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

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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.

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) 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 meta-plastic 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
The present review was performed using a computer-assisted literature review. Follow-up will be performed over an extended period. After a follow-up period of 21 months, there was no evidence of recurrence or malignant transformation reported. 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. 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 potential to differentiate into cartilage.
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               |
| Mohanty et al, 2016                        | 18         | M   | Right    | LMO, A   | Extraoral         | 36                | None               |
| Dandriyal et al, 2015                      | 20         | M   | Left     | LMO, P   | Intraoral         | 54                | None               |
| Sawada et al, 2015                         | 14         | M   | Left     | LMO      | Intraoral         | 6                 | None               |
| Losa-Munoz et al, 2014                     | 42         | M   | Right    | LMO, A   | Intraoral         | NA                | NA                 |
| Fan et al, 2014                            | 20         | M   | Left     | LMO, A   | Combined          | 20                | None               |
| Mohanty et al, 2016                        | 18         | M   | Right    | LMO, A   | Intraoral         | 36                | None               |
| Dandriyal et al, 2015                      | 20         | M   | Left     | LMO, P   | Intraoral         | 54                | None               |
| Sawada et al, 2015                         | 14         | M   | Left     | LMO      | Intraoral         | 6                 | None               |
| Losa-Munoz et al, 2014                     | 42         | M   | Right    | LMO, A   | Intraoral         | NA                | NA                 |
| Fan et al, 2014                            | 20         | M   | Left     | LMO, A   | Combined          | 20                | None               |
| Stringer et al, 2013                       | 27         | M   | Left     | LMO, A   | Intraoral         | NA                | None               |
| Aoki et al, 2013                           | 18         | M   | Right    | A, P     | Intraoral         | 15                | None               |
| Ruiz and Lara, 2012                        | 28         | M   | Left     | LMO      | Combined          | 36                | None               |
| Dandriyal et al, 2015                      | 20         | M   | Left     | LMO, A   | Intraoral         | 20                | None               |
| Coll-Anglada et al, 2011                   | 52         | F   | Right    | LMO, A   | Intraoral         | 6                 | None               |
| D'Ambrosio et al, 2011                     | 39         | M   | Left     | LMO      | Intraoral         | Several years     | None               |
| Acosta-Feria et al, 2011                   | 55         | M   | Right    | LMO, A   | Extraoral         | 20                | None               |
| Sreeramaneni et al, 2011                   | 45         | F   | Left     | LMO, A   | Combined          | 3                 | None               |
| Yesildag et al, 2010                       | 16         | M   | Right    | LMO, A   | Extraoral         | 14                | None               |
| Zhong et al, 2009                          | 39         | F   | Bilateral| LMO, A   | Intraoral         | 9                 | None               |
| Etoz et al, 2009                           | 43         | F   | Right    | LMO, A   | Intraoral         | 6                 | None               |
| Thota et al, 2009                          | 15         | M   | Bilateral| LMO, A   | Intraoral         | 14                | None               |
| Akan and Mehrelyeva, 2006                  | 24         | M   | Bilateral| LMO      | Intraoral         | NA                | NA                 |
| Villanueva et al, 2006                     | 44         | F   | Left     | LMO, A   | Intraoral         | 10                | None               |
| Capote et al, 2005                         | 23         | F   | NA       | LMO, A, P| Intraoral         | 12                | None               |
| Emekli et al, 2002                         | 21         | M   | Right    | LMO, A, P| Extraoral         | NA                | NA                 |
| Escuder et al, 2001                        | 24         | M   | Left     | LMO, A   | Intraoral         | NA                | NA                 |
| Roychoudhury et al, 2002                   | 32         | M   | Left     | LMO, A   | Extraoral         | 12                | None               |
| Hernandez-Alfar et al, 2000                 | 22         | M   | Left     | LMO, A   | Extraoral         | NA                | NA                 |
| Chichareon et al, 1999                     | 5          | M   | Right    | LMO, A   | NA                | NA                | NA                 |
| Manganaro, 1998                           | 26         | F   | Left     | LMO, A   | Intraoral         | 6                 | None               |
| Chen et al, 1998                           | 57         | F   | Left     | LMO, A   | Extraoral         | Several weeks     | None               |
| Gross et al, 1997                          | 22         | M   | Left     | LMO, A   | NA                | NA                | NA                 |
| Constantinides et al 1997                  | 31         | M   | Right    | LMO, A   | Extraoral         | 12                | None               |
| Kerscher et al, 1993                       | 45         | M   | Left     | LMO      | Intraoral         | NA                | NA                 |
| Kierscher et al, 1993                      | 45         | M   | Left     | LMO      | Intraoral         | NA                | NA                 |
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.

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.
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).

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 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.

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

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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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