RESEARCH ARTICLE

CT and MR imaging characteristics of histological subtypes of head and neck ossifying fibroma

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Objectives: The current study aimed to assess CT and MRI characteristics of histological subtypes of head and neck ossifying fibroma (OF).

Methods: 12 patients with histopathologically-proven head and neck OF were included in this study. Lesions were pathologically classified into three histological subtypes: eight cement-OFs (COFs), three juvenile psammomatoid OFs (JPOFs), and one juvenile trabecular OF (JTOF). All patients underwent CT examination, while seven also underwent MRI. Imaging characteristics were retrospectively assessed.

Results: On CT images, the lesion margins were well-defined in nine patients (75%) (seven COFs and two JPOFs), partially ill-defined in two (17%) (one COF and one JTOF), and ill-defined in one (8%) (one JPOF). The continuity of the eroded overlying bone cortex was maintained in nine patients (75%) (seven COFs and two JPOFs) but disrupted in three (25%) (one COF, one JPOF, one JTOF). With respect to lesion density, homogeneous ground-glass opacity was observed in five patients (42%) (five COFs), target-like appearance in three (25%) (two COFs, one JPOF), and mixture of hyper- and hypodense areas were observed in four (33%) (one COF, two JPOFs, one JTOF). MR signal intensity was homogeneous in two patients (29%) (two COFs) and heterogeneous in five (71%) (two COFs, two JPOFs, one JTOF).

Conclusions: COFs tended to exhibit well-defined margins and preserved continuity of the overlying bone cortex. COFs were usually homogeneous, whereas JPOFs and JTOF were always heterogeneous. Target-like appearance was one of the characteristics of OFs, but it was observed in both COF and JPOF.

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Introduction

Ossifying fibromas (OFs) are benign fibro-osseous neoplasms that affect the jaws and the craniofacial skeleton. The World Health Organization (WHO) classification of head and neck tumors (2017) categorizes OFs into fibro-osseous and osteochondromatous lesions.¹

Based on the age of onset, clinical presentation, potential behavior, pattern of mineralization, and histopathological characteristics, three clinicopathological variants have been identified: cemento-OF (COF) and two distinct juvenile OFs (JOFs) [juvenile psammomatoid OF (JPOF) and juvenile trabecular OF (JTOF)].¹ COFs exhibit a definite female predilection (female-to-male ratio as high as 5:1),^{1,2} while JOFs exhibit no gender

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predilection.¹ COFs commonly occur in the third and fourth decades of life.^{1,2} JPOFs have been reported more commonly in adults (mean age: 16–33 years) compared with JTOFs, whereas JTOFs are usually seen in children and adolescents (mean age: 8.5–12 years).^{1,3}

COFs occur exclusively in the tooth bearing areas of the jaw, and the most common sites are the mandibular premolar and molar areas. JPOFs may occur in the jaw but predominantly affect the extragnathic craniofacial bones, particularly the periorbital frontal and ethmoid sinus.^{4,5} JTOFs usually occur in the jaw, with a predilection for maxilla; however, extragnathic occurrence is extremely rare.³

COFs are slow-growing benign neoplasms, and they do not tend to recur after complete excision.¹ On the contrary, JOFs, particularly in children, exhibit rapid growth and are associated with a high recurrence rate (30–56%).⁶ Because of the considerable difference between the recurrence rates of COP and JOF, it is clinically important to differentiate between the two entities for surgical planning and prognostic prediction. Although differentiation between the CT findings of JPOF and JTOF has already been reported,^{6–8} we did not find any studies that compared the CT and MRI characteristics of the three histological subtypes, particularly between COF and JOF. Therefore, the aim of this study was to assess the CT and MRI characteristics of histological subtypes of head and neck OF with an emphasis on the differentiation between COF and JOF.

Methods and materials

Patients

The present study was approved by the human research committee of the institutional review board of Gifu University Hospital, and complied with the guidelines of the Health Insurance Portability and Accountability Act of 1996. The requirement for informed consent was waived due to the retrospective nature of this study. Between December 2006 and June 2017, we searched our hospital's (Gifu University Hospital) electronic medical chart system and found 12 consecutive patients [5 males and 7 females; median age, 26 years (range, 6–65 years)] with histopathologically-proven head and neck OF. The histopathological diagnosis was based on surgical specimens (total resection) in five patients and on excisional biopsy specimens in seven. The tumor locations were mandibular bone in four patients, sphenoid bone in three, maxillary bone in two, inferior nasal turbinate in two, and temporal bone in one. All 12 patients underwent CT examination, while 7 also underwent MRI.

