Clinical Case Report

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Characteristics of chondroid lipoma

A case report and literature review

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Abstract

Rationale: Chondroid lipoma (CL) is a rare benign tumor. No relevant epidemiological reports have been published on CL, and there is a lack of uniform diagnostic and treatment criteria for the tumor.

Patient concerns: Here, we report a case of CL with a mass on the left buttock for 2 weeks, and further illuminate its diagnosis and treatment.

Diagnosis: The diagnosis of CL was rendered according to the pathological indices.

Interventions: The tumor was resected completely under spinal anesthesia.

Outcomes: The patient was followed-up for 6 months and showed no tumor recurrence or metastasis and there was resolution of the patient's lower-limb numbness and pain.

Lessons: The case study presented here provides evidence that CL could be effectively diagnosed by using ultrasound, puncture or biopsy, and/or magnetic resonance imaging. Furthermore, the patient recovered without any complications after completely resecting the tumor.

Abbreviations: CDFI = color Doppler flow imaging, CL = chondroid lipoma, CT = computed tomography, FISH = fluorescence in situ hybridization, HE = hematoxylin-eosin, HU = Hounsfield unit, MRI = magnetic resonance imaging, RT-PCR = reverse transcription-polymerase chain reaction, SPECT = single-photon emission computed tomography, STIR = short-time inversion recovery, T1WI = T1-weighted image, T2WI = T2-weighted image.

Keywords: case report, chondroid lipoma, epidemiology, genetics, imaging, pathology

1. Introduction

Chondroid lipoma (CL) is a rare benign tumor, and was first reported by Meis and Enzinger[1] in 1993. To date, there is no consensus regarding for its diagnosis or treatment. It is often misdiagnosed and mistreated by clinicians, who frequently have a poor understanding of these tumors, resulting in significant economic loss and physical and mental harm to patients. In the present study, after describing a case of CL, we search the relevant literature and integrate what is known about the epidemiological, pathological, imaging, and genetic characteristics of CL, as well as its treatment and prognosis.

2. Case report

A 56-year-old man was admitted due to a mass on the left buttock. Physical examination revealed a 4×3 cm rounded mass. The mass was tough with no fluctuation, and no adhesion to the surface of skin. The patient felt radiative pain and numbness around the knee joint when the mass was pressed. Prehospital ultrasound examination revealed a heterogeneous low echo of 30×24-mm-sized zone in the left gluteal muscle layer with irregular morphology and blurred boundaries; color Doppler flow imaging (CDFI) revealed no obvious blood flow signal in the mass. In-hospital ultrasound examination (Fig. 1A) revealed a heterogeneous $86 \times 47 \times 34$-mm-sized zone with a slightly high intensity echo, irregular morphology, blurred boundaries, and uneven internal echo in the left gluteal muscle layer. Computed tomography (CT) examination (Fig. 1B) revealed a 48×28-mm-sized mass-like low-density shadow in the left gluteus maximus, with a CT value of approximately −80 Hounsfield units (HU). A nodular soft tissue density shadow and dot-like, high-density shadow were also visible. Magnetic resonance imaging (MRI) examination revealed a mixed high and low signal density on T1-weighted image (T1WI) of the left gluteus maximus (Fig. 2A, B). A high signal density was observed on T2-weighted image (T2WI) (Fig. 2C) and a mixed high and low signal density was observed on fat-suppression sequence T2WI (T2WI-FS) of the left gluteus maximus (Fig. 2D, E, F).

Ultrasound-guided puncture pathology revealed that there was adipose tissue in the left gluteus maximus and that adipocytes had differentiated and matured, in some regions differed in size. There also seemed to be adipoblasts present. Cartilage and bony tissue were observed partially. It did not rule out an atypical lipoma-like mass accompanied by cartilage and ossification.

