Osteochondroma of the coronoid process: A case report and review of the literature

TIANJUN LAN[1,2*], XIN LIU[1,2*], PEI-SHENG LIANG[1,2] and QIAN TAO[1,2]

[1]Department of Oral and Maxillofacial Surgery, Guanghua School of Stomatology, Hospital of Stomatology; [2]Guangdong Provincial Key Laboratory of Stomatology, Sun Yat-Sen University, Guangzhou, Guangdong 510055, P.R. China

Received November 4, 2018; Accepted May 13, 2019

DOI: 10.3892/ol.2019.10537

Abstract. Osteochondroma (OC) is considered the most common tumor of the axial skeleton, although it is relatively uncommon in the craniofacial region. The present study describes an atypical case of OC of the coronoid process. A 34-year-old woman presented with severely limited mouth opening (5 mm) and swelling of the right zygoma. Cone-beam computed tomography (CBCT) revealed a mushroom-shaped outgrowth from the coronoid process to the inner surface of the zygomatic arch, forming a pseudojoint. The patient was treated with coronoidectomy via an intraoral approach. Histopathological examination revealed features suggestive of OC. Subsequently, the patient was able to open their mouth, and there was no evidence of recurrence or post-operative complications in the 21-month follow-up. A review of the literature revealed only 38 histologically proven cases of coronoid OC in the past 30 years (1989-2018). The incidence of the disease was higher in men compared with that in women (male:female, 2.17:1), and the median age at onset was 28.7 years, with a range of 5-57 years. Gradual limitation of mouth opening and facial asymmetry are the most noticeable symptoms. Water's view and submentovertex projection of the zygomatic arch may be useful in identifying the tumor and its association with the zygoma, while CT and CBCT permit a detailed visualization of the location and density of the tumor. Coronoidectomy is the preferred treatment option, and the prognosis is excellent, with no evidence of recurrence or malignant transformation.

Correspondence to: Dr Qian Tao, Department of Oral and Maxillofacial Surgery, Guanghua School of Stomatology, Hospital of Stomatology, Sun Yat-Sen University, 56 Lingyuanxi Road, Guangzhou, Guangdong 510055, P.R. China
E-mail: taoqian@mail.sysu.edu.cn

*Contributed equally

Key words: osteocondroma, coronoid process, clinical characteristics, treatment, prognosis

Introduction

Osteochondroma (OC) or osteocartilagenous exostosis, a cartilage-capped osseous lesion that protrudes from the surface of the affected bone, is the most common tumor of the axial skeleton, accounting for 35-50% of benign bone tumors, and 8-15% of bone tumors overall (1,2). OC frequently arises from the long bones (3), such as the proximal metaphysis of the tibia or the distal metaphysis of the femur, and rarely occurs in the craniofacial region (<1% of cases) (4,5). The embryonic development of the mandibular condyle from cartilaginous ossification makes it the most frequent facial site of this type of tumor (5). Although extremely rare, involvements of the coronoid process (6), the posterior maxillary region (7), the maxillary sinus (8) and the body (9), symphysis (10) and ramus of the mandible (11) were also reported. Different from OCs of the long bones, craniofacial OCs occur at older ages (mean age, 36.4 years), and grow slowly long after the end of puberty (12). The etiology of the tumor is not fully understood, and the most accepted theory was hypothesized by Lichtenstein (13), which suggests that periosteum had the pluripotentiality to give rise to chondroblasts or osteoblasts, and that OC results from metaplastic change in the periosteum.

The present study reviewed the literature concerning coronoid OCs from 1989-2018 and also describes the case of a patient treated surgically and followed up for 21 months in the Hospital of Stomatology (Guangzhou, Guangdong, China). The case involved a giant OC on the coronoid process, and the patient presented with facial asymmetry and a limited ability to open her mouth.

Case report

A 34-year-old woman presented to the Hospital of Stomatology with progressive restriction of mouth opening over a period of 20 years and facial asymmetry with swelling in the right zygomatic region within the past 2 years. No history of trauma was reported. A physical examination revealed swelling in the right zygomatic arch region, facial asymmetry and the ability to open their mouth only 5 mm. There were no associated temporomandibular joint (TMJ) complaints such as pain or clicking when opening their mouth.