An experienced pathologist with 28 years of post-training experience reviewed the histological specimens macroscopically and microscopically, and classified the OFs into three histological subtypes according to the World Health Organization classification of head and neck tumors: COF, JPOF, and JTOF.¹ Eight OFs were

Table 1 Patient characteristics

Number of patients	12
Age (year)	
Median	26
Range	6–65
Gender	
Male	5
Female	7
Histological subtype	
COF	8
JPOF	3
JTOF	1
Location	
Mandibular bone	4
Sphenoid bone	3
Maxillary bone	2
Inferior nasal turbinate	2
Temporal bone	1
Examination	
CT	12
MRI	7

COF, cemento-ossifying fibroma; JPOF, juvenile psammomatoid ossifying fibroma; JTOF, juvenile trabecular ossifying fibroma.

classified as COF [two males and six females; median age, 34 years (range, 6–65 years)], three as JPOF [two males and one female; median age, 13 years (range, 10–63 years)], and one as JTOF (male; age, 22 years). Patient characteristics are summarized in [Table 1](#).

CT imaging

CT examination was performed in all patients. A 16-slice CT scanner (LightSpeed Ultra 16, GE Healthcare, Milwaukee, WI) was used for three patients, a 64-slice CT scanner (Brilliance CT 64, Philips Medical Systems, Best, Netherlands) for six, 64-slice CT scanner (Discovery CT 750HD, GE Healthcare, Milwaukee, WI) for one, and cone beam CT for the remaining two. Transverse CT images were reconstructed using a 2.5 mm section thickness and no overlap.

MRI

Seven patients (four patients with COF, two with JPOF, and one with JTOF) were examined using a 1.5 T MRI system (Intera Achieva 1.5 T Pulsar; Philips Medical Systems, Amsterdam, Netherlands, in five patients and Signa Excite Twin Speed, GE Healthcare, Milwaukee, WI, in the remaining two). All images were obtained at section thickness of 4 mm with a 1 mm intersection gap, and a 20 × 20 cm field of view. Axial non-fat-suppressed T_2 weighted fast spin echo [3000–4300/89–102 repetition time/echo time (TR/TE), 512 × 512 matrix] and axial non-fat-suppressed T_1 weighted spin echo [566–827/9–12 (TR/TE), 512 × 512 matrix] images were obtained in all seven patients. In five patients (three patients with COFs and two with JPOFs), axial

gadolinium-enhanced fat-suppressed T_1 weighted spin echo [584–666/9–13 (TR/TE), 512×512 matrix] images were obtained after intravenous injection of 0.1 mmol kg^{-1} of (Magnevist, Bayer HealthCare, Leverkusen, Germany) or Gadobutrol (Gadavist, Bayer HealthCare, Leverkusen, Germany).

Image assessment

Two radiologists with 18 and 4 years of post-training experience of head and neck imaging individually reviewed all MR images, and any disagreements were resolved by consensus.

CT images were reconstructed using bone algorithm and were evaluated with respect to tumor margin, presence of expansile growth, status of adjacent overlying bone cortex, and intratumoral density. The intratumoral density was visually classified into three groups: homogeneous ground-glass opacity; target-like appearance; and mixture of hyper- and hypodense areas. Ground-glass opacity was defined as a granular or pebbled density that resembles pulverized glass. Target-like appearance on CT images was defined as central dense ossification surrounded by hypodense area or ground-glass opacity.

The uniformity of the intratumoral signal intensity was assessed on T_2 and T_1 weighted images. T_2 weighted images were evaluated for the predominant signal intensity, presence of hypointense area similar to that of the muscles, hyperintense area similar to that of the cerebrospinal fluid, target-like appearance, and fluid-fluid level. The predominant signal intensity was compared with that of the gray matter. Target-like appearance on T_2 weighted images was composed of central hypointensity surrounded by iso- or hyperintense area. The uniformity and the degree of intratumoral enhancement were also assessed on gadolinium-enhanced T_1 weighted images.

Results

The maximum diameter of the 12 OFs measured from CT images ranged from 7 to 40 mm (mean, 22.6 mm). The maximum diameter for each subtype was as follows: eight COFs [mean, 20.3 mm (range, 7–33 mm)]; three JPOFs [mean, 23.0 mm (range, 11–33 mm)]; and one JTOF (40 mm). The maximum diameter of JOFs tended to be larger than that of COFs.

