Case Report

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Management of temporomandibular joint osteochondroma with facial asymmetry: a case report

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Osteochondroma is rare in the facial region and osteochondroma of the mandible typically causes facial asymmetry and malocclusion. Resection of the affected condyle and orthognathic surgery has been frequently reported as the treatment of choice. The aim of this study was to manage a case of osteochondroma of the temporomandibular joint (TMJ) causing facial asymmetry. After extensive treatment plan discussions between the surgeon and the patient, the patient was reluctant to undergo the surgeries and chose to wait and see if there was cessation of the condylar hyperplasia instead of undergoing orthognathic surgery and condylectomy. Thus, this case was followed up by performing clinical examinations, radiographs, CT, and bone scintigraphy for approximately eight years. Subsequently, the facial asymmetry and the skeletal discrepancy became more severe. Furthermore, the bone scan revealed evidence of ongoing condylar hyperplasia. Based on these findings, the patient chose to undergo surgery. Low condylectomy with bimaxillary orthognathic surgery were selected for this case. The intraoperative sequence, however, differed from other studies in that the maxilla orthognathic surgery was performed prior to resecting the condyle and bilateral sagittal split osteotomy (BSSRO). The histopathologic results indicated that the tumor was consistent with osteochondroma. His facial asymmetry and malocclusion had been appropriately corrected. The patient did not experience restricted mouth opening, whereas postoperative complications, such as lip paresthesia and facial weakness were reported after three months. However, three months later, these complications had resolved. Moreover, the radiographic evaluation demonstrated no recurrence of the osteochondroma.

Keywords: condylectomy, mandibular condyle, orthognathic surgery

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Introduction

Osteochondroma (OC) or osteocatilagenous exostosis is an exophytic lesion arising from the bone cortex and is capped with cartilage [1]. OC is one of the most common benign tumors of the long bones, occurring in 35%–50% of all benign bone tumors and 8%–15% of all primary bone tumors [2]. While frequently occurring in the general skeleton, it is rarely associated with the facial bones because these bones develop via intramembranous formation [3]. The coronoid and condylar processes are the most common sites of the facial skeleton for OC to occur [4-6]. Facial asymmetry may result from various mandibular pathologies, including hemi-mandibular hyperplasia or elongation, mandibular condylar hyperplasia or elongation, and benign tumors or cysts of the mandible [7-8]. With its distinct border, radiographic investigations, such as radiography and computerized tomography (CT) have been commonly used to evaluate OCs [3, 9]. In addition to the problems of facial appearance, patients have trouble chewing due to malocclusion, speech disturbances, and temporomandibular joint dysfunction. The typical condylar OC clinical

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features include progressive facial asymmetry, prognathic deviation of the chin, cross-bite to the contra-lateral side, changes in condylar morphology, and malocclusion with an open-bite on the affected side [10-11]. Correcting these deformities can be achieved by surgically excising the pathology and/or orthognathic surgery depending on the etiology and severity of the asymmetry. This clinical report describes the management of a facial deformity due to OC of the left TMJ by a combination of a low condylectomy on the affected side and bimaxillary orthognathic surgery (Le Fort I and Bilateral Sagittal Split Ramus Osteotomy (BSSRO)). To manage osteochondroma of the TMJ that is causing facial asymmetry, several factors influence the type of treatment appropriate for each patient. Those factors include age (growing children or adults), location of the mass (superior or inferior), an extension of the mass (medial or lateral), the morphology of the mass, and the degree of dentofacial deformity (mild or severe) [12].

Case report

A 28-year-old male patient first visited the Department of Oral and Maxillofacial surgery, Faculty of Dentistry, Mahidol University in 2014, with the chief complaints of a progressively asymmetrical face and malocclusion. Left condylar hyperplasia was the diagnosis from the patient's clinical and radiographic examination at that time. After extensive treatment plan discussions between the surgeon and the patient, the patient chose to wait and see if there was cessation of the condylar hyperplasia instead of undergoing orthognathic surgery and condylectomy. Thus, this case was followed up by performing clinical examinations, radiographs, CT, and bone scintigraphy for approximately eight years. Subsequently, the facial asymmetry and the skeletal discrepancy became more severe (Figure 1). Furthermore, the periodic bone scintigraphy evaluation demonstrated evidence