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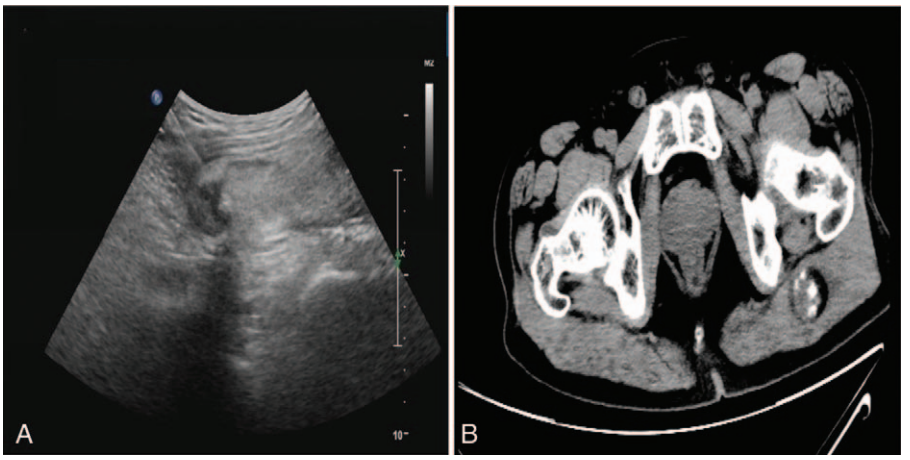

Figure 1. A, Preoperative ultrasound image of the mass revealed a heterogeneous zone with a slightly high intensity echo, irregular morphology, blurred boundaries, and uneven internal echo. B, Preoperative computed tomography (CT) image of the mass revealed a nodular soft-tissue-density shadow and dot-like high-density shadow.

Considering that a malignant tumor was not excluded, single-photon emission computed tomography (SPECT)/CT imaging was performed, which depicted the proximal part of the left femur as a radioactivity concentrated shadow and the left gluteus maximus as an oval low-density shadow, with a CT value of about −86 HU. The maximum cross-sectional area was approximately 52×28 mm. Separation, irregular soft tissue density, and high-density changes were further detected, with a CT value as high as approximately 166 HU. A high-density shadow with enhanced radioactivity uptake was also observed.

Under spinal anesthesia, the mass was resected. During the surgery, we found that the mass, about $10 \times 10 \times 4$ cm in size, located in the deep part of the gluteus maximus, was movable, had an intact capsule, and had no adhesions to the adjacent