CT imaging findings are summarized in Table 2. On CT images, the tumor margin was well-defined in nine patients (75%) (seven COFs and two JPOFs), partially ill-defined in two (17%) (one COF and one JTOF), and ill-defined in one (8%) (one JPOF). Expansile growth was observed in 10 patients (83%) (6 COFs, 3 JPOFs, and 1 JTOF). The adjacent overlying bone cortex was observed to be eroded in all patients; the continuity of the eroded overlying bone cortex was maintained in nine patients (75%) (seven COFs and two JPOFs) but disrupted in three patients (25%) (one COF, one

Table 2 The summary of CT imaging findings

No.	Age	Sex	Diagnosis	Location	Size (mm)	Margin	Expansile growth	Adjacent overlying bone cortex	Intratumoral density
1	6	M	COF	Maxillary bone	26	Partially ill-defined	+	Eroded, continuous	Homogeneous GGO
2	16	M	COF	Inferior nasal turbinate	7	Well-defined	+	Eroded, disrupted	Homogeneous GGO
3	18	F	COF	Mandibular bone	15	Well-defined	+	Eroded, continuous	Homogeneous GGO
4	30	F	COF	Inferior nasal turbinate	33	Well-defined	+	Eroded, continuous	Homogeneous GGO
5	37	F	COF	Mandibular bone	8	Well-defined	+	Eroded, continuous	Heterogeneous, mixed
6	42	F	COF	Mandibular bone	23	Well-defined	+	Eroded, continuous	Heterogeneous, target
7	46	F	COF	Mandibular bone	13	Well-defined	+	Eroded, continuous	Homogeneous GGO
8	65	F	COF	Sphenoid bone	37	Well-defined	+	Eroded, continuous	Heterogeneous, target
9	10	M	JPOF	Sphenoid bone	33	Well-defined	+	Eroded, disrupted	Heterogeneous, mixed
10	13	M	JPOF	Temporal bone	25	Ill-defined	+	Eroded, continuous	Heterogeneous, target
11	63	F	JPOF	Sphenoid bone	11	Well-defined	+	Eroded, continuous	Heterogeneous, mixed
12	22	M	JTOF	Maxillary bone	40	Partially ill-defined	+	Eroded, disrupted	Heterogeneous, mixed

COF, cemento-ossifying fibroma; GGO, ground-glass opacity; JPOF, juvenile psammomatoid ossifying fibroma; JTOF, juvenile trabecular ossifying fibroma; Mixed, mixture of hyper- and hypodense areas; Target, target-like appearance.

Figure 1 An 18-year-old female with COF (Case 3). (a) CT image with bone algorithm shows a well-defined lesion (arrow) of the right mandibular bone. The adjacent overlying bone cortex is eroded, but the continuity of the eroded overlying bone cortex was maintained (arrow head). Intratumoral density is homogeneous ground-glass opacity. (b) T_2 weighted image shows a homogeneously hypointense lesion (arrow). COF, cemento-ossifying fibroma.

JPOF, one JTOF). With respect to lesion density, homogeneous ground-glass opacity was observed in five patients (42%) (five COFs) (Figure 1a), target-like appearance in three (25%) (two COFs, one JPOF) (Figures 2a and 3), and mixture of hyper- and hypodense areas were observed in four (33%) (one COF, two JPOFs, one JTOF) (Figures 4a and 5a).

MRI findings are summarized in Table 3. The intratumoral signal intensity on T_2 and T_1 weighted images was homogeneous in two patients (29%) (two COFs) and heterogeneous in five (71%) (two COFs, two JPOFs, and one JTOF). Among the three patients (three COFs) with homogeneous ground-glass opacity on CT images, the signal intensity on T_2 and T_1 weighted images was homogeneous in two patients (Figure 1b) and heterogeneous in one. The predominant signal intensity on T_2 weighted images was hypointense in six patients (86%) (three COFs,

Figure 2 A 65-year-old female with COF (Case 8). (a) CT image with bone algorithm shows a well-defined lesion (arrow) of the right sphenoid bone. The expansile growth into the clivus is observed (arrow head). The lesion is composed of central dense ossification surrounded by hypodense area. (b) T_2 weighted image shows a heterogeneous lesion (arrow) with central hypointensity surrounded by iso- or hyperintense area (target-like appearance). COF, cemento-ossifying fibroma.