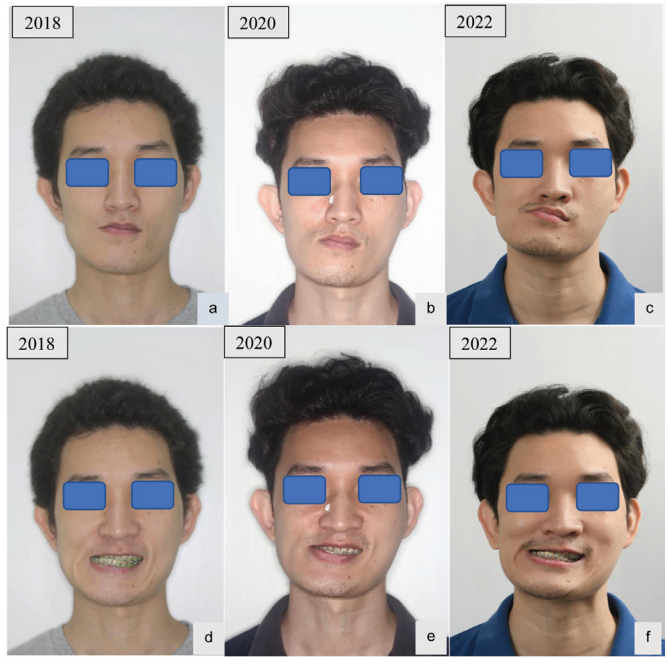


Figure 1 Photographs of the patient's facial profiles taken in 2018, 2020, and 2022. There was progression of facial asymmetry with chin deviation to the right (a, b, c) together with mandibular and maxillary canting (d, e, f) over the period of time.

of active lesions. Based on these findings, the patient chose to undergo low condylectomy concomitant with bimaxillary orthognathic surgeries. The preoperative evaluation revealed that the patient had received orthodontic treatment for 6–8 years. The patient had no medical problems and denied a history of craniofacial trauma. The facial examination revealed facial asymmetry with the chin deviated to the right ~6 mm from the midline of the face, and mandibular and maxillary canting (Figure 1). No TMJ symptoms were detected, except for a clicking sound on both sides. The patient's intraoral examination revealed fixed orthodontic appliances, anterior and right posterior crossbites, and the midline of the lower tooth was shifted 14.5 millimeters to the right of upper dental midline (Figure 2).

Imaging findings (Figure 3):

A panoramic radiograph demonstrated a well-circumscribed radiopaque lesion with a thin corticated rim at the left condyle. The lesion was larger, and the lower dental midlines were shifted to the right compared with the previous radiograph.

Cone beam CT (large FOV) demonstrated exophytic proliferation of the left condyle in the vertical and medial directions, and the size of the condyle was ~2x2x2.5 cm.

Figure 2 Increasing severity of the crossbites, and discrepancy of the dental midlines had been observed over the period. (2018 (a, b, c), 2020 (d, e, f), and 2022 (g, h, i)
The preoperative intraoral examination (g, h, i), revealed anterior crossbite, right posterior crossbite, and discrepancy of the dental midlines (the lower dental midline (green vertical line) was 14.5 mm shifted to the right of the upper dental midline (black vertical line)).