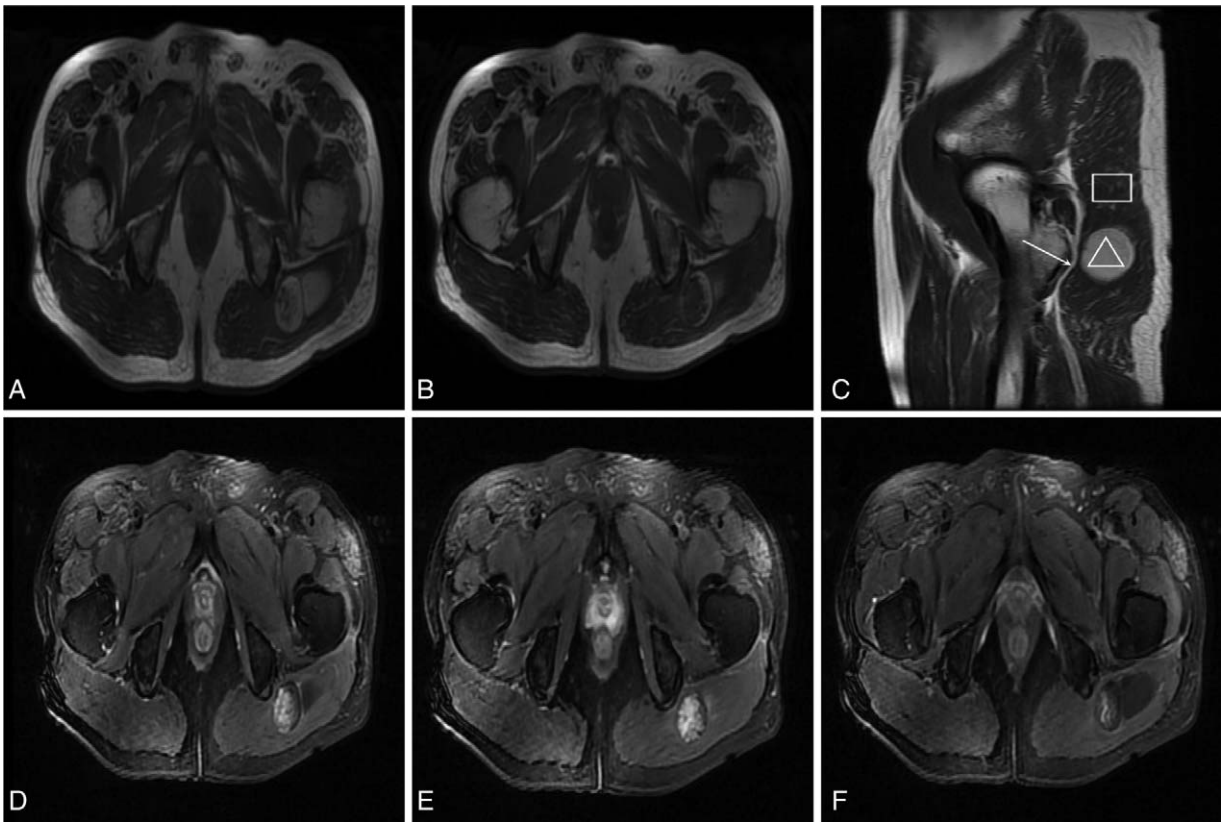

Figure 2. Magnetic resonance imaging (MRI) examination of CL. A, B, Preoperative T1-weighted image (T1WI) of the mass revealed mixed high and low signal density. C, A high signal intensity was revealed on T2-weighted image (T2WI). White arrow indicates the sciatic nerve, "Δ" indicates the mass, and "□" indicates the gluteus maximus. D, E, F, Mixed high and low signal intensity on T2WI-FS.

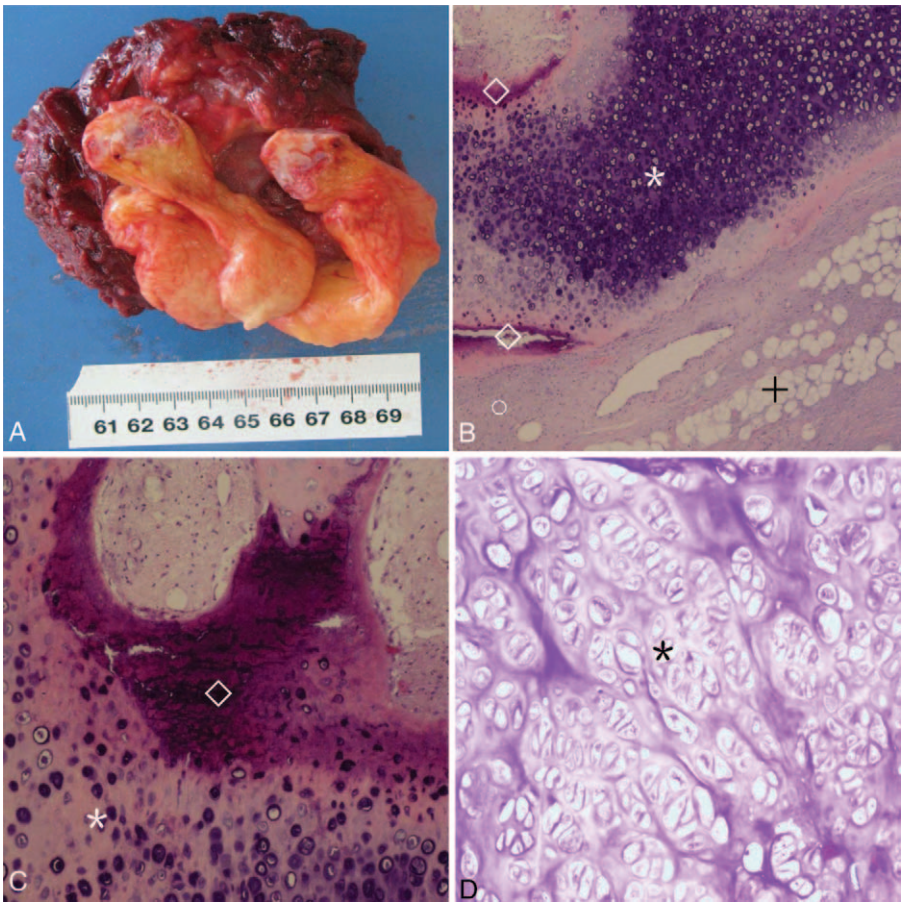

Figure 3. A, Postoperative tumor appearance: 10 × 10 × 4 cm-sized yellowish or reddish-brown irregular mass. B, (4 × 10 times), C, (10 × 10 times), D, (10 × 20 times). Postoperative hematoxylin-eosin (HE) staining image via bright field microscopy. "*" indicates chondrocytes, "◇" indicates osteogenic tissue, "○" indicates fibrous tissue, and "+" indicates adipocytes.