A panoramic radiograph showed an enlarged right coronoid process (Fig. 1). Cone-beam computed tomography (CBCT) revealed a mushroom-shaped outgrowth from the

lateral aspect of the coronoid process to the inner surface of the zygomatic arch, with outward expansion, forming a pseudojoint (Fig. 2). A diagnosis of OC of the right coronoid process was made according to the clinical and radiographic features. The patient was then scheduled for right coronoidectomy, performed through an intraoral approach as previously described (5,14).

Considering that the patient suffered from a serious limitation of mouth opening, all procedures were conducted under general anesthesia via naso-tracheal intubation. The patient was taken to the operating room and, after naso-tracheal intubation, was prepped and draped for transoral incisions. An incision was made along the anterior border of the ramus to the tip of the coronoid process. The mucoperiosteal flaps were raised superiorly to the sigmoid notch and lower portion of the coronoid, and by blunt dissection, the coronoid process and the tumor were visualized. The tumor and the coronoid process were removed without difficulty using a fissure bur and a chisel.

Post-operative panoramic radiograph and CBCT showed that the tumor and the right coronoid process were totally excised (Figs. 3 and 4). The mass measured ~4x1.5x1.5 cm (Fig. 5A). Histologically, the tumor showed the presence of three layers from the surface to the inside: Fibrous tissue, cartilaginous tissue and cancellous bone (Fig. 5B and C), which confirmed the diagnosis of OC. Three highly qualified pathologists made the diagnosis independently. Hematoxylin staining was performed as follows: The specimen was fixed in 4% paraformaldehyde at 4˚C for 24 h and transferred to 19% ethylene diamine tetraacetic acid (EDTA) solution for decalcification at room temperature for ~2 months. After thoroughly decalcification, the specimen was then dehydrated as follows: 70% ethanol (60 min), 80% ethanol (40 min), 95% ethanol (30 min), 100% ethanol (25 min) at room temperature, and embedded in paraffin. Following embedding, the specimen was sliced sagitally in to 7-μm thick slices. The slices were deparaffinized in xylene, rehydrated in 100% ethanol, 95% ethanol, 80% ethanol and 70% ethanol for 2 min each, and then stained with hematoxylin and eosin for 1 min, all at room temperature.

Postoperatively, the patient was able to open their mouth 36 mm. After a follow-up period of 21 months, there was no evidence of recurrence and the patient was asymptomatic. The patient is still being followed up every 6 months, and the follow-up will be performed over an extended period.

Literature review

The present review was performed using a computer-assisted search of Medline Industries, Inc. (www.medline.com). The literature published in the English language on OC of the mandibular coronoid process from January 1989 to December 2018, concerning clinical characteristics, histopathological features and treatment, were reviewed. The criterion for inclusion was any case with a histopathological description or photomicrograph indicating OC. Cases were excluded if histopathological characteristics of the lesion were not shown, even those with a diagnosis of OC.

The review of the literature revealed only 39 reports of OC involving the coronoid process within the last 30 years (5,6,15-48), plus the present case (Tables I and II). The median age at onset was 28.7 years (range, 5-57 years), with the largest number of cases observed in patients 20-30 years old. Men were more commonly diagnosed (66.7%) than women. Unilateral masses were more frequently described (32 cases, 82.1%) and a slight tendency for involvement of the left coronoid process (51.3%) was revealed. These findings were generally in agreement with previous studies (6,28).

As mentioned in several reported cases, the disease was predominately characterized by a lengthy history (ranging from 3 months to 20 years) of progressive reduction in the ability of mouth opening (37 cases, 94.9%). Later signs included total trismus and appreciable swelling in the zygomatic region, visible as facial asymmetry (32 cases, 82.1%). Pain was not a common symptom (4 cases, 10.3%).

Panoramic radiography usually showed a sessile or pedunculated bony mass in the affected coronoid process. Water's view may be useful in identifying coronoid tumors and their relation to the wall of the maxillary sinus and the zygoma (25). To visualize the exact shape, location and density of the tumor, 3-dimensional CT and CBCT were performed and are considered as the 'gold standard' for an accurate diagnosis (6,36). A pseudojoint formation between the mass and the protruded zygoma (Fig. 2) was observed in the majority of the cases (38 cases, 97.4%), a condition that was first described by Oscar Jacob in 1899 and was hence termed Jacob's disease (49).

From the literature, coronoidectomy was recorded as the preferred treatment. Data on the surgical approaches were present in 38 cases, and were used as follows: Extraoral in 24.3% of cases; intraoral in 64.9% of cases and combined intra- and extraoral approaches in 10.8% cases, with the intraoral approach used most often.