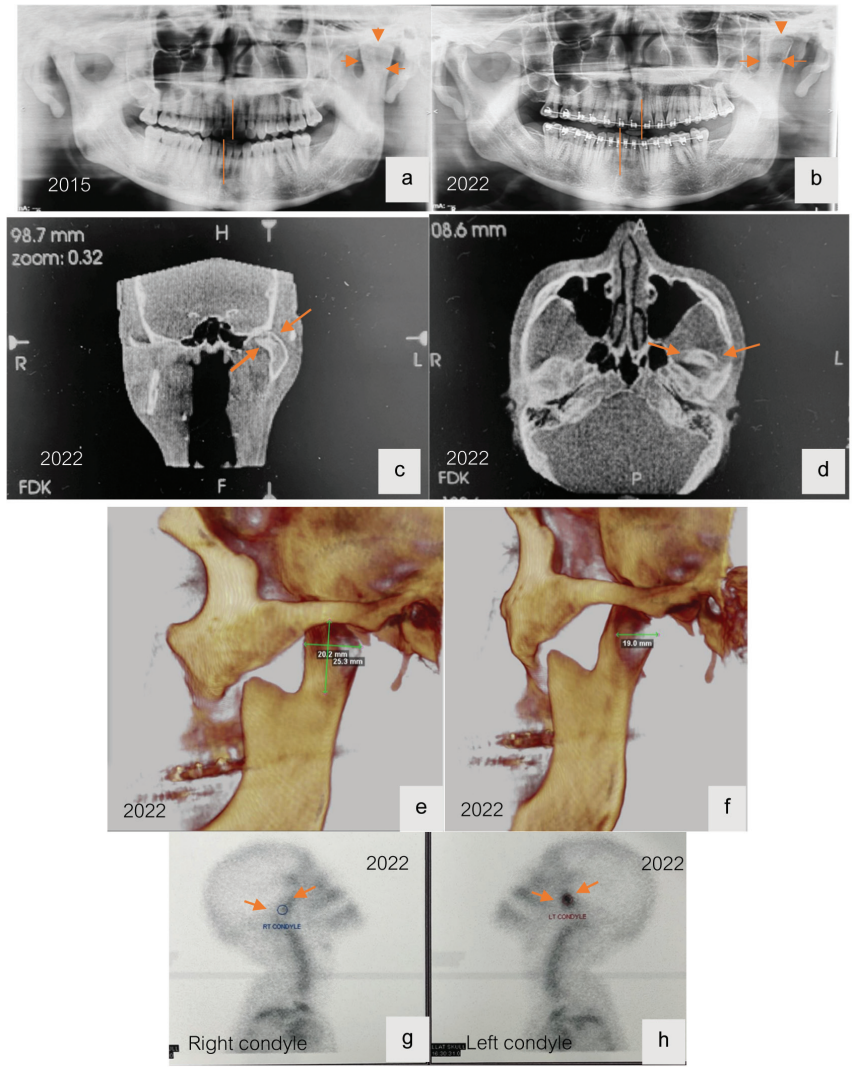


Figure 3 The preoperative panoramic radiograph showed a well-circumscribed radiopaque lesion with a thin corticated rim at the left condyle (b). The lesion became larger, and the lower dental midlines progressively shifted to the right compared with the previous radiograph (a, b). Large FOV cone beam CT coronal and axial views revealed medial expansion of the lesion with a thin cortical rim at the left condyle of the mandible (c, d). The size of the affected condyle was ~20.2 mm, 25.3 mm, and 19.0 mm in sagittal, vertical, and medio-lateral dimension, respectively (e, f). The latest bone scintigraphy performed in 2022 showed active growth of the left condyle (h).

Bone Scintigraphy had been performed from 2014 to 2022 revealing ongoing growth of the left mandibular condyle.

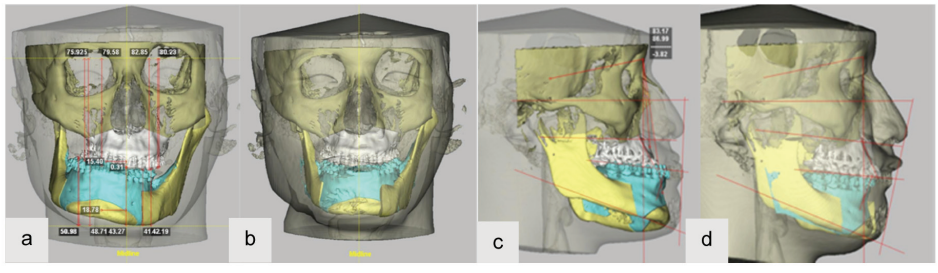
The patient was diagnosed with left condylar hyperplasia type 2B [13] with skeletal class III deformity (canting of the maxilla and mandible, orthognathic maxilla, close configuration, prognathic mandible with normal configuration, and prognathic chin), angle's class III malocclusion (molar and canine) with proclination and protrusion of both upper and lower teeth, and facial asymmetry (according to the cephalometric analysis, Table 1).

