

Ossifying fibroma in the mandibular angle mimicking metastatic clear cell renal cell carcinoma

A case report

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Abstract

Rationale: Ossifying fibroma is benign fibro-osseous neoplasm. The authors report a case of ossifying fibroma in the mandibular angle suspected as metastasis of clear cell renal cell carcinoma.

Patient concerns: A 74-year-old man presented to the primary hospital complaining of frequent urination. A tumor in the left kidney was detected via an abdominal computed tomography scan. The patient then visited the Department of Urology at our hospital.

Diagnoses: According to whole-body imaging examinations, the patient was suspected of having renal cancer with mandibular metastasis. Also, a cystic lesion of the maxilla was revealed.

Interventions: Left nephrectomy was performed by urologists, and the patient was diagnosed with clear cell renal cell carcinoma of the left kidney. Approximately 1 month later, resection with a safety margin of the mandibular lesion and removal of the maxillary lesion were performed by oral and maxillofacial surgeons.

Outcomes: The patient was diagnosed with ossifying fibroma of the mandible and an odontogenic keratocyst of the maxilla via a histopathological examination. Eighteen months have passed since the operation without clinical and imaging findings associated with recurrence.

Lessons: Ossifying fibroma in the mandibular angle of elderly patients is extremely rare. Surgeons should consider the possibility of metastasis when osteolytic lesions of the jaw are found in patients with cancer.

Abbreviations: CT = computed tomography, FDG PET/CT = 18F-fluorodeoxyglucose positron emission tomography/computed tomography, MRI = magnetic resonance imaging, OF = ossifying fibroma, RCC = renal cell carcinoma, Tc-99m MDP WBBS = whole-body bone scintigraphy using technetium-99m methylene diphosphonate.

Keywords: clear cell renal cell carcinoma, imaging examination, mandible, metastasis, ossifying fibroma

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Ethical approval: Not applicable. This is a case report.

Written informed consent was obtained from the patient for publication of the case and any accompanying images.

The authors have no conflicts of interest to disclose.

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1. Introduction

Ossifying fibroma (OF) is an uncommon benign fibro-osseous neoplasm affecting the jaws and the craniofacial skeleton.^[1–3] It is included in fibro-osseous and osteochondromatous lesions in the World Health Organization's classification of head and neck tumors.^[1] OF is mainly composed of fibrous stroma and bone elements with various degrees of maturation.^[4] It commonly occurs in the mandibular premolar-molar region and occurs in the second to fourth decades of life.^[2–7] Additionally, it has a female predilection with a male to female ratio of 1:5.^[2,3,6,7] It is usually initially asymptomatic, and pain and paresthesia are rare.^[2] Most cases are small and incidentally detected by routine dental radiographs, and there are few cases of multiple occurrences associated with familial inclination.^[2] However, it can cause facial deformity, displacement of the teeth, pathological fracture, and extend into the intracranial and intraorbital regions due to progressive and destructive growth.^[2–5] Previous studies suggested that OF arises from the periodontal ligament.^[2,4,7,8] Radiographically, OF is usually a well-defined unilocular lesion with or without a sclerotic margin overlapping the roots with or without root resorption, and the internal structure is often a mixture of radiolucent and radiopaque



Figure 1. An abdominal computed tomography scan detected a tumor in the left kidney (▲).

densities.^[2-4,6] However, these radiographic findings are inconclusive.^[6] The differential diagnosis of such lesions might include benign maxillofacial bone and cartilage tumors, benign epithelial odontogenic tumors, odontogenic and non-odontogenic developmental cysts, benign mesenchymal odontogenic tumors, chronic sclerosing osteomyelitis, primary and metastatic malignant tumors, and so on.^[3,6,7] In jaw lesions without significant expansion or destruction of the cortical bone and without displacement of the inferior mandibular canal, it is especially difficult to differentiate between benign and malignant lesions via radiological examinations.^[6] In such cases, the lesions are often not easily accessible for biopsies due to soft tissues and thick cortical bone. Therefore, image assessments such as magnetic resonance imaging (MRI) and nuclear medical examination may play an important role in the preoperative differential diagnosis.

The authors report a case of ossifying fibroma in the mandibular angle suspected as metastasis of clear cell renal cell carcinoma in an elderly man.

2. Consent

Written informed consent was obtained from the patient for publication of the case and any accompanying images.

