excision + orthodontic treatment	Stable occlusions after orthodontic treatment.	Without orthodontic treatment occurs an malocclusion with open bite
Kwon et al. (2017)	Preauricular	Conservative condylectomy	<ul style="list-style-type: none"> <li>• Preservation of the vertical height</li> <li>• Stable occlusion</li> <li>• Additional reconstruction is not required</li> </ul>	Recurrence after 3 years
Li et al. (2014)	Retromandibular	Condylectomy + vertical ramus osteotomy + sagittal split ramus osteotomy + ipsilateral inferior border osteotomy + genioplasty + orthodontic therapy	<ul style="list-style-type: none"> <li>• Good remodelling of the mandibular angle</li> <li>• Remission of pain symptoms</li> <li>• Stable occlusions</li> <li>• Facial aesthetics</li> <li>• No obvious bone resorption</li> </ul>	Not reported
Li et al. (2016)	Not reported <ul style="list-style-type: none"> <li>• Virtual surgical planning and guiding templates</li> </ul>	Resection of the tumour and the mandibular condyle + vertical ramus osteotomy ipsilateral and in the contralateral side + osteotomy in inferior border + genioplasty	Facial aesthetics Stable occlusions	Not reported
Lim et al. (2014)	Circumvestibular + vestibular in mandible + endaural	Resection of the tumour with preservation of the medial pole of the condyle + orthognathic surgery	No sign of recurrence	Mouth opening limitation at 29 mm
Lim et al. (2014)	Circumvestibular, vestibular in mandible + preauricular	Condylectomy + ipsilateral coronoidectomy	No sign of recurrence after 3 years	Deviation in opening mouth
Luo et al. (2016)	Preauricular and submandibular	Condylectomy + vertical ramus osteotomy + orthodontic treatment with elastics	<ul style="list-style-type: none"> <li>• No sign of recurrence</li> <li>• Proper mouth opening</li> <li>• Remission of pain symptoms</li> <li>• Stable occlusions</li> <li>• Facial aesthetics</li> <li>• No obvious bone resorption</li> </ul>	Malocclusion could be resolved partially

**Table 1** (Continued)

Authors	Access	Treatment	Positive results	Complications
Ma <i>et al.</i> (2018)	Modified preauricular	Resection of the tumour and preservation of the condylar head + orthodontic traction	<ul style="list-style-type: none"> <li>• Facial aesthetics</li> <li>• No sign of recurrence</li> <li>• Stable occlusions</li> <li>• Proper mouth opening</li> <li>• Remission of pain symptoms</li> </ul>	Mandibular deviation was not completely corrected in several patients
Morey-Mas <i>et al.</i> (2011)	Preauricular and Submandibular	Condylectomy + immediate reconstruction with stock total TMJ prosthesis	<ul style="list-style-type: none"> <li>• Remission of pain symptoms</li> <li>• Proper mouth opening</li> <li>• No sign of recurrence</li> </ul>	Not reported
Park <i>et al.</i> (2017)	Preauricular with Zygomatic arch osteotomies	Conservative resection of the tumour and preservation of the condyle	Symptoms remission	Because the short follow-up period, they could evaluate recurrence for this case
Ramos-Murguialday <i>et al.</i> (2011)	Preauricular and submandibular	Condylectomy and discectomy + reconstruction with chondrocostal graft	<ul style="list-style-type: none"> <li>• No sign of recurrence</li> <li>• Stable occlusions.</li> <li>• Proper mouth opening</li> <li>• Remission of pain symptoms</li> </ul>	Not reported
Ramos-Murguialday <i>et al.</i> (2011)	Preauricular and submandibular	Resection of the tumour + reconstruction with stock total TMJ prosthesis	<ul style="list-style-type: none"> <li>• No sign of recurrence</li> <li>• Proper mouth opening</li> <li>• Remission of pain symptoms</li> </ul>	Not reported
Sun <i>et al.</i> (2013)	Intraoral	Tumour resection with endoscopy combined with a navigation system	<ul style="list-style-type: none"> <li>• Minimum risk of damaging vital structures</li> <li>• No facial nerve injury or salivary fistula</li> <li>• No sign of recurrence</li> </ul>	Not reported
Tantanapornku <i>et al.</i> (2017)	Extraoral – not specified	Condylectomy and extraoral vertical ramus osteotomy	<ul style="list-style-type: none"> <li>• Remission of pain symptoms</li> <li>• Posterior cross-bite</li> <li>• Proper mouth opening</li> <li>• Appropriate contralateral</li> </ul>	<ul style="list-style-type: none"> <li>• Slight deviation in opening mouth</li> <li>• Midline deviation</li> </ul>
Yang <i>et al.</i> (2015)	Submandibular	Resection of the tumour	<ul style="list-style-type: none"> <li>• Remission of pain symptoms</li> <li>• Stable occlusions</li> <li>• Facial aesthetics</li> </ul>	Restriction of mouth opening after 1 year



**Table 1** (Continued)

Authors	Access	Treatment	Positive results	Complications
Zhou <i>et al.</i> (2016)	Retromandibular	Resection of the tumour with the condyle + vertical ramus osteotomy of the posterior border	Facial aesthetics and stable occlusions during the first 10 months	<ul style="list-style-type: none"> <li>• Slight facial asymmetry after 11–12 months</li> <li>• Mandibular deviation to the operated side in mouth opening</li> <li>• Neocondylar resorption and reduction in height are apparent in the radiographs</li> </ul>

treatment was successful, being considered a satisfactory alternative, since the patient is still without signs of relapse and with satisfactory function and aesthetics. However, new studies on this treatment modality are necessary, considering that there are few studies published in the literature.

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## Conflict of Interest

Author 1 declares that she has no conflict of interest. Author 2 declares that she has no conflict of interest. Author 3 declares that he has no conflict of interest.

## Ethical approval

This article does not contain any studies with animals performed by any of the authors.

## Informed consent

Informed consent was obtained from all individual participants included in the study.

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