Table 1 Cephalometric analysis and postoperative prediction

	Measurement	Male norm	SD	Preop	Postop		Measurement	Male norm	SD	Preop	Postop
Maxilla	FH-SN (dg)	5.36	2.63	7.5	7.5	Lower tooth	1-NB(dg)	29.13	6.09	25	23
	SNA (dg)	85.66	4.38	82.5	85		1- NB (mm)	6.19	2.43	6	4.5
	COA (mm)	97.03	4.15	87	90		1-MP (dg)	97.34	5.45	87	90
	A to perpend. plane (mm)	4.33	4.6	2	3	Jaw relation	ANB (dg)	3.69	2.05	-4	5
	SN-PP (dg)	8.18	3.52	4	18		MP-PP (dg)	20.36	6.79	18	15
	PNS ANS -FH	-0.18	3.55	4	9		SN-OP (dg)	14.01	4.17	18	17
Mandible	SNB	81.96	3.74	87	80		WITS (mm)	-3.24	2.37	-12.5	1
	Co-GN (mm)	126.35	4.95	127	118.5	Tooth relation	1 to 1 (dg)	125.55	8.34	136	135
	Pg. to Perpend. Plane (mm)	0.3	7	14	0		OVB (mm)	1.6	1.1	3	2.5
	SN-GoGN	28.55	5.88	30.5	31		OVJ (mm)	2.66	1	-7.5	4
	MP-FH	21.58	5.57	22	25	Soft tissue	NLA (dg)	91.12	7.98	80	95
	Go.angle (dg)	117.35	6.79	132	125		FCA (dg)	9.1	4.08	-1	12
Chin	SN-Pog (dg)	82.17	3.77	88	82		UFH (mm)	50.66	3.34	47	48
	Pog-NB (mm)	0.43	1.62	3	3		LFH (mm)	74.44	4.55	72	67
Upper tooth	1 - NA (dg)	21.61	5.41	25	17		ULL (mm)	25.18	1.97	19	20
	1 - NA (mm)	3.26	2.08	4.5	2		LLL (mm)	49.02	3.16	58	45
	1 – Npog (mm)	8.66	3.25	-3	6		LCT (dg)	109.7	9.14	87	86
	1 - PP (dg)	119.68	6.26	121	115		L-lip to E-line (mm)	1.59	2.06	0	2
	ADH	30.6	2.8	23	24						
	PDH	20.3	1.98	19	22						

Frankfurt Horizontal plane-FH, Sella and Nasion – SN line, A point- A, B point- B, Sella-Nasion-A Point angle-SNA, Condylion to A point – COA, Palatal plane – PP, Posterior nasal spine- PNS, Anterior nasal spine- ANS, Perpendicular plane- Perpend. Plane, (Gonion-Gnathion) distance – GoGn, Mandibular plane- MP, Gonion angle - Go.angle, Pogonion - Pog upper incisor – 1, Anterior dental height- ADH, Posterior dental height- PDH, Occlusal plane- OP, lower incisor – 1, Overbite- OVB, Overjet- OVJ, Nasolabial angle- NLA, Facial angle- FCA, Upper facial height- UFH, Lower facial height- LFH, Upper lip length- ULL, Lower lip length- LLL, Lip-chin-throat angle- LCT, Lower lip- L-lip, Esthetic line- E-line

The treatment plan for this case was low condylectomy of the left mandibular condyle to eliminate the pathology and simultaneous correction of the facial deformity by bimaxillary orthognathic surgery. The intraoperative sequences began with a Le fort I osteotomy, repositioning of the maxilla and plates and screw fixation, low condylectomy on the left mandibular condyle, then Bilateral Sagittal Split Ramus Osteotomy (BSSRO). The orthognathic surgical plan details and its predicted tracing are presented in Figure 4 and Table 1. A maxillary vestibular incision was made and a Le fort I osteotomy was performed. The maxilla was repositioned with an intermediate splint, then the segments were immobilized with plates and screws. The tumor and the left condylar head were exposed using an endaural approach, low condylectomy, and hemostasis was achieved. The resected condyle was sent for histopathological investigation. Bilateral mandibular vestibular incisions were made, and



Maxilla					Mandible				
Right side			Left side		Right side			Left side	
First Molar	Caine	Incisor	Canine	First molar	First Molar	Caine	Incisor	Canine	First molar
0 mm	↓1mm	0 mm	↑2mm	↑4.5 mm	↓3.5 mm	↓3 mm	↓1 mm	↑2 mm	↑5.5 mm