3. Case report

A 74-year-old man presented to the primary hospital complaining of frequent urination. A tumor in the left kidney was revealed via an abdominal computed tomography (CT) scan (Fig. 1). The patient then visited the Department of Urology at our hospital. Whole-body bone scintigraphy using technetium-99m methylene diphosphonate (Tc-99m MDP WBBS) demonstrated an abnormally increased uptake in the left mandibular angle (Fig. 2). The patient was referred to the Department of Dentistry and Oral

Surgery at our hospital for further evaluation. A panoramic radiograph and CT scan of the maxillofacial region revealed an osteolytic lesion accompanied by a slight expansion of the cortical bone in the mandibular angle and a cystic lesion accompanied by expansion of the cortical bone in the maxillary anterior region (Fig. 3). He had no subjective symptoms in the maxillofacial region, and a physical examination revealed no symptoms. An intraoral examination demonstrated swelling of the maxillary anterior region. MRI of the mandibular lesion showed low signal intensity on a T1 weighted image and high signal intensity on a T2 weighted image in the central area (Fig. 4). The maxillary lesion showed high signal intensity on the

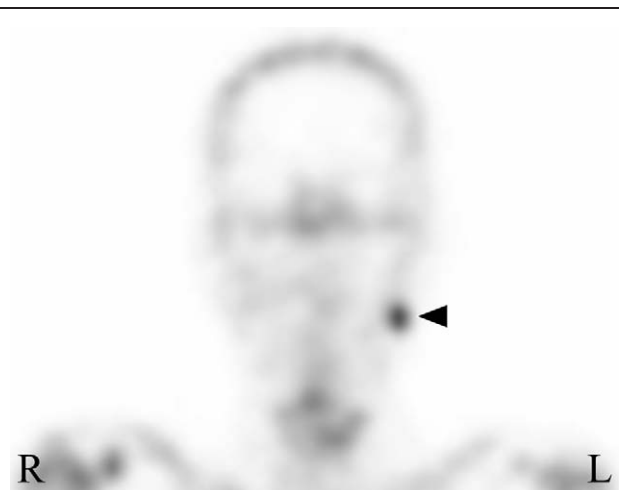


Figure 2. Whole-body bone scintigraphy using technetium-99m methylene diphosphonate showed abnormally increased uptake in the left mandibular angle (▲).