Follow-up data were included in 25 of the 39 cases and showed that the prognosis of coronoid OC was excellent, with no recurrences or malignant transformations reported.

Discussion

An extensive review of the English literature within the last 30 years revealed a total of 435 patients with OC in the craniofacial region. The most frequently affected site was the mandibular condyle (384 cases, 88.3%), followed by the coronoid process (8.7%). However, involvement of the posterior maxillary region (7), maxillary sinus (8), and the body (9), symphysis (10) and ramus (11) of the mandible were also reported. A previous review of the literature by Sreeramaneni *et al* (6) identified 39 cases of coronoid OC up until December 2010, after which there were only 12 new cases reported. Reports with only photographic evidence of OC were not included in the present review.

The pathogenesis of OC has not yet been elucidated. Langenskiold (50) hypothesized that such lesions resulted from cells in the undifferentiated layer that were displaced from the epiphysis to the metaphyseal area. However, this may only explain the emergence of lesions in the condylar region. Another theory hypothesized that there were accumulations of embryonic cells at the points of tendon attachments, and that the continuous strain on tendons may stimulate the cartilaginous potential of the embryonic cells (51). The most widely accepted theory was hypothesized by Lichtenstein (13), who suggested that pluripotential cells in the periosteum have the

Table I. Clinical characteristics, treatments and outcomes of cases of coronoid osteochondromas reported in Medline.

First author/s, year	Age, years	Sex	Location	Symptoms	Surgical approach	Follow-up, months	Recurrence	(Refs.)
Mohan Choontharu *et al*, 2018	16	F	Left	A	Intraoral	6	None	(48)
Mohanty *et al*, 2016	18	M	Right	LMO, A	Extraoral	36	None	(47)
Dandriyal *et al*, 2015	20	M	Left	LMO, P	Intraoral	54	None	(5)
Sawada *et al*, 2015	14	M	Left	LMO	Intraoral	6	None	(46)
Losa-Munoz *et al*, 2014	42	M	Right	LMO, A	Intraoral	NA	NA	(45)
Fan *et al*, 2014	20	M	Left	LMO, A	Combined	20	None	(44)
Stringer *et al*, 2013	27	M	Left	LMO, A	Intraoral	NA	None	(43)
Aoki *et al*, 2013	18	M	Right	A, P	Intraoral	15	None	(42)
Ruiz and Lara, 2012	28	M	Bilateral	LMO	Combined	36	None	(41)
Ajila *et al*, 2012	28	M	Left	LMO, A	Intraoral	12	None	(40)
Coll-Anglada *et al*, 2011	52	F	Right	LMO, A	Intraoral	6	None	(38)
D'Ambrosio *et al*, 2011	39	M	Left	LMO	Intraoral	Several years	None	(39)
Acosta-Feria *et al*, 2011	55	M	Right	LMO, A	Extraoral	20	None	(37)
Sreeramaneni *et al*, 2011	45	F	Left	LMO, A	Combined	3	None	(6)
Yesildag *et al*, 2010	16	M	Right	LMO, A	Extraoral	14	None	(36)
Zhong *et al*, 2009	39	F	Bilateral	LMO, A	Intraoral	9	None	(35)
Etoz *et al*, 2009	43	F	Right	LMO, A	Intraoral	6	None	(33)
Thota *et al*, 2009	15	M	Bilateral	LMO, A	Intraoral	14	None	(34)
Dede *et al*, 2007	20	M	Bilateral	LMO, A	Intraoral	NA	NA	(32)
Akan and Mehreliyeva, 2006	24	M	Bilateral	LMO	Intraoral	NA	NA	(30)
Villanueva *et al*, 2006	44	F	Left	LMO, A	Intraoral	10	None	(31)
Capote *et al*, 2005	23	F	NA	LMO, A, P	Intraoral	12	None	(29)
Emekli *et al*, 2002	21	M	Right	LMO, A, P	Extraoral	NA	NA	(27)
	26	F	Right	LMO, A	Intraoral	6	None	
Escuder *et al*, 2001	24	M	Left	LMO, A	Intraoral	NA	NA	(26)
	16	NA	Bilateral	LMO	Intraoral	12	None	
Roychoudhury *et al* 2002	32	M	Left	LMO, A	Extraoral	12	None	(28)
Hernandez-Alfaro *et al*, 2000	22	M	Left	LMO, A	Extraoral	NA	NA	(25)
Chichareon *et al*, 1999	5	M	Right	LMO, A	NA	NA	NA	(24)
Manganaro, 1998	26	F	Left	LMO, A	Intraoral	Several weeks	None	(23)
Chen *et al*, 1998	57	F	Left	LMO, A	Extraoral	72	None	(22)
Gross *et al*, 1997	22	M	Left	LMO, A	NA	NA	NA	(21)
Constantinides *et al* 1997	31	M	Right	LMO, A	Extraoral	12	None	(20)
Kermer *et al*, 1996	40	M	Left	LMO, A	Extraoral	NA	NA	(19)
Çenetoğlu *et al*, 1996	19	M	Left	LMO, A	Intraoral	NA	NA	(18)
Kerscher *et al*, 1993	45	M	Left	LMO	Intraoral	NA	NA	(17)