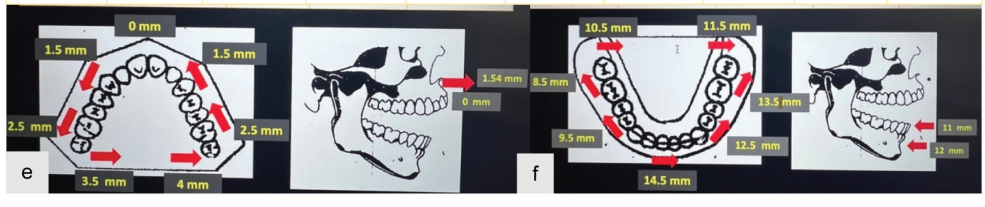


Figure 4 Three-dimensional postoperative prediction for the bimaxillary orthognathic surgery was performed to give more information to the patient about the outcome of the surgical plan (a and c were the preoperative facial profiles while c and d were the postoperative prediction). The surgical plan was to correct the maxillary canting by superior positioning the left side of the maxilla (the new positions of the left first maxillary molar and the left maxillary canine were 4.5 mm and 2 mm upward, respectively, while the right maxillary canine was 1 mm downward). The maxilla was also moved forward and rotated counterclockwise, resulting in an advancement of point A (1.54 mm), while the position of the maxillary central incisors was not changed (e). According to the adjusted maxilla, the mandible was repositioned backward and counterclockwise, resulting in a setback of point B and the mandibular central incisor (12 mm and 11 mm, respectively) (f).

BSSRO was performed. The mandible was rearranged to the appropriate position using the final splint and the mandibular segments were fixed with miniplates and screws. The buccal fat pad was harvested from the right cheek to complete the interposition graft on the condylectomy site, then flap approximation and suturing was performed (Figure 5). The tumor was histopathologically consistent with osteochondroma (Figure 6). Postoperative follow-ups performed every week in the first month, monthly for the first three months,

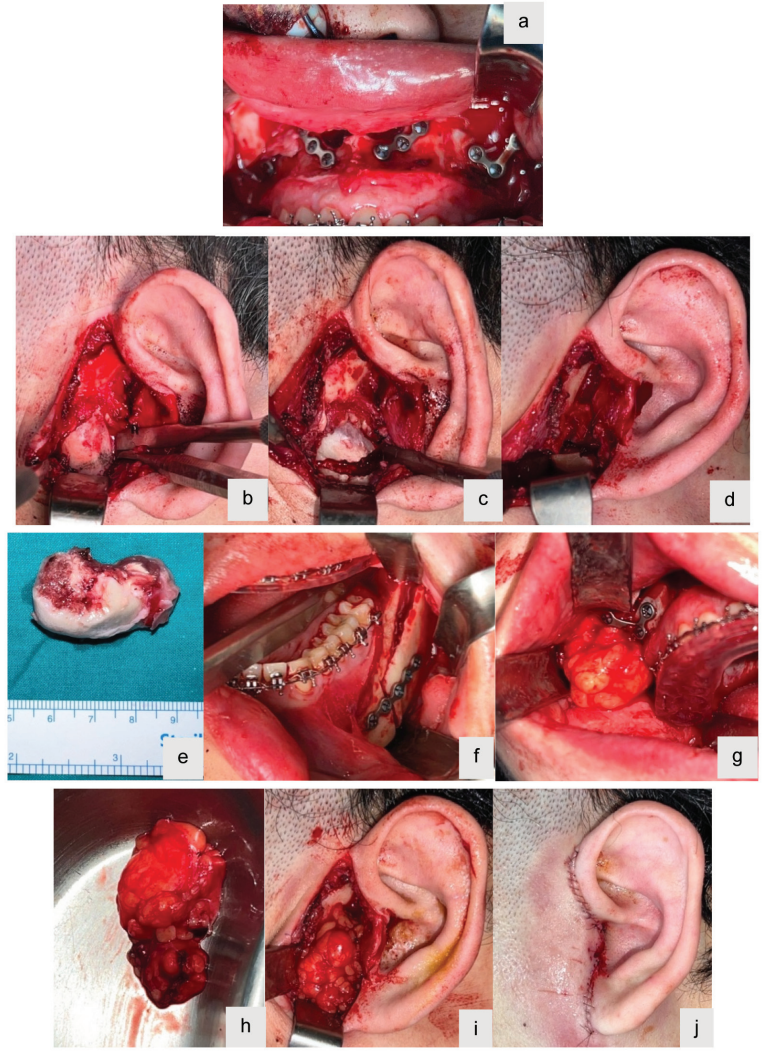


Figure 5 Le fort I osteotomy was approached using a vestibular incision. Guided by the intermediate splint, the maxilla was repositioned, and the segments were fixed with plates and screws (a). The tumor and left condylar head were exposed via an endaural approach and low condylectomy was achieved (b, c, d). The resected condyle was further delivered for histopathological investigation (e). Performing BSSRO, the mandible was rearranged to the planned position by applying the final splint, then the mandibular segments were fixed with miniplates and screws (f). The buccal fat pad was harvested from the right cheek to complete the interposition graft on the condylectomy site (g, h, i) prior to flap approximation and suturing (j).