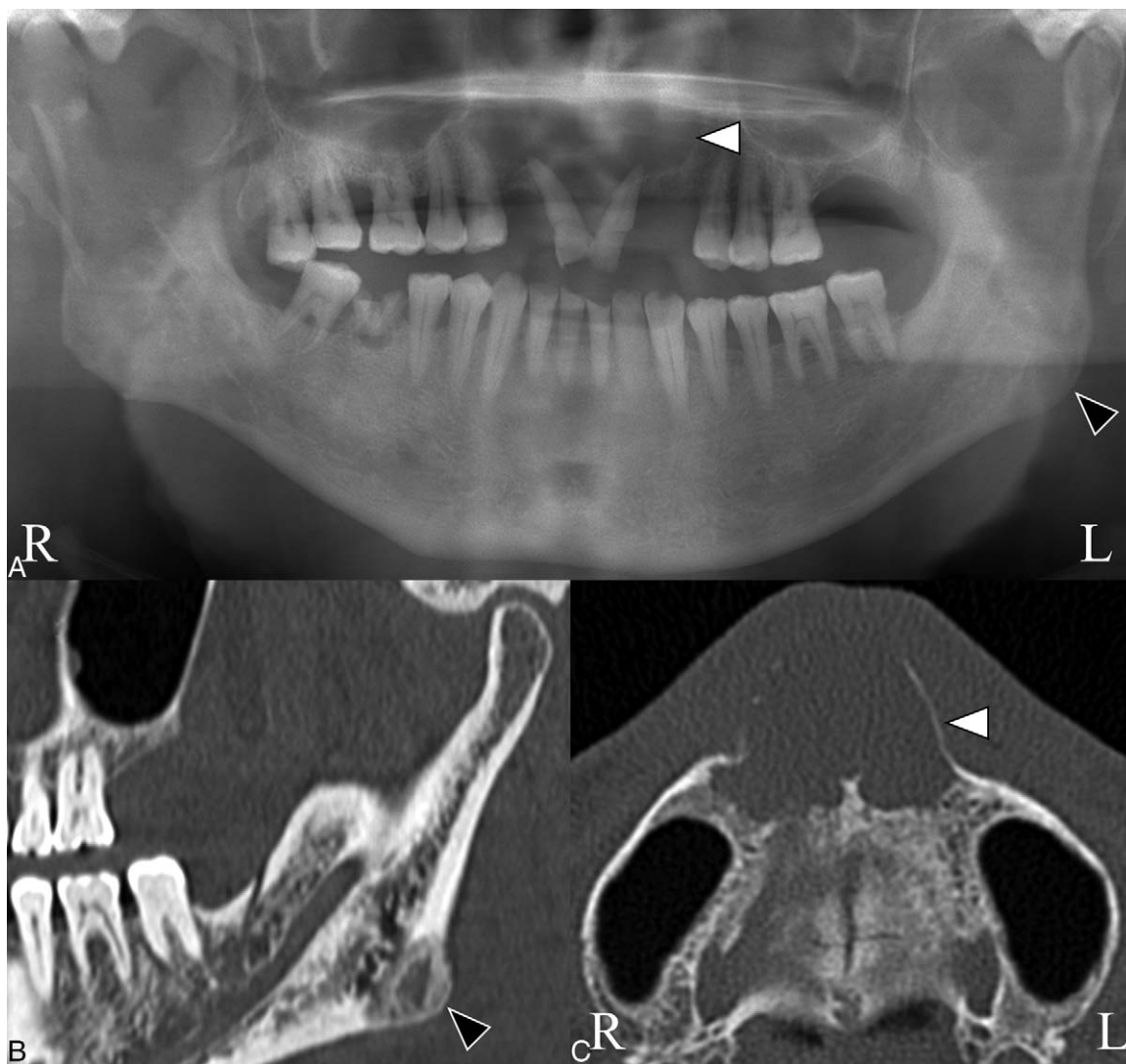


Figure 3. A preoperative panoramic radiograph (A) and computed tomography scan of the maxillofacial region (B and C) revealed an osteolytic lesion in the mandibular angle (▲) and a cystic lesion in the maxillary anterior region (△).

T2 weighted image. Whole-body 18F-fluorodeoxyglucose positron emission tomography/computed tomography (FDG-PET/CT) showed abnormally increased FDG uptake in the left kidney, but there was no abnormal uptake in the maxillofacial region. The results of the imaging examinations suggested a diagnosis of left kidney cancer (cT2aN0M1, Stage IV). They also revealed an osteolytic lesion of the mandible and a cystic lesion of the maxilla. The possibility of kidney cancer with mandibular metastasis could not be denied by the results. Left nephrectomy was performed by urologists under general anesthesia, and the patient was diagnosed with clear cell renal cell carcinoma (RCC) of the left kidney.

Approximately 1 month later, resection of the mandibular lesion with a safety margin and removal of the maxillary lesion were performed under general anesthesia by oral and maxillofacial surgeons. The findings of an intraoperative rapid-frozen pathological assessment of the mandibular lesion suggested that it was not a metastatic lesion from RCC. Concomitantly, mandibular reconstruction using a titanium plate and screws was performed to prevent mandibular fracture. The patient was

diagnosed with ossifying fibroma of the mandible and an odontogenic keratocyst (OKC) of the maxilla via a postoperative histopathological examination (Figs. 5 and 6). His postoperative course has been uneventful (Fig. 7).

There was no evidence of recurrence of RCC and jaw lesions 18 months after the second surgery.

4. Discussion

OF is a benign neoplasm, and the surgical approach remains controversial. Treatment methods for OF have been reported including curettage, enucleation, and radical resection.^[2,4] Previous studies reported the rate of recurrence after curettage was 28%, and that after partial or incomplete resection ranging from 30% to 56%.^[5,8] The residual outer lamella of the OF is considered as a factor for recurrence.^[5] Therefore, complete removal of the lesion is widely supported considering the possibility of recurrence although the size and location of the lesion often affects the choice of surgical approach.^[2-5] However, previous studies suggested that for asymptomatic OF, a wait-and-scan strategy can be applied in selected cases considering the