Figure 3 A 13-year-old male with JPOF (Case 10). CT image with bone algorithm shows an ill-defined lesion (arrow) of the left temporal bone. The expansile growth into the external auditory canal is observed (arrow head). The lesion is composed of central dense ossification surrounded by ground-glass opacity. JPOF, juvenile psammomatoid ossifying fibroma.

two JPOF, and one JTOF) and isointense in one (14%) (one COF). On T_2 weighted images, hypointense area similar to that of the muscles was observed in five patients (71%) (two COFs, two JPOFs, and one JTOF) and hyperintense area similar to that of the cerebrospinal fluid was observed in three (43%) (one COF, one JPOF, and one JTOF) (Figures 4b and 5b). Target-like appearance on T_2 weighted images was observed in one patient (14%) (one COF) (Figure 2b), who also showed target-like appearance on CT images. Fluid-fluid level was not observed in any of the OF lesions.

Figure 4 A 10-year-old male with JPOF (Case 9). (a) CT image with bone algorithm shows a well-defined lesion (arrow) of the left sphenoid bone. The expansile growth and disrupted overlying bone cortex is observed (arrow head). Intratumoral density is heterogeneous mixture of hyper- and hypodense areas with ground-glass opacity. (b) T_2 weighted image shows a heterogeneous lesion (arrow) with predominant hypointense area similar to that of the muscles and focal hyperintense area (arrow head) similar to that of the cerebrospinal fluid. JPOF, juvenile psammomatoid ossifying fibroma.

Figure 5 A 22-year-old male with JTOF (Case 12). (a) CT image with bone algorithm shows a partially ill-defined lesion (arrow) of the right maxillary bone. The expansile growth and disrupted overlying bone cortex is observed (arrow head). Intratumoral density is heterogeneous mixture of hyper- and hypodense areas with ground-glass opacity. (b) T2 weighted image shows a heterogeneous lesion (arrow) with predominant iso- and hypointense area and focal hyperintense area (arrow head) similar to that of the cerebrospinal fluid. JTOF, juvenile trabecular ossifying fibroma.

On gadolinium-enhanced T_1 weighted images, heterogeneous slight enhancement was observed in all five patients.

Discussion

In comparison with COF, JOF exhibits a tendency for locally aggressive behavior, including invasion and destruction of surrounding tissues, bony erosion, and post-operative recurrence. Histopathologically, COF is encapsulated, whereas JPOF and JTOF are not encapsulated. On CT images, COF typically appears as a well-defined expansile mass with no destruction of contiguous anatomical structures and is completely covered by bone cortex.² JOFs are also expansile masses with well-defined borders and thin corticated margin,⁷ however, cortical disruption and involvement of adjacent anatomical structures may be seen.⁹ In the present study, almost all COFs [seven of eight (88%)] exhibited a well-defined margin, whereas a partially ill-defined or ill-defined margin was observed in half of JOFs [two of four (50%)]. In addition, the continuity of the eroded overlying bone cortex was maintained in almost all COFs [seven of eight (88%)], whereas it was disrupted in half of JOFs [two of four (50%)]. These differences of imaging characteristics are attributable to the locally aggressive nature of JOFs.

COFs are characterized by hypercellular fibroblastic stroma containing variable amounts of calcified structures. The calcified structures are composed of variable amount of osteoid or bone and lobulated basophilic masses of cementum-like tissue.¹ The ratio of osteoid or bone to cementum-like tissue varies. Ossification is rare, and woven bone is the predominant component.² In our study, COFs frequently showed homogeneous ground-glass appearance similar to that observed in fibrous dysplasia [five of eight (63%)]; however, large COFs

may appear as heterogeneous masses.^{2,10,11} The relatively small size of COFs in our study [mean maximum diameter, 20.3 mm (range, 7–33 mm)] may have affected the intratumoral homogeneity.

JPOF are characterized by concentric or laminated ossicles called psammomatoid bodies. Psammomatoid ossicles contain osteocytes, which indicates its osteogenic origin; their morphology may vary from small round or oval shape to large and irregular shape. Extensive psammomatous calcification may account for the ground-glass opacity.¹² JPOFs are primarily radiolucent and exhibit irregular and scattered calcification. The internal structure of JPOFs can be radiolucent, mixed, or radiopaque, depending on the degree of calcification and the extent of the cystic changes.⁵ Because large cystic areas can be seen on the cut surface of JPOFs, cystic areas sometimes appear on CT and MR images. These cystic areas may contain hemorrhage and represent secondary aneurysmal bone cysts. In our study, all three JPOFs were heterogeneous on CT images, and cystic areas on T_2 weighted images were observed in one of two (50%) JPOFs.

