Calcifying cystic odontogenic tumour: CT imaging

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Objectives: Calcifying cystic odontogenic tumour (CCOT) is a rare disorder of the jaw. A comparison between conventional radiographs and CT images in CCOTs has not been reported. The purposes of this study were to analyse conventional radiographs and CT images of CCOTs, establish CT images of CCOTs and assess the utility of CT in the diagnosis of CCOTs.

Methods: Nine patients with a histopathologically confirmed CCOT who had both conventional radiographs and CT images were enrolled.

Results: CT was superior to conventional radiographs in detecting buccolingual expansion, odontomas and radio-opaque bodies.

Conclusion: The characteristic CT appearances of CCOT were that radio-opaque bodies were typically located in the periphery of the lesion and the shape of radio-opaque bodies was linear and/or spotted. CT was useful in diagnosing a CCOT.

Received 30 March 2010

Revised 29 June 2010

Accepted 15 July 2010

DOI: 10.1259/bjr/19841479

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The calcifying odontogenic cyst (COC) was first recognised as a distinct pathological entity by Gorlin et al [1] in 1962. COC is a rare disorder of the jaw [2], and the reported frequency of COCs varies from 0.37 to 2.1% of all odontogenic tumours [3]. In 2005, COC was classified as a tumour and designated as a “calcifying cystic odontogenic tumour” (CCOT) by the World Health Organization (WHO) [4].

Histopathologically, the cyst wall is lined by a thin ameloblastomatous epithelium with the formation of ghost cells. These ghost cells may calcify [4], and the frequency of calcification in some of the ghost cells varies from 19 to 77% [5].

Radiographically, a CCOT generally appears as a unilocular lesion with a well-defined margin and contains calcification [3]. Few studies have reported on the radiographic features of CCOTs in the English language literature [3, 6], although CT image findings of CCOTs have been described [2, 3, 7]. A comparison between conventional radiographs and CT image findings in CCOTs, however, has not been reported.

The purposes of this study were to analyse conventional radiographs and CT images of CCOTs, establish CT images of CCOTs and assess the utility of CT in the diagnosis of CCOTs.

Methods and materials

We evaluated conventional radiographs and CT image findings of CCOTs and sought to demonstrate characteristic CT image findings and the utility of CT images in the diagnosis of CCOTs in patients referred to our departments between 1997 and 2007.

Nine patients with a histopathologically confirmed CCOT who had both conventional radiographs and CT images were enrolled. The subjects consisted of seven males and two females, with ages ranging from 11 to 57 years (median, 14 years).

Imaging examinations were performed with conventional radiographs and CT. Conventional radiographs included panoramic radiograph and intra-oral radiograph. CT was performed with a single-slice CT scanner (CT9200; YMS, Tokyo, Japan or Hispeed Advantage SG; GE Healthcare, Waukesha, WI) or a multislice CT scanner (Light Speed QX/i; GE Healthcare) at 120–140 kV or 140–250 mA. CT scans were performed with a 256 × 256 or 512 × 512 matrix size and 1–5 ml of iohexol in two cases. Coronal and sagittal reformatted images were made after examination.

We evaluated the site, locus, border, margin of tumour, root resorption, tooth divergence, impacted teeth and radio-opaque bodies on conventional radiographs. On CT, in addition to the items evaluated on conventional radiographs, the size of the tumour, buccolingual bony expansion, contrast enhancement, CT value inside the tumour, CT value outside the

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tumour, and the location and shape of radio-opaque bodies were evaluated. "CT value" was measured by setting a region of interest (ROI) inside the tumour or radio-opaque bodies. In the radio-opaque body, we also set a ROI. The tumour ROI was a circle and it was about a quarter of the tumour in size. The radio-opaque body ROI was a cross shape. The CT value was measured three times and the average was recorded as the CT value inside the tumour and the radio-opaque body.

Results

Results of conventional radiographs are shown in Table 1. By location, six cases involved the maxilla and three cases involved the mandible.

