A huge osteolipoma involving the coronoid process: a case report

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Abstract: A 28-year-old man visited our hospital with the chief complaint of trismus. Computed tomography revealed a well-defined, soft tissue tumor, 66 × 45 × 21 mm, with a distinct boundary in the inner region of the zygomatic arch. The mass contained various sizes of bone-like hard tissue, some of which adhered to the right coronoid process. A contrast-enhanced magnetic resonance image showed that the mass was composed mainly of adipose tissue. Tumorectomy was performed, and the histopathological diagnosis was osteolipoma. At 2-year follow-up, mouth opening had increased from 31 mm to 50 mm.

Keywords: osteolipoma; coronoid process; tumorectomy.

Introduction

Lipoma is a benign, mesenchymal neoplasm of the soft tissue that can occur in any part of the body. It is composed of mature adipose tissue encapsulated in thin connective tissue and is divided into lobules by a fibrous septum. The first case of lipoma with ossification was reported by Plaut et al. in 1959 (1). Histopathologically, they are classified on the basis of the formation of interstitial and tumor cells into various types, including the normal type, fibrolipoma, angiolipoma, myxolipoma, spindle-shaped lipoma, and others. On rare occasions, lipomas show osteogenesis, and they are then known as osteolipomas. Allen et al. reported that only 6 of 635 cases with benign lipomas in their study showed ossification (2). Clinically, an osteolipoma, containing mature lamellar bones irregularly arranged in a significant fat component, is quite rare (3,4). In contrast, an ossifying lipoma usually occurs within the intrathecal bone and rarely within the cortical bone. Although “ossifying lipoma” and “osteolipoma” have been confused in the literature, the present tumor was more appropriately defined as an “osteolipoma” (5). A very rare case of a huge osteolipoma involving the coronoid process is reported.

Case Report

In December 2012, a 28-year-old man visited our hospital with the chief complaint of trismus since his junior high school days.

Findings outside the oral cavity

Maximum mouth opening was 31 mm, and a bone-like hard mass was palpated around the lower edge of the right zygomatic bone.

CT findings

Computed tomography revealed a well-defined soft tissue tumor, 66 × 45 × 21 mm, with a distinct boundary in the inner region of the zygomatic arch (Fig. 1A). Because of the enlargement of the tumor, the temporal muscle and zygomatic arch were thinner, and the posterior wall of the maxillary sinus was pressed and absorbed (Fig. 1B). The mass contained various sizes of unstructured bone-like
hard tissue, some of which adhered to the right coronoid process, which was slightly hyperplastic compared to the left side (Fig. 1C, 1D).

**MRI findings**
A contrast-enhanced magnetic resonance image showed that the mass was mainly composed of adipose tissue, and other than these tissues, no organization was present in the mass. In addition, this image showed that the surrounding soft tissues including the temporal muscle were shifted by the tumor (Fig. 2).

**Treatment and clinical course**
The mass was diagnosed to be a benign tumor involving the coronoid process, and tumorectomy under general anesthesia was performed in June 2013. Considering the anatomical positional relationship between the tumor and the surrounding tissue, the surgery was performed with both extra-oral and intra-oral approaches. An extra-oral incision line was made following the Al-Kayat method, and the tumor under the deep temporal fascia was accessed following exposure of the zygomatic arch. The temporal muscle was only slightly present near the zygomatic arch, and the lower layer of the temporal fascia was filled with adipose tissue with rich blood vessels. Removal of adipose tissue on the outside of the tumor and peeling of its coating led to exposure of the surface of the tumor. In the oral cavity, the anterior edge of the mandibular ramus was seen by incising the buccal mucosa. The tumor partly adhered to the temporal muscle and the coronoid process. It was therefore resected by cutting the coronoid process at the level of the mandibular notch and removed by pushing from the oral cavity side toward the skin side (Fig. 3).
The extracted tumor of $63 \times 44 \times 28$ mm, was covered with a thin fibrous capsule, the temporal muscle was attached to the lower surface, and the cut surface of the coronoid process was observed (Fig. 4A, 4B). On histopathological examination, diffuse proliferation of mature adipose cells was observed, and mature bone tissue was widely distributed within the tumor. In part of these tissues, a lining of osteoblasts was observed, with no lipoblasts (Fig. 5). The histopathological diagnosis was osteolipoma. At 2-year follow-up, mouth opening had increased to 50 mm, and no postoperative neuropathy or recurrence was observed.
Discussion

With respect to the developmental mechanism of osteolipoma, several theories have been proposed. Castillo et al. presumed that an osteolipoma is caused by differentiation of multipotent, mesenchymal cells within the adipose tissue (metaplasia in pre-existing lipoma) (6). This phenomenon may be caused by systemic and local metaplasia (trauma or long continued ischemia). Blanshard and Veitch proposed that the transformation of osteoblasts to fibroblasts is led by bone-inducing factors released by blood-borne monocytes that enter into adipose tissue (7). Beranek proposed an alternative pathogenesis and suggested that, in some complex vascular tumors such as a lipoma with vascular origin, the proliferation of two or three clear cell populations occurs simultaneously, originating from undifferentiated endothelial cells (8). Chromosomal abnormalities such as translocations in 11q13 have also been reported in osteolipomas (6). Under normal conditions, the existence of adipose tissue in some regions can explain the occurrence of a lipoma and subsequent osteolipoma (9). Ohno et al. stated that fibrous tissue is the origin for bone changes in a lipoma (3). It is still controversial whether this disorder is a hamartoma, a neoplasm, or metaplastic. In the present case, it can be presumed that rich nutrition brought by hyperplasia of the blood vessels promoted the augmentation of this osteolipoma. As a result, the coronoid process was caught by the tumor and became slightly longer, probably induced by similar mechanisms of bone formation within the tumor.

Surgical excision is usually chosen as treatment for osteolipoma (6,10). For surgery in this case, extraoral and intra-oral approaches were used. There was a possibility that the coronoid process could be removed by passing through the extra-oral route, cutting the zygomatic arch, and inverting the masseter muscle. However, in this case, it appeared that the zygomatic arch could not be reconstructed with screws and plates, because of the significant thinness of this bone. In addition, there was a risk of damage to the maxillary artery with this method. Since the tumor had strong adhesions only with the coronoid process, by pushing upward from the oral cavity after cutting the coronoid process, the tumor could be extracted en bloc with minimum invasiveness without cutting the zygomatic arch. Although recurrences have not yet been reported (6,10), detailed monitoring and long-term follow-up are recommended because of the lack of clinical information. Thus, we have continued careful observation of this case.

References