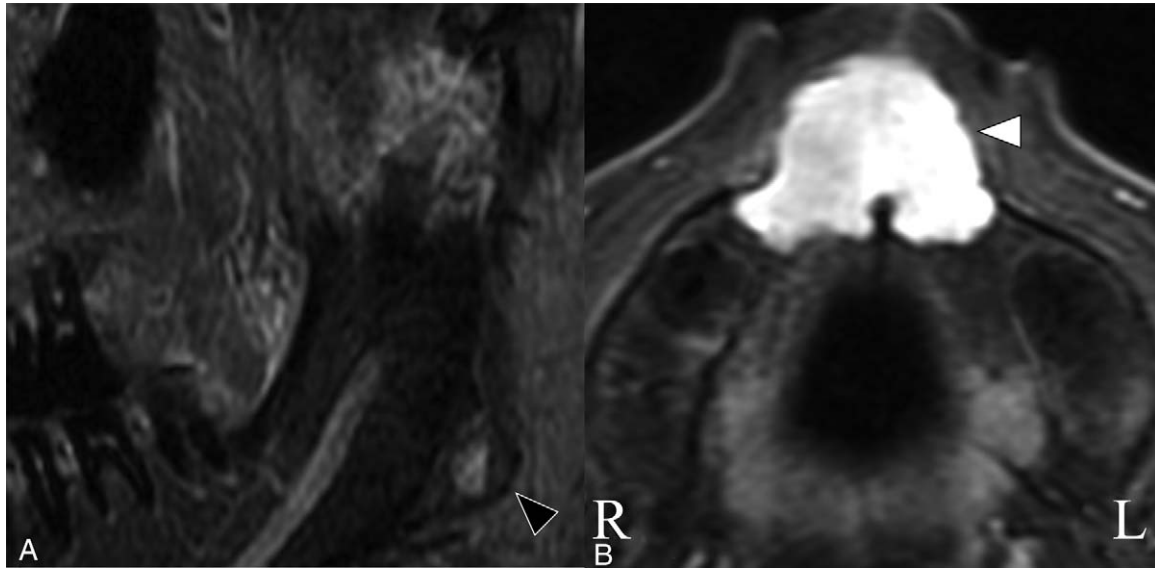


Figure 4. On T2 weighted magnetic resonance imaging, the central area of the mandibular lesion (A, \blacktriangle) and the maxillary lesion (B, \blacktriangle) showed high signal intensity.

anatomical location.^[5] It is difficult to differentiate between benign and malignant lesions of the jaw using only imaging findings, and biopsy usually plays an important role in differential diagnosis. In the present case, OF occurred in the mandibular angle away from the root-apex area in the elderly patient, and there was a problem of a differential diagnosis between the benign and metastatic malignant lesions because the patient was diagnosed with clear cell RCC. Therefore, the patient was treated via radical resection with a safety margin combined with an intraoperative rapid-frozen pathological assessment because there is the possibility of shelling out the tumor during surgical procedures, and mandibular reconstruction using a titanium plate and screws was performed to prevent mandibular fracture. Recurrence of OF did not occur during the follow-up period in this case.

Metastases to the oral and maxillofacial region from gastrointestinal or respiratory cancer are uncommon conditions and represent <1% of all malignancies in that region.^[9–11] The lung is the most common primary site metastasizing to that region, and metastatic RCC is extremely rare.^[10,11] There are reports showing that metastatic RCC can be the only presenting sign for RCC.^[12] The site will be most commonly angle of mandible (molar) as blood supply is abundant. Some studies reported metastatic clear cell RCC to the head and neck region affecting the parotid and submandibular glands, mandible, maxilla, paranasal sinuses, intraoral soft tissues, and so on.^[11] Although head and neck metastatic clear cell RCC is usually detected 1 to 7 years after the diagnosis of the primary tumor, some studies reported that metastases to the oral and maxillofacial region were detected synchronically or before the detection of

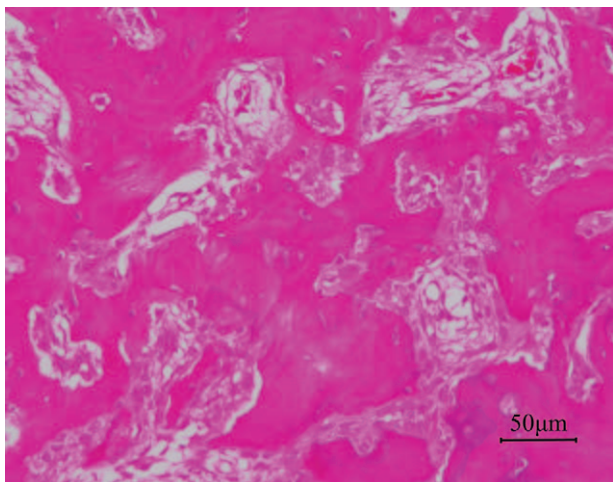


Figure 5. Fibrous connective tissue with variable amounts of calcified structures was observed on the hematoxylin and eosin stained mandibular specimen.