In all cases, the loculus was unilocular, borders were well defined and margins were regular (Figures 1a, 2a and 3a). Root resorption was seen in seven of nine cases and teeth divergence in eight of nine (Figures 1a,b, 2b and 3a). Impacted teeth were found in six of nine cases; in three cases, they were outside the lesion, in two cases they were inside the lesion and in one case, they were both outside and inside the lesion. An odontoma was found in one case.

The results of CT scanning are shown in Table 2. Regarding the loculus, border, margin, root resorption and teeth divergence, no difference was observed between the conventional radiographs and CT images (Figures 1c, d, 2c-e and 3b,c). Odontomas were detected in four cases on CT images; a single case was determined by conventional radiographs. In Cases 5 (Figure 3c), 6 and 7, odontomas were found only by CT. On CT images,

radio-opaque bodies were detected in eight cases, but six were observed by conventional radiographs. In Cases 2 and 8, radio-opaque bodies were found only by CT.

Table 3 shows the results of CT imaging findings. Buccolingual bony expansion was found in seven of nine cases. Contrast enhancement was not found in two cases (Figure 2c,d). CT values inside the tumour ranged from 18.6 to 64.2 HU (mean, 37.9 ± 12.8 HU). CT values of radio-opaque bodies ranged from 170.3 to 371.0 HU (mean, 245.5 ± 69.4 HU). Radio-opaque bodies were located at the periphery of the tumour in eight of nine cases. The shape of the radio-opaque body was linear in five cases (Figures 1c,d, and 2e) and linear and spotted in three cases (Figure 3b,c). Histopathological images of CCOTs are shown in Figure 4.

Discussion

A variable number of radio-opaque bodies is seen in about 50% of CCOT cases [4]. Root resorption and root divergence are common, and an associated unerrupted tooth is seen in about one-third of cases [4].

Tanimoto et al [6] reported that root resorption was one of the most important radiographic findings and that others were impacted teeth, odontomas and/or some other accompanying calcification when evaluating radiographs of 142 cases with COCs.

A COC generally appears as a unilocular lesion with a well-defined margin [3, 7]. In our series, by conventional radiographs and CT images, all cases had well-defined unilocular forms with regular margins. Teeth divergence and root resorption were common. Impacted teeth were found in six of nine cases. In our series, conventional

Table 1. Results of conventional radiography

Case	Site	Loculus	Border	Margin	Root absorption	Teeth divergence	Impacted tooth	Odontoma	Radio-opaque bodies
1	Right MD canine to moral	U	W	R	(+)	(+)	(-)	(-)	(+)
2	Right MD lateral incisor to canine	U	W	R	(+)	(+)	(-)	(-)	(-)
3	Right MX molar	U	W	R	(+)	(-)	OUT	(+)	(-)
4	Right MX lateral incisor	U	W	R	(-)	(+)	(-)	(-)	(+)
5	Right MX canine	U	W	R	(+)	(+)	OUT	(-)	(+)
6	Left MX canine	U	W	R	(+)	(+)	OUT	(-)	(+)
7 ^a	Right MX canine	U	W	R	(+)	(+)	IN	(-)	(+)
8 ^a	Right MX lateral incisor	U	W	R	(-)	(+)	IN	(-)	(-)
9	Right MD canine to left first molar	U	W	R	(+)	(+)	IN/OUT	(-)	(+)
Total	MX 6, MD 3	U 9	W 9	R 9	7	8	OUT 3, IN 2, IN/OUT 1	1	6

IN, impacted tooth inside the tumour; MD, mandible; MX, maxilla; OUT, impacted tooth outside the tumour; R, regular; U, unilocular; W, well-defined.

^aPreviously reported cases [8].

(+), positive results; (-), negative results.