tissue. After opening the capsule, a yellowish or reddish-brown lobulated, slightly tough mass was revealed (Fig. 3A). Frozen sections (thickness, 5 μm) of the mass revealed that there was a large amount of cartilage tissue accompanied by ossification, and the proliferation of the peripheral fibrous and adipose tissue with no obvious cellular atypia. Paraffin-embedded sections of the mass (Fig. 3B, C, D) revealed an adipose-derived tumor with chondro-osseous metaplasia. Heterologous interstitial cells were not observed in the mass. According to these pathological indices, a diagnosis of CL was rendered, without performing further immunohistochemistry. After surgery, adjuvant treatments were not administered. A follow-up examination 6 months later revealed no tumor recurrence or metastasis, with resolution of the patient's lower-limb numbness and pain. Written informed consent was obtained from the patient before the submission of the manuscript.

3. Discussion

CL is a rare benign tumor, first reported by Meis.[1,2] There is no consensus regarding for its diagnosis or treatment. To better understand the published context of the current study, we performed an online search of Embase, Web of Science, Wanfang ,and PubMed biomedical databases to retrieve relevant case reports or series of case reports. After excluding nonoriginal publications or those without original data, a final of 64 publications[1–64] including 88 cases (including the case presented here), were analyzed for epidemiological, histopathological, imaging, and genetic characteristics, as well as diagnosis, treatment, and prognosis. Collectively, this review provides an evidence-based reference for future clinical treatment and basic research into CL.

3.1. Epidemiological characteristics

Consistent with previous reports,[1,2] CL tends to occur at just about 40 (42.85 ± 16.97; median, 41) years of age and has no obvious geographical or ethnic trends. The difference is that in our review we found a male-to-female ratio of 1:1.6, and there is no obvious sex difference. The course of this disease ranges from 6 days[52] to 10 years.[24] CL tends to occur in the trunk (23/88, 26.14%), limbs (lower limbs 32/88, 36.4%, upper limbs 12/88, 13.64%), also in the mouths (7/88, 7.95%), feet (4/88, 4.55%), and uterus (1/88, 1.14%). The tumors are mainly found in skeletal muscle, subcutaneous fat, and occasionally in the superficial fascia and submucosa. Park et al[40] also reported a case of CL in the phrenic nerve. CL in the trunk and limbs mostly manifests as a painless mass, but CL with tenderness,[41,52,55] tingling,[40] and radiating pain (in the present case) are also noted in a few cases (12/71, 16.90%). When accompanied by pain, it

may be often misdiagnosed as neurofibromatosis[32] or schwannomas.[15,18] CL in the mouth manifests as polyps[54] while intrauterine CL presents with unexplained vaginal bleeding.[22] In the present case, upon the application of pressure to the tumor, the adjacent sciatic nerve was stimulated and the patient-reported numbness of the limbs and an electric shock–like radiating pain around the knee joint.

3.2. Histopathological characteristics

Generally, the performances of CL are mostly solid, yellowish, or reddish-brown masses of different sizes, with clear boundaries and an intact capsule. The cut surfaces appear mostly yellow-brow, white, pearly gray, or reddish-brown in color, gel-like in texture, and are occasionally accompanied by bleeding. In the past, CL was thought to be composed of white adipocytes, without any true cartilage differentiation.[3] Kindblom and Meis-Kindblom,[2] however, firstly confirmed the existence of cartilage differentiation in CLs. In the present review study, hematoxylin-eosin staining revealed that CLs are mainly composed of different proportions of mature adipocytes, adipoblasts, and mucus-transparent cartilage-like matrices, some of which are accompanied by ossification.

The volume of mature adipocytes is large, with cytoplasm and nucleus squeezed to the edges of the cells. There are also large fat droplets can be seen in the mature adipocytes. Adipoblasts are smaller than mature adipocytes, and often arranged in flakes and lines, as well as in nest- and cord-like structures under high magnification. The cytoplasm is eosinophilic and contains vacuoles of various sizes and numbers, and the nuclei are small in size, diverse in shape, and have no heteromorphism. Nucleolar and intranuclear pseudoinclusion bodies can be seen, and nuclear cleavage images are rare or absent. There are different amounts of mucus-transparent cartilage-like matrices, as well as partial cellulose depositions, between the mature adipocytes and adipoblasts. Vascular networks[15,17,18,42,47] are also present but lacking a typical plexiform vascular morphology, as in myxoid liposarcoma.