JTOFs are characterized by hypercellular stroma composed of spindle cells with little collagen production. Irregular mineralization occurs at the center of the strands, which results in the production of immature woven bone trabeculae that are devoid of osteoblastic rimming.¹ Progressive calcification of the osteoid results in anastomosing trabeculae of immature woven bone. Cystic degeneration and aneurysmal bone cyst formation have been described.⁶ Radiographically, JTOF are usually mixed radiolucent/radiopaque lesions, especially radiolucency with irregular scattered calcifications; however, these may present as completely radiolucent lesions with areas of faint internal radiopacity.^{6,7} Areas of calcification occasionally appear as fine specks and produce ground-glass appearance.⁶ A case of JTOF in our study showed mixture of hyper- and hypodense

Table 3 The summary of MRI findings

No.	Age	Sex	Diagnosis	Uniformity of intratumoral SI on T_1/T_2WI	Predominant SI on T_2WI	Hypointense area similar to the muscles on T_2WI	Hyperintense area similar to the CSF on T_2WI	Target-like appearance on T_2WI	Fluid–fluid level on T_2WI
1	6	M	COF	NA	NA	NA	NA	NA	NA
2	16	M	COF	Heterogeneous	Hypointensity	+	–	–	–
3	18	F	COF	Homogeneous	Hypointensity	–	–	–	–
4	30	F	COF	NA	NA	NA	NA	NA	NA
5	37	F	COF	NA	NA	NA	NA	NA	NA
6	42	F	COF	NA	NA	NA	NA	NA	NA
7	46	F	COF	Homogeneous	Isointensity	–	–	–	–
8	65	F	COF	Heterogeneous	Hypointensity	+	+	+	–
9	10	M	JPOF	Heterogeneous	Hypointensity	+	+	–	–
10	13	M	JPOF	NA	NA	NA	NA	NA	NA
11	63	F	JPOF	Heterogeneous	Hypointensity	+	–	–	–
12	22	M	JTOF	Heterogeneous	Hypointensity	+	+	–	–

COF, cemento-ossifying fibroma; CSF, cerebrospinal fluid; JPOF, juvenile psammomatoid ossifying fibroma; JTOF, juvenile trabecular ossifying fibroma; NA, not available; SI, signal intensity; T_1WI , T_1 weighted image; T_2WI , T_2 weighted image.

areas on CT images and heterogeneous intensity on T_2 and T_1 weighted images. In addition, hypointense area similar to that of the muscles and hyperintense area similar to that of the cerebrospinal fluid were both observed within the JTOF on T_2 weighted images.

Most aneurysmal bone cysts in the paranasal sinus might be secondary and frequently occur in combination with OFs. Indeed, fluid–fluid levels have been reported in OFs with secondary aneurysmal bone cysts.^{8,10,13–16} In a study, 15 out of 438 (3.42%) patients with OFs in the paranasal sinus showed fluid–fluid levels; the histological subtypes included COF in 10 patients (66.7%), JPOF in 4 (26.7%), and JTOF in 1 (6.7%).¹³ However, in our study, fluid–fluid level was not observed in any OF.

We defined target-like appearance as central dense ossification surrounded by hypodense area or ground-glass opacity on CT images or central hypointensity surrounded by iso- or hyperintense area on T_2 weighted images. However, to the best of our knowledge, we failed to find any reports that have described fibro-osseous lesions (including OFs) with target-like appearance. Therefore, target-like appearance (centrally located dense ossification) may be one of the characteristic imaging findings of OFs. In the present study, target-like appearance on CT images was observed in both COP [two of eight (25%)] and JPOF [one of three (33%)].

The present study has several limitations. First, the cohort was small because the study was conducted at a single institution. Second, MR images were available only for seven patients, because of the retrospective nature of this study. However, MRI usually plays a supplemental role in the diagnosis of fibro-osseous lesion.

COFs tended to exhibit well-defined margins and preserved continuity of the overlying bone cortex, whereas half of JOFs had partially ill-defined or ill-defined margins with disruption of the overlying bone cortex. COFs were usually homogeneous, whereas JOFs were always heterogeneous. Therefore, homogeneous lesions with well-defined margins are suggestive of COFs. Target-like appearance was one of the characteristics of OFs, but it was observed in COF and JPOF.

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