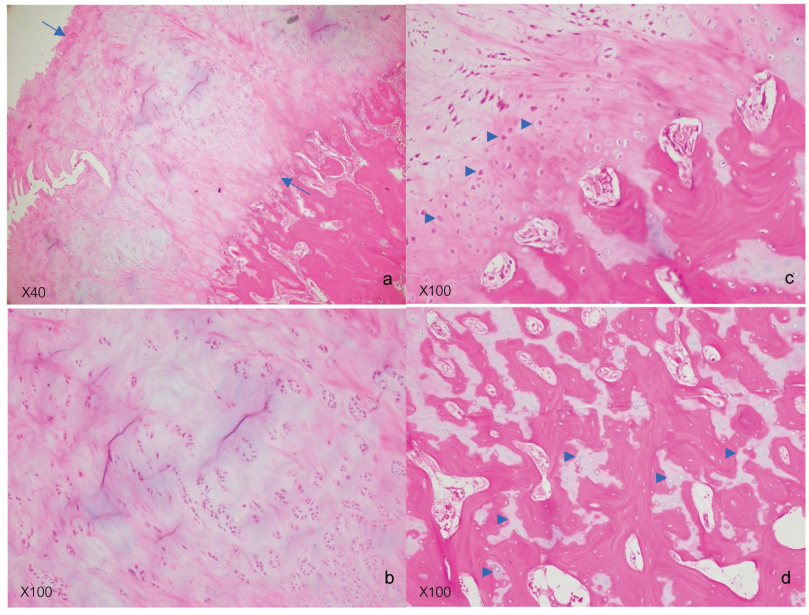


Figure 6 (Courtesy of Dr. Theerachai Kosanwat) Histopathologic features (hematoxylin and eosin stained) showed the bony structure is covered by a thick layer of a cartilaginous cap (a) (area between the blue arrows). The cap consists of cartilaginous tissue that is frequently arranged in a haphazard manner (b). In some places, the cartilaginous tissue is arranged in four layers including a fibrous layer, undifferentiated mesenchymal layer, cartilage layer, and calcified cartilage layer (c). The hyperplastic chondrocytes (blue arrowheads) of the cartilage layer are infrequently observed. Endochondral ossification is presented. (d) The cartilage is continuous with the underlying bone and cartilage islands (blue arrowheads) in the trabeculae bone are frequently detected. These histopathological features are consistent with osteochondroma.

every 3 months in the first year, every 6 months in the next two years, then annual follow-up. The patient's lip paresthesia and facial weakness (House Brackmann Grade III) was detected in the first 3 months however, three months later, these complications had resolved. The facial asymmetry was corrected, and the correct occlusion was achieved. The patient was satisfied with his improved mastication and was more confidence in his appearance, had no difficulty in mouth opening with a maximum mouth opening of ~3.5 cm and the latest radiographic examination (6-month postoperation) demonstrated that the tumor had not recurred (Figure 7).

Discussion

Osteochondroma (OC) is a relatively common finding in the skeleton, occurring frequently in the metaphyseal region of the long bones. It is also found in the ribs, scapulae, clavicles, and vertebrae [6]. The occurrence of OC is rarely reported in the maxillofacial region. Most extracondylar OCs in this region occur in the coronoid process. However, cases have been reported in the posterior maxilla, maxillary sinus, zygomatic arch, mandibular body, and symphysis [9]. Mandibular condyle OC is rare. Those involving the mandibular condylar region have been found most often on the medial aspect of the mandibular condyle (52%), followed