Immunohistochemical staining reveals that the more mature the adipoblast differentiation is, the higher degree of the positive expression rate of vimentin (30/32, 93.75%) and S-100 (42/45, 93.33%) is. In addition, the nuclear proliferation index is <1%, and Ki-67 proliferation index is low. In a few cases, KP1 (8/10, 80.00%), LAM (9/13, 69.23%), CD68 (9/14, 64.29%), and CK (7/27, 25.93%) are positive, whereas EMA (0/24), SMA (0/20), MSA (0/3), HMB45 (0/9), and GFAP (0/19) are negative. Furthermore, Gomez-Ortega et al[4] found that although most of the patients were female, it did not express the sex hormone–related estrogen receptor, progesterone receptor, or androgen receptor, indicating that CL was not associated with sex hormones. These pathological findings demonstrate that early puncture biopsy or tissue resection biopsy can provide evidence for the diagnosis of CL.

3.3. Imaging characteristics

There is evidence[30,45] that cases of CL often lack characteristic imaging findings due to inconsistent proportions of adipose tissues and cartilage-like tissues in the matrix. In the present study, we analyzed imaging data from 23 cases and have found that MRI is of significance to the diagnosis of CL. Mixed high and low signal intensity is appeared on T1WI and short-time inversion recovery (STIR) sequences, high-intensity signals is appeared on T2WI, and heterogeneous fat saturation is also appeared on T2W fat-saturated images. The low signal intensity on T1WI and high signal intensity on T2WI indicate the presence of chondroid tissue. The high-intensity signals on T1WI are inhibited on STIR images, indicating the presence of fat tissue,[5] which shows nonuniform fat saturation on T2W fat-saturated images. There was a "fat ring" sign on the contrast-enhanced and fat-saturated image in a previous study. This sign reflected cartilage tissue in the center of a mass with low signal intensity on T1WI, high signal intensity on T2WI, and delayed ring- or arc-shaped enhancement patterns after intravenous injection of a contrast agent. The surrounding adipose tissue exhibited a signal stronger than that of muscle on the T1WI and T2WI, and heterogeneous fat saturation on the T2W fat-saturated image.[58]

Green et al[19] reported positive x-ray changes in the imaging of CL, suggesting the presence of calcification-like structures in the mass. Patne et al,[32] however, reported that CL should not be excluded when the findings of x-ray were negative. This is because the x-ray images can be used for the identification of ossification but not cartilage. Similarly, a diagnosis of CL cannot be excluded when negative CT findings are obtained. Therefore, routine auxiliary diagnosis of CL by x-ray and CT is not recommended because of the high false negative rate associated with these 2 imaging methods.

Recently, SPECT/CT has been widely used in the differential diagnosis of tumors. In a study by Escobar et al,[51] CL was shown to have a high affinity for fludeoxyglucose (18F-FDG), the accumulation of which is positively correlated with the level of glucose metabolism in tissues and organs. High affinity for 18F-FDG is more common in malignant tumors and inflammatory tissues. So, SPECT/CT with 18F-FDG lacks specificity for the diagnosis of CL. In our case presented here, the patient's mass has a high affinity for methylenediphosphonate (99mTc-MDP), which is specific for bone tissue or cartilage tissue.[65] Therefore, 99mTc-MDP is of more value than 18F-FDG in the diagnosis of CL. However, because of the high cost associated with SPECT/CT and lack of sufficient evidence support for its use in this application, we do not recommend it as a routine examination of CL.

In our case, we investigated the application of ultrasound in the diagnosis of CL. CL appears lobulated due to the presence of a large number of fibrous membranes, resulting in poor ultrasound image quality. In addition, the mass often contains a large amount of cartilage-like tissues, resulting in a strongly echogenic mass. The mass often shows a heterogeneous mixed imaging signal, due to that the content and morphology of cartilage-like tissues vary greatly, and the shape is irregular. In addition, due to the lack of a typical vascular plexus, CDFI has no obvious blood flow signal. As an inexpensive, noninvasive imaging examination, ultrasound should be further validated and promoted for the use in the diagnosis of CL.