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Table I. Continued.

First author/s, year	Age, years	Sex	Location	Symptoms	Surgical approach	Follow-up, months	Recurrence	(Refs.)
Asanami *et al*, 1990	17	M	Left	LMO, A	Combined	NA	NA	(15)
Totsuka *et al*, 1990	37	F	Left	LMO, A	Intraoral	NA	NA	(16)
Present study	34	F	Right	LMO, A	Intraoral	21	None	

M, male; F, female; LMO, limitation of mouth opening; A, asymmetry; P, pain; NA, not available.

Table II. Summary of clinical features of coronoid osteochondromas.

Clinical features	Value
Side, n (%)	
Left	20 (51.3)
Right	12 (30.8)
Bilateral	6 (15.4)
NA	1 (2.6)
Sex, n (%)	
Male	26 (66.7)
Female	12 (30.8)
NA	1 (2.6)
Age, years	
Mean	28.7
Range	5-57
Symptoms, n (%)	
Limitation of mouth opening	37 (94.9)
Asymmetry	32 (82.1)
Pain	4 (10.3)

Percentage values are based on the 38 patients reviewed, and 1 case currently reported (n=39).

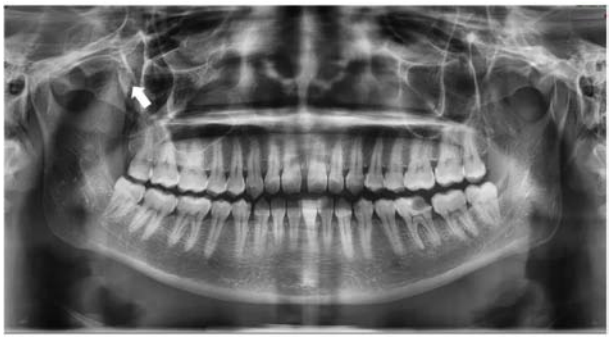

Figure 1. Panoramic radiograph revealing a bulge in the right coronoid process.

potential to form chondroblasts or osteoblasts and result in OC.

OCs can occur independently or as part of an autosomal dominant disorder known as hereditary multiple OC (HMO) syndrome (41). In the literature, of the patients with HMO syndrome, only 2 had lesions in the craniofacial region (41,52). The discrimination of these two types is important, as sarcomatous changes are rare in solitary OCs (1-2%), but do occur in 5-25% of HMO cases (53,54).

Due to the rarity of its occurrence and insidious onset, OC arising from the coronoid process is often overlooked. A coronoid OC should be suspected when patients present with a progressively worsening ability to open their mouth and facial deformity. Due to the limitation in the ability to open the mouth, it is important to differentiate this disease from TMJ disorders or masticatory muscle tendon-aponeurosis hyperplasia (55), the latter of which is more rarely observed clinically.