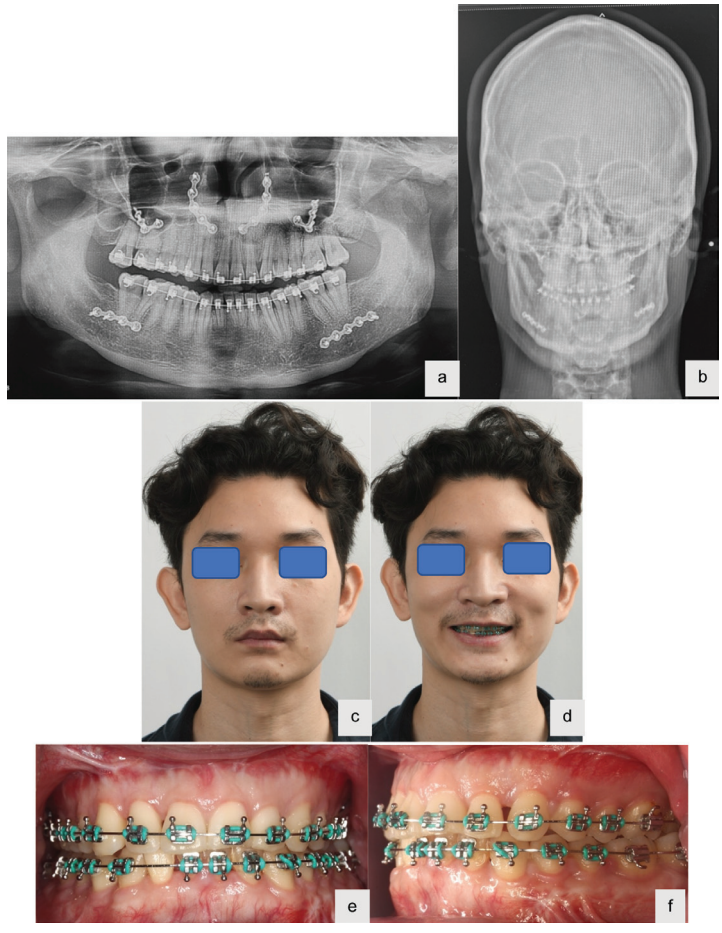


Figure 7 The six-month- postoperative follow-up. The panoramic and PA skull radiograph indicated that there was no recurrence of the tumor (a, b). No facial weakness was observed, and the facial asymmetry was corrected together with achieving the correct occlusion (c, d, e, f).

by an anterior location (20%), but rarely on the lateral or superior positions (1%) [8].

Osteochondromas are often slow growing. The presentation of condylar OC includes the development of facial asymmetry, malocclusion, crossbite on the contralateral side and lateral open bite on the affected side, deviation on opening, hypomobility, pain, and clicking [6]. When considering a diagnosis of mandibular condyle OC, other differential diagnoses must be considered. The clinical differential diagnosis of slow-growing masses of the mandibular condyle should include giant cell tumor, condylar hyperplasia, fibro-osseous lesion, vascular malformation, osteoma, chondroma, and osteochondroma [9].

Considerable confusion exists between OC and unilateral condylar hyperplasia. However, the treatment plan for each these conditions are different. It is mandatory that clinic-pathological and radiological differentiation should be performed between the two pathologies. Unilateral condylar hyperplasia manifests clinically and radiographically as an enlarged condylar process, whereas the OC is seen in most cases as a globular projection extending from the medial margins of the condylar head. Osteochondroma growth continues and progresses much longer, including after skeletal growth ceases, whereas, with condylar hyperplasia, skeletal growth ceases [14]. Bone scintigraphy is a valuable tool for assessing the activity of condylar growth. The radionuclide uptake ratio of the bilateral

condyles can be generated. An uptake difference above 10% is indicative of condylar hyperactivity [15].

Pre-operative CT plays a crucial role in treatment planning for these tumors. CT clearly depicts the continuation of the cortex and medulla of the parent bone with that of the tumor, the most diagnostic feature of OC. Although 3-D CT can also demonstrate a normal condylar head, the coronal CT view is the most diagnostic. The coronal CT view reveals a growth arising medially from the morphologically normal condyle however, other views may not be useful in demonstrating this picture. Condylar hyperplasia will be seen only as a uniform enlargement of the condylar head [14]. In this case, large FOV cone beam CT coronal and axial views revealed the medial growth of the lesion with a thin cortical rim at the left condyle of the mandible.