3.4. Cellular genetics

Genetic diagnosis has attracted increasing attention for the diagnosis of rare and refractory diseases. In the present study, we have found that CL has the characteristics of chromosome t (11; 16) (q13; p13) translocations and *C11*or *f95-MKL2* fusion genes. Thomson et al[10] firstly performed a genetic study of CL and found chromosome t (11;16) (q13;p12–13) equilibrium translocation in these tumors. Gisselsson et al[8] had successively verified those findings. Although the common type of lipomas, other

Table 1

Genetic characteristics of chondroid lipoma.

Case	Reference	Year	Karyotype
1	Huang et al[37]	2010	46,XX,t(6;12)(p21;q13),t(11;16)(q13;p13.2)[23]/47,idem,þr,0–5dmin[8]/46,XX[11]
2	Huang et al[37]	2010	46,XX,t(11;16)(q13;p13)[13]/46,XX[4]
3	Huang et al[37]	2010	46,XY,t(11;16)(q13;p13)[5]/46,XY[10]
4	Ballaux et al[17]	2004	46,XX,t(11;16)(q13;p13)[20]/47–48,XX,idem,+2–3r[cp2]
5	Gisselsson et al[8]	1999	46,XY,t(1;2;5)(q32;q37;q31),t(11;16)(q13;p13)[24]/46,XY[1]
6	Thomson et al[10]	1999	46,XY,t(11;16)(q13;p12–13)

types of lipomas, and hibernating tumors could also be found to have chromosome abnormalities of 11q13, they do not reorganize with 16p12–13, allowing for differentiation of this particular tumor type (Table 1).

Huang et al[37] and Flucke et al[48] used the novel fluorescence in situ hybridization (FISH) and reverse transcription-polymerase chain reaction (RT-PCR) techniques to study chromosome t (11;16) (q13;p13) translocations, and found a specific expression of the fusion gene *C11* or *f95-MKL2* in CL, which had not been found in other tumors. As such, they proposed that FISH and RT-PCR assays could serve as valuable diagnostic adjuncts for this rare disease entity, particularly when evaluating small samples or when fresh tissue was not available.

3.5. Differential diagnoses

In the past, due to the lack of correct understanding, CLs were often misdiagnosed as lipomas,[16,24,25,35] neurofibromas,[32] schwannomas,[15,18] teratomas,[52] and even malignant tumors, resulting in unnecessary lymph node dissection,[1,47] enlarged lesion resection,[1,44] and toe amputation.[1] This is a waste of medical resources and unnecessary physical and mental damage to patients and could be avoided with an adequately precise diagnostic rubric for CL. For malignant tumors, such as extraskeletal chondrosarcoma or myxoid chondrosarcoma also contain mature chondrocytes, adipoblasts, mature adipocytes, and myxoid matrix,[1] so they need to be identified through using a combination of the aforementioned pathological, MRI, and genetic assessments.

3.6. Treatment and prognosis

The treatment of CL is mainly based on simple lesion resection, and does not require adjuvant treatment postoperative. Diode lasers can be used for the treatment of affected tissues in sensitive areas, such as in the oral cavity. The advantages of this technique include that it causes no intraoperative bleeding, requires no suturing, and is associated with less postoperative pain, bleeding, swelling, and inflammation than other techniques. In addition, the therapeutic use of lasers leads to rapid wound healing.[33] Of all 88 cases, during the longest follow-up period of 36 years,[1] only 1 case[1] of CL had a recurrence (no specific information was provided in the original literature). As in our study, Boets et al[18] presented that the confirmed CL with good postoperative recovery and no need for follow-up.

4. Conclusions

In conclusion, CL is a rare benign tumor that can be accurately diagnosed by puncture or biopsy, MRI, and genetic examination. For those not accompanied by symptoms such as pain, numbness, and rapid growth, which may impact the quality of patients' life, a simple follow-up may be performed. Finally, a simple focal resection is effective treatment for CL.

Author contributions

All authors substantially contributed to the manuscript. Chao Huang and Wen-Lai Guo designed the study and performed the literature review, extraction of the data, and analysis of the pooled data. Wen-Rui Qu and Zhe Zhu performed surgical resection and perioperative care. Rui Li and Zhe Zhu reviewed and edited the manuscript. All authors read and approved the final manuscript.

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