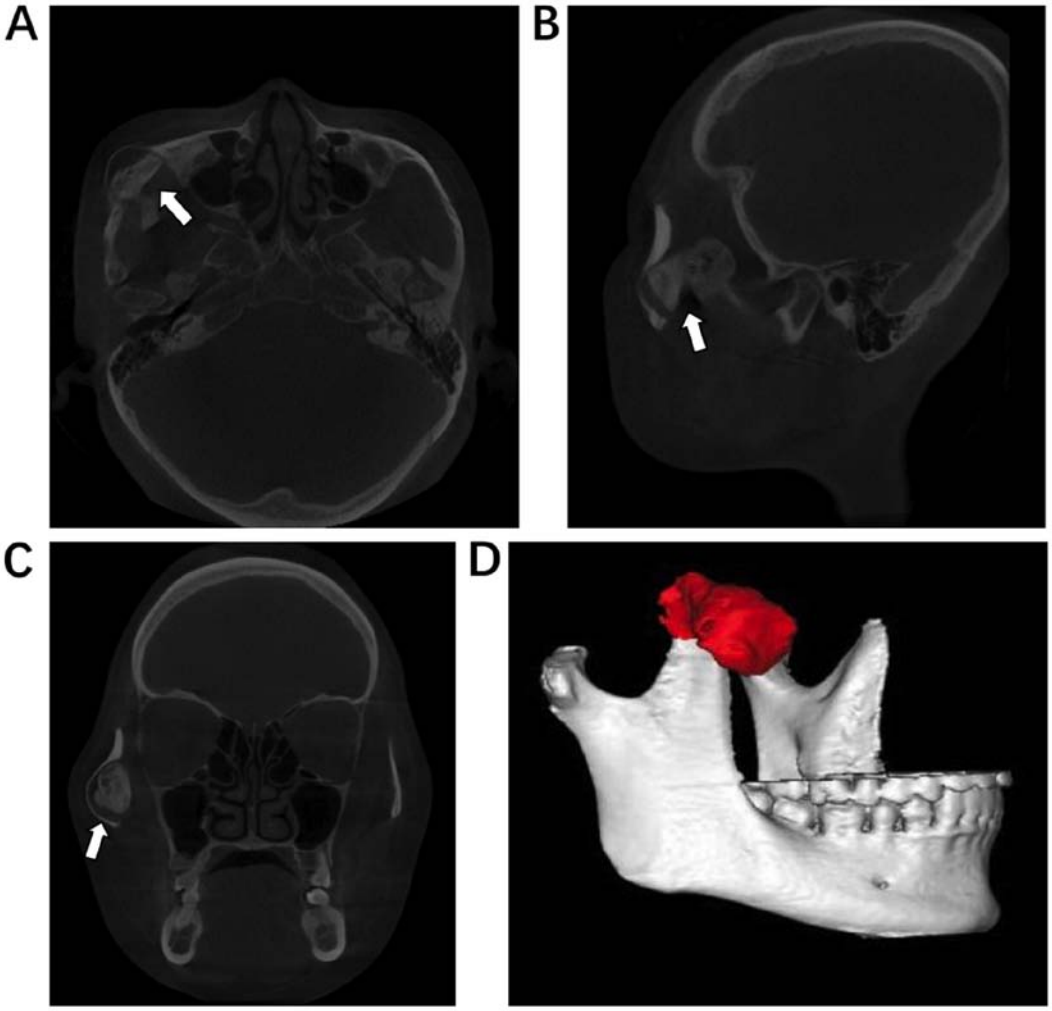

Figure 2. Preoperative CBCT and 3-dimensional reconstruction. A CBCT scan showed a mushroom-shaped mass forming a pseudojoint with the enlarged and protruded right zygoma. (A) Axial plane, (B) sagittal plane and (C) coronal plane. (D) 3-Dimensional reconstruction of the tumor and the mandible. The tumor is marked in red. CBCT, cone-beam computed tomography.

CT is considered as the gold standard for diagnosing OC and provides accurate details regarding the location of the tumor, its density and its relation to adjacent structures (30,36), all of which are valuable when planning the course of treatment. However, CT exposes patients to high doses of radiation, and thus, its use should comply with appropriate guidelines. For younger patients, or those with small morphological alternations that can be clearly discerned by image examinations with less radiation exposure, the unnecessary use of CT should be prevented. Recently, CBCT, being an ideal substitute for CT for the diagnosis of abnormalities in the craniofacial region, has been extensively applied, owing to its lower radiation dosage. Furthermore, submentovertex projection of the zygomatic arch permits a clear visual of the coronoid tumor and the zygomatic arch, which may be more economical and less time consuming for an early diagnosis of tumors in the coronoid process.

Histologically, OC reveals the presence of bony trabeculae covered by a cartilaginous cap and fibrous tissue (56). When considering the differential diagnosis of OC, the possibilities of other lesions, such as bizarre parosteal osteochondromatous proliferations, osteoma, hyperplasia, giant cell tumors and chondroma, must also be considered (5,57). Rarer bony tumors have included chondroblastoma, osteoblastoma, chondrosarcoma, osteosarcoma and metastatic tumors (12).