According to a classification system for conditions causing condylar hyperplasia [13], the patient was classified as condylar hyperplasia type 2B because the abnormality was found at the left condyle of the mandible causing facial asymmetry and malocclusion. The radiographic features indicated an enlargement of the left mandibular condyle caused by exophytic tumor growth, resulting in a predominantly vertical growth direction. The histopathological report confirmed that the exophytic mass was osteochondroma. Condylar hyperplasia is mainly diagnosed from the patient's clinical and imaging examination, whereas osteochondroma of the mandibular condyle requires histopathological investigation [14]. Osteochondroma is the most common benign tumor of the axial skeletal bones, however, is rarely related to the facial bones. Osteochondroma of the mandibular condyle and condylar hyperplasia can cause asymmetric facial deformities, altered dental occlusion, and facial appearance that motivate the patients to seek treatment. Determining the type of condylar hyperplasia is crucial for treatment planning concerning whether condylectomy should be included (e.g., type 2B, 3, and 4 are tumors requiring condylectomy). TMJ osteochondroma and their management have been reported depending on the severity of the malocclusion, facial asymmetry, the extent of the tumor, age, and the patient's health status. The types of surgical treatments for osteochondroma vary from resection without reconstruction (low-condylectomy), resection (total condylectomy) with TMJ reconstruction (autogenous/alloplastic), combined surgery, including orthognathic correction and low-condylectomy or TMJ reconstruction [12]. There were several reports of TMJ osteochondroma with no or mild facial deformity. Their management required condylectomy combined with orthodontic treatment or only postoperative maxillomandibular fixation without concomitant surgeries [17-19]. In cases of large masses and their advanced extension, radical condylectomy with immediate total joint alloplastic reconstruction was also reported [19].

In our case, the patient chose to wait and see if the abnormal condylar growth ceased. A treatment plan comprising condylectomy on the left mandibular condyle (in case there was continuous growth of the left mandibular condyle) concomitant with bimaxillary orthognathic surgery was discussed with the patient at his first visit. There was increased facial asymmetry and evidence of ongoing active growth of the left condyle from the bone scan, hence the patient was urged to undergo the treatment at every follow-up visit. The final decision was made, and the operation was performed after he had been followed up for ~8 years. Condylectomy was included in the treatment plan rather than only bimaxillary orthognathic surgery. In contrast, in cases where there is no evidence of ongoing growth of the left mandibular condyle, only orthognathic surgery might be sufficient to correct the patient's facial deformity. Moreover, the severity of facial deformity would be much less than the existing deformity and the orthognathic surgical plans would

have been less complicated if the surgical interventions had been performed earlier. However, consideration of the patient's readiness was also required.

There are studies that suggested an intraoperative sequence of initially performing condylectomy, followed by orthognathic surgery with a Le Fort I osteotomy after BSSRO to achieve positional stability of the mandible and the maxilla during the operative procedure [20, 21]. Based on the surgical model set-up, a Le Fort I osteotomy was initially considered. Because low condylectomy was initially performed, the position of the mandible that was used as the reference for the intermediate splint for the maxilla repositioning would be unreliable. Therefore, in this case, the low condylectomy was accomplished after the maxilla was rearranged and fixed so that we could obtain a further reference for the mandibular orthognathic surgery. Additionally, from the same incision for BSSRO, the buccal fat pad was harvested from the right cheek and placed into the gap on the condylectomy site to eliminate a dead space and reduce the chance of TMJ ankylosis. The buccal fat pad was selected to be an interposing material for this case because acquiring it was not invasive and it was adequate for filling the gap. Due to the surgical approaches for BSSRO and condylectomy (via preauricular approach), the inferior alveolar nerves and the facial nerve could be injured. In this case, lower lip paresthesia and facial weakness (House Brackmann Grade III) was reported in the first three months of the postoperative follow-up. However, three months later, these complications had resolved. Compassionate communication helps the patients in the decision making and being cooperative during case management. Correcting the facial deformity simultaneously with eliminating the neoplasm was the goal for this patient with TMJ osteochondroma, then the patient underwent bimaxillary orthognathic surgery with low condylectomy. Although osteochondroma is rare in the facial region, the clinical examination and radiographic features, including periodic scintigraphy, play a crucial role in identifying the abnormality leading to the appropriate management. Additionally, the consideration of the intraoperative sequence depends on the stability of the relevant structures; maxilla, mandible, and the affected condyle because the orthognathic surgery requires a constant positioning reference. Postoperative follow-ups to evaluate the postoperative outcomes (e.g., correcting the facial deformity/malocclusion), complications (paresthesia, facial weakness) and the recurrence of the tumor should be considered. Long-term observation of the tumor recurrence by annual radiographic examination has been planned and CT will be performed if there is a suspected recurrence found on the radiograph.

Declaration of patient's consent

The patient understands that his name will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed. The patient's pictures were illustrated in the report with permission from the patient. The consent form was obtained.

Conclusion

Facial asymmetry arising from TMJ osteochondroma needs precise information from the clinical examination and imaging findings for the diagnosis, resulting in an accurate treatment plan.

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