Figure 1. (a) (Case 1) Panoramic view shows unilocular radiolucencies with a well-defined border in the right mandible canine to molar area. Root absorption (arrowhead) and root divergence are observed (arrow), with radio-opaque material seen in the lesion. (b) Intra-oral view shows buccolingual bony expansion in the right mandible canine to molar area and radio-opaque material in the lesion (arrow). (c) Axial CT image shows unilocular radiolucencies with a well-defined border in the right mandible canine to molar area and buccolingual bony expansion. Radio-opaque materials are located at the periphery. The shape appears to be linear (arrow). The CT value of this radio-opaque material is 170.3 HU. (d) Coronal CT image shows unilocular radiolucencies with a well-defined border in the right mandible canine to molar area and buccolingual bony expansion. Radio-opaque materials are located at the periphery. The shape appears to be linear (arrow). The CT value of this radio-opaque material is 170.3 HU.

radiographic features were similar to those in Tanimoto et al's [6] report. Thus, conventional radiographic features of CCOTs are thought to be root resorption and teeth divergence. This suggests that a CCOT might have features more like a tumour than a cyst. Whether odontomas are frequently associated with CCOTs is a matter of debate [3]. In our series, odontomas were found in four cases on CT images, whereas a single case was detected by conventional radiographs. The detection of odontomas within CCOTs

has been thought to depend on radiographic examinations. Moreover, CCOTs are held to be associated with odontomas. With CT values, radio-opaque bodies within the lesion were diagnosed as odontomas; the value of the radio-opaque body was 1000–2000 HU. Thus, CT was useful for detecting odontomas.

In our series, by CT, radio-opaque bodies were found in eight of nine cases and even with conventional radiographs, they were found in six of nine. The frequency of

Figure 2. (a) (Case 4) Panoramic view shows unilocular radiolucencies with a well-defined border in the right maxilla lateral incisor area (arrow). (b) Intra-oral view shows root divergence and radio-opaque material (arrow) in the lesion. (c) Axial CT image before contrast enhancement. (d) Axial contrast-enhanced CT image shows no enhancement in the lesion. (e) Axial CT image shows that radio-opaque material is located at the periphery and the shape appears to be linear (arrow). The CT value of this radio-opaque material is 280.2 HU.

Figure 3. (a) (Case 6) Panoramic view shows unilocular radiolucencies with a well-defined border in the right maxilla canine area. Root divergence and an impacted tooth are observed outside the lesion, with radio-opaque materials seen in the lesion (arrows). (b) Axial CT image shows radio-pacity located at the periphery and the shape appears to be linear (arrows). The CT value of this radio-pacity material is 220.6 HU. (c) In the sagittal CT image, the CT value of this radio-opaque material is >1700 HU (arrow) and thought to indicate an odontoma (arrow).

radio-opaque bodies in our series was higher than that reported by the WHO (50%) [4].

No contrast enhancement of the lesion after administration of contrast was found in two cases. But, buccolingual bony expansion was seen in seven of nine cases. Thus, this also suggests that a CCOT might be more like a tumorous lesion than a cystic lesion.

Rushton and Horner [7] reported that a peripheral band was a characteristic pathological finding on CT in two cases of CCOTs, although the CT value of the peripheral band was not measured. Yoshiura et al [2] reported radiographic findings, including CT images, for four cases of COCs. The CT value was measured in three of the four cases, and as the CT value in two cases was about 30 HU, no calcification was thought to have occurred. In our series, CT values of radio-opaque bodies were measured in all nine cases. On CT, in eight of nine cases,

radio-opaque bodies were found within the tumour and in the remaining case, the wall of the tumour was calcified. On conventional radiographs, radio-opaque bodies were detected in six of nine cases. However, the locations and shapes of the radio-opaque bodies were not determined in detail. By CT, radio-opaque bodies were located in the periphery of the tumour in eight of nine cases and the shape of the radio-opaque body was linear in five cases and linear and spotted in three. The CT values of the radio-opaque bodies ranged from 170.3 to 371.0 HU, indicating that radio-opaque bodies might include calcification. Linear calcification has been reported to occur in 40% of COC cases [7]. The frequency of linear calcification was higher than this in our series. Our findings suggest that a characteristic CT image of a CCOT was a radio-opaque body located in the periphery of the tumour, with the shape of the radio-opaque body