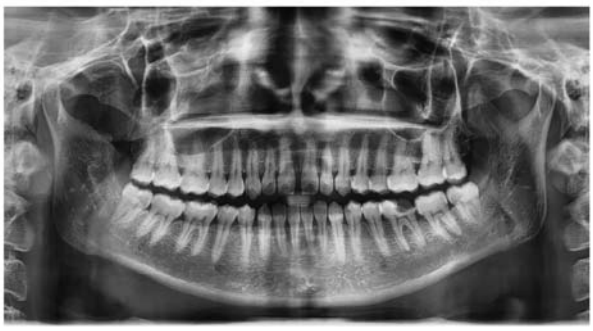

Figure 3. Postoperative panoramic radiograph showing the complete excision of the tumor and the right coronoid process.

Different from OCs of the long bone, the majority of which are asymptomatic and do not require any treatment (12), the functional and cosmetic problems resulting from OCs of the craniofacial bone necessitate their resection. The definitive treatment of coronoid OC is coronoidectomy. No reconstruction of the face is needed, which contrasts with the requirements for

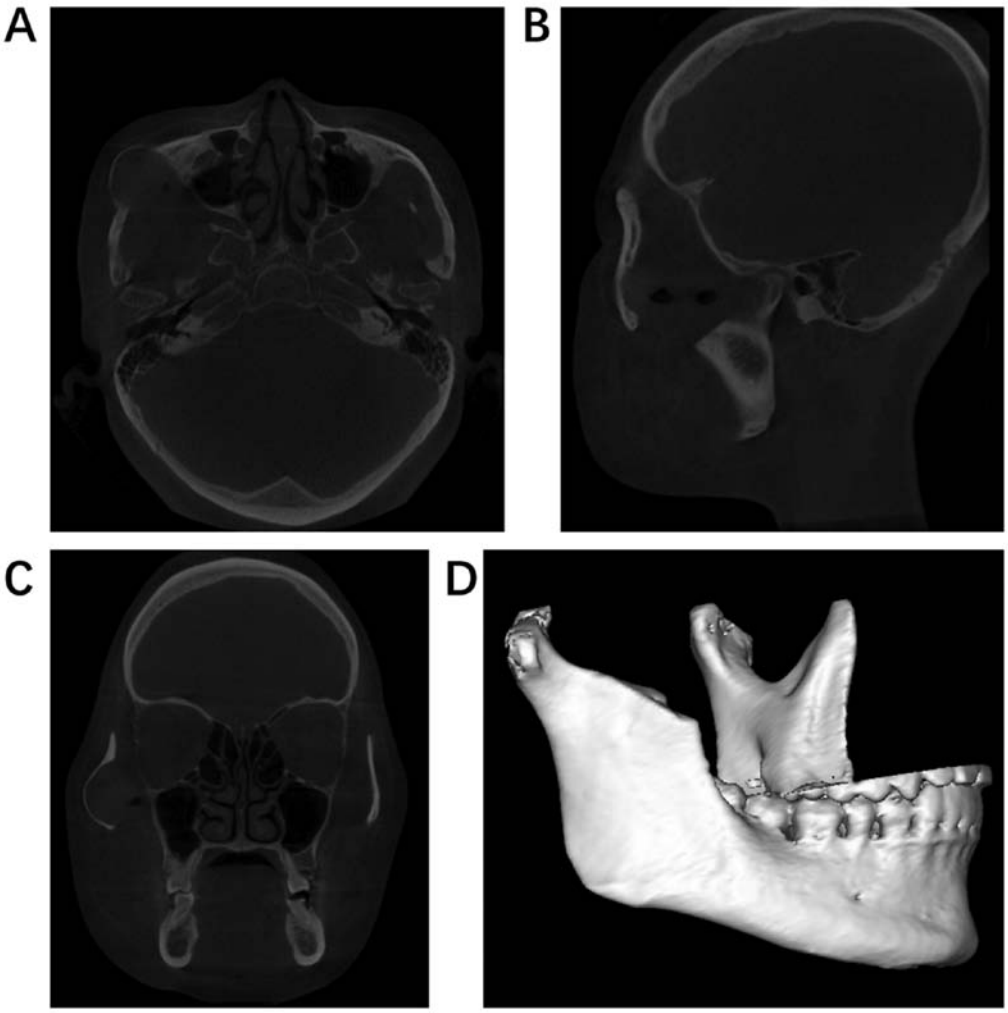

Figure 4. Postoperative CBCT and 3-dimensional reconstruction. Post-operative CBCT showed that the tumor was totally excised. (A) Axial plane, (B) sagittal plane and (C) coronal plane. (D) 3-Dimensional reconstruction of the tumor and the mandible. CBCT, cone-beam computed tomography.