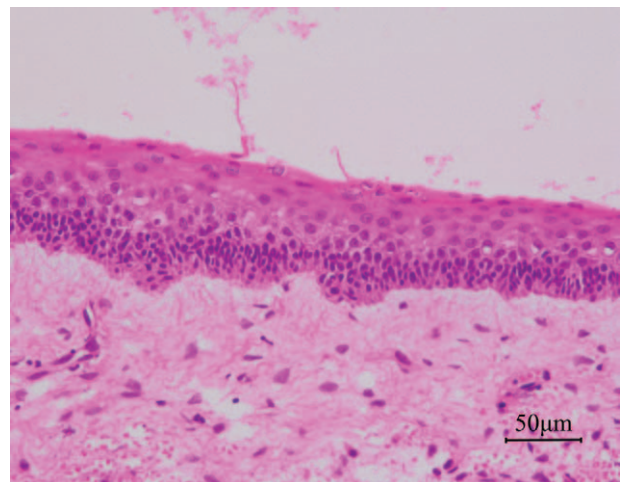


Figure 6. A cystic wall lined by a parakeratinized epithelium and fibrous connective tissue were observed on the hematoxylin and eosin stained maxillary specimen.



Figure 7. Postoperative panoramic radiograph.

the distant primary tumor.^[10,11] Therefore, clinicians should consider the possibility that the symptoms or results of imaging examinations in those regions may be the first clinical signs of an undiscovered distant primary tumor, and also the previous history of malignant disease is very important.^[10,11] The differential diagnosis of metastatic clear cell RCC to the head and neck region may vary depending on the location of the metastasis.^[11] If metastatic clear cell RCC to the jaws is suspected, clear cell odontogenic carcinoma and other odontogenic tumors containing clear cells should be considered using histopathological and immunohistochemical findings.^[11]

Although OF is best imaged by CT, additional imaging examinations might play an important role in the differential diagnosis of osteolytic lesions in patients with malignant diseases. OF usually has low to intermediate signal intensity on T1 weighted images and variable signal intensity on T2 weighted images on MRI.^[13] On T2 weighted images, low signal intensities may be predominant depending on the degree of calcification.^[13] Furthermore, low signal intensity is typically observed in the ossified peripheral areas, and high signal intensity is observed in the non-ossified central areas on T2 weighted images in cases of OF.^[13] Although the MRI findings of the mandibular lesion in this case were consistent with the previous report of OF, these could not deny the possibility of metastasis from clear cell RCC to the mandible because these were not specific findings. Therefore, this case suggested that MRI may play a limited role in the differential diagnosis of benign and metastatic lesions of the mandible in patients with malignant tumors.

Bone lesions related to RCC are typically observed in osteolytic lesions.^[14] Wu et al^[15] evaluated the diagnostic utility of FDG-PET and Tc-99m MDP WBBS for detecting bone metastases in 18 patients with RCC. They reported that the diagnostic sensitivity and accuracy of FDG-PET were 100% and 100% and those of Tc-99m MDP WBBS were 77.5% and 59.6%, respectively.^[15] Wu et al^[15] evaluated the role of FDG-PET for the detection of distant metastases in 24 patients with clear cell RCC. They reported the sensitivity, specificity, and positive predictive values were 63.6%, 100%, and 100%, respectively.^[16] They also reported that the mean lesion size of distant metastases with false-negative FDG-PET images was 1.0 cm.^[16] Tc-99m MDP WBBS show an abnormally increased uptake in benign lesions such as OF, fibrous dysplasia, and enchondroma.^[17] Therefore, it is difficult to differentiate these benign osteolytic lesions from bone metastases using Tc-99m MDP WBBS.^[17] Although imaging examination for detecting bone metastases in patients with RCC remains controversial, FDG-PET/CT may be more useful for detecting bone metastases than Tc-99m MDP WBBS.^[14,15] In the

present case, Tc-99m MDP WBBS demonstrated abnormally increased uptake in the left mandibular angle, and FDG-PET/CT showed no abnormal FDG uptake in that region. As a result, the rare condition that OF occurred in the mandibular angle of an elderly patient with RCC presented a preoperative diagnostic challenge.

The maxillary lesion with cortical bone expansion showed high signal intensity on a T2 weighted image. Additionally, Tc-99m MDP WBBS and FDG-PET/CT did not show an abnormal uptake. These findings suggested that the lesion was a benign cystic lesion, and it was diagnosed as OKC via a histopathological examination. Recurrence of OKC did not occur during the follow-up period in this case.

In conclusion, OF in the mandibular angle of elderly patients is extremely rare. Surgeons should consider the possibility of metastasis when an osteolytic lesion of the jaw is revealed in patients with cancer.

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