Figure 4. (a) The cystic wall of the tumour is lined by stratified odontogenic epithelium with ghost cells and dystrophic calcification (haematoxylin and eosin; bar, 200 μ m). (b) The ghost cells are anucleate keratinised cells and retain the outline of the cell membrane (haematoxylin and eosin; bar, 50 μ m).

being linear and/or spotted. Thus, CT was useful in distinguishing a CCOT from the other calcifying lesions.

Conclusions

On conventional radiograph and CT images, a CCOT was a well-defined unilocular lesion with regular margins. Teeth divergence and root resorption were

common. Impacted teeth were found in six of nine cases. CT was superior to conventional radiographs in detecting buccolingual expansion, odontomas and radio-opaque bodies. While CT values inside the tumour were 18.6–64.2 HU, the CT values of radio-opaque bodies were 170.3–371.0 HU. On CT images, radio-opaque bodies were typically located in the periphery of the tumour and the shape was linear and/or spotted. CT imaging was useful in diagnosing a CCOT.

Table 2. Results of CT scanning

Case	Site	Locus	Border	Margin	Root absorption	Teeth divergence	Impacted tooth	Odontoma	Radio-opaque bodies
1	Right MD canine to molar	U	W	R	(+)	(+)	(-)	(-)	(+)
2	Right MD lateral incisor to canine	U	W	R	(+)	(+)	(-)	(-)	(+)
3	Right MX molar	U	W	R	(+)	(-)	OUT	(+)	(-)
4	Right MX lateral incisor	U	W	R	(-)	(+)	(-)	(-)	(+)
5	Right MX canine	U	W	R	(+)	(+)	OUT	(+)	(+)
6	Left MX canine	U	W	R	(+)	(+)	OUT	(+)	(+)
7 ^a	Right MX canine	U	W	R	(+)	(+)	IN	(+)	(+)
8 ^a	Right MX lateral incisor	U	W	R	(-)	(+)	IN	(-)	(+)
9	Right MD canine to left first molar	U	W	R	(+)	(+)	IN/OUT	(-)	(+)
Total	MX 6, MD 3	U 9	W 9	R 9	7	8	OUT 3, IN 2, IN/OUT 1	4	8

IN, impacted tooth inside the tumour; MD, mandible; MX, maxilla; OUT, impacted tooth outside the tumour; R, regular; U, unilocular; W, well-defined. (+), positive results; (-), negative results.

Table 3. Results of CT images findings

Case	Maximal length of the tumour (mm)	Buccolingual bony expansion	Contrast enhancement	CT value (inside tumour)	CT value (radio-opaque bodies)	Location of radio-opaque bodies	Shape of radio-opaque bodies
1	40	(+)		18.6	170.3	Peripheral	Linear
2	15	(+)	(-)	37.1	215.0	Peripheral	Linear, spotted
3	23	(-)		36.8	NA	NA	NA
4	15	(+)	(-)	35.8	280.2	Peripheral	Linear
5	23	(+)		41.8	230.3	Peripheral	Linear
6	13	(-)		25.0	220.6	Peripheral	Linear, spotted
7 ^a	17	(+)		46.0	371.0	Peripheral	Linear
8 ^a	22	(+)		64.2	232.0	Peripheral	Linear
9	60	(+)		35.4	245.0	Peripheral	Linear, spotted
	Mean, SD: 25.3 ± 15.3	7	0	Mean, SD: 37.9 ± 12.8	Mean, SD: 245.5 ± 69.4	Peripheral: 8	Linear: 5, Linear, spotted: 3

Dot, calcification; Linear, linearcalcification; NA, not accountable; SD, standard deviation.

^aPreviously reported cases [8].

(+), positive results; (-), negative results.

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