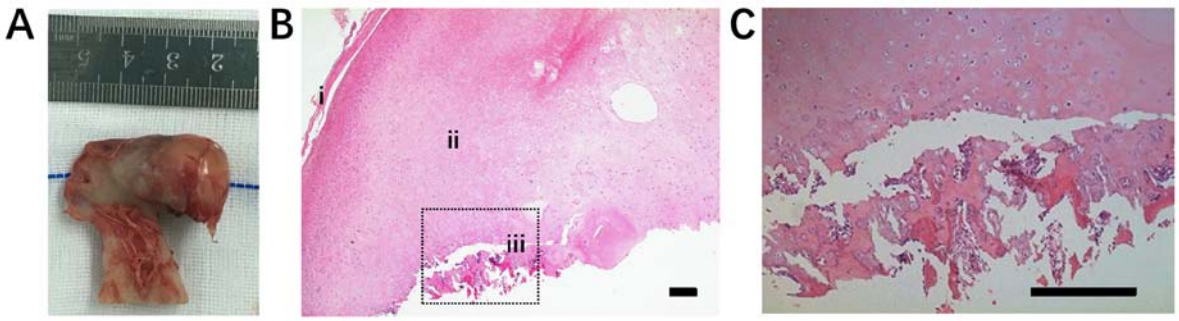

Figure 5. Excised specimen and histological hematoxylin and eosin photomicrographs. (A) The tumor measured ~4.0x1.5x1.5 cm. (B) Histologically, the mass was divided into three layers from the surface to the inside: i) Fibrous tissue; ii) cartilaginous tissue; and iii) cancellous bone. Magnification, x5. (C) Higher magnification of the region marked 'iii' in (B). Magnification, x20. Scale bar, 250 μm.

condylar OC. Surgical approaches primarily include intraoral and extraoral approaches, or a combination of both techniques. The intraoral approach is more favorable, as it allows direct access to the coronoid process while eliminating the potential of injuring the facial nerve and scarring (27). However, problems may occur when facing patients with severe trismus, which could prevent or hinder surgical access. Additionally, if the mass is large and in close proximity to the zygomatic arch, an extraoral approach allows better access and visualization (5). In the present case, although the tumor was extremely large and the patient presented with a serious limitation of mouth opening, considering the patient's young age and that the coronoid process was not firmly trapped in the zygomatic arch, an intraoral approach was successfully performed.

Recurrence and malignant transformations of OC are extremely rare (5,12). For OCs in the craniofacial region, only

6 recurrences (12,58-62) and 2 malignant transformations (63) were reported. All cases with recurrence of malignant change were associated with OCs in the extracoronoid region and were initially treated in a conservative way, namely local resection of the tumor. The excellent prognostic outcome of treating patients with coronoid OCs may be due to the relatively radical surgical procedure in which the tumor, as well as the coronoid process, are removed. These findings suggest that a complete resection of the tumor should be ensured to prevent recurrence or malignant change.

In conclusion, a diagnosis of coronoid OC should be taken into consideration when facing patients with a limited ability to open their mouth, especially in patients with no other symptoms. CT or CBCT scans may serve an important role in an accurate diagnosis. Timely treatment can prevent possible complications such as facial swelling and asymmetry. Coronoidectomy is the ideal treatment. The prognosis of the disease is excellent, as no recurrence or malignant changes were reported.

Acknowledgements

Not applicable.

Funding

The present study was funded by The Science and Technology Planning Project of Guangzhou, China (grant no. 2015100110268).

Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Authors' contributions

QT and XL conceived and designed the study. XL and PSL collected the data. XL and PSL wrote the manuscript. TL critically revised the article, reanalyzed the data, solved problems with the 3D reconstruction and edited the figures.

Ethics approval and consent to participate

Not applicable.

Patient consent for publication

Written informed consent for publication was provided by the patient.

Competing interests

The authors declare that they have no competing interests.

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