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WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Langerhans cell histiocytosis presenting as an isolated brain tumour: A case report

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Abstract

BACKGROUND

Langerhans cell histiocytosis (LCH) is a rare proliferative histiocyte disorder. It can affect any organ or system, especially the bone, skin, lung, and central nervous system (CNS). In the CNS, the hypothalamic-pituitary is predominantly affected, whereas the brain parenchyma is rarely affected. LCH occurring in the brain parenchyma can be easily confused with glioblastoma or brain metastases. Thus, multimodal imaging is useful for the differential diagnosis of these intracerebral lesions and detection of lesions in the other organs.

CASE SUMMARY

A 47-year-old man presented with a headache for one week and sudden syncope. Brain computed tomography (CT) and magnetic resonance imaging showed an irregularly shaped nodule with heterogeneous enhancement. On ^{18}F -fluorodeoxyglucose (^{18}F -FDG) positron emission tomography/CT, a nodule with ^{18}F -FDG uptake and multiple cysts in the upper lobes of both lungs were noted, which was also confirmed by high-resolution CT. Thus, the patient underwent surgical resection of the brain lesion for further examination. Postoperative pathology confirmed LCH. The patient received chemotherapy after surgery. No recurrence was observed in the brain at the 12-mo follow-up.

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CONCLUSION

Multimodal imaging is useful for evaluating the systemic condition of LCH, developing treatment plans, and designing post-treatment strategies.

Key Words: Langerhans cell histiocytosis; Brain neoplasms; Lung; Computed tomography; Magnetic resonance imaging; Positron emission tomography/computed tomography; Case report

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Core Tip: Langerhans cell histiocytosis (LCH) is a rare hematological disease characterized by a clonal proliferation of abnormal langerhans cells. It can affect any organ or system, especially the bone, skin, lung, and central nervous system (CNS). In the CNS, the hypothalamic-pituitary is predominantly affected, whereas the brain parenchyma is rarely affected. Cases of LCH involving the brain parenchyma and presenting as an isolated brain tumour have been reported, but all the reports lack complete multimodal imaging. In this manuscript, we have reported a case of LCH involving the brain parenchyma and bilateral lungs, which was assessed using computed tomography (CT), high-resolution CT, magnetic resonance imaging, and ¹⁸F-fluorodeoxyglucose positron emission tomography/CT. Furthermore, we have reviewed the relevant literature.

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INTRODUCTION

Langerhans cell histiocytosis (LCH) is an uncommon disease characterized by clonal proliferation of myeloid precursors that differentiate into cluster of differentiation (CD)1a⁺/CD207⁺ (Langerin) cells in lesions[1,2]. It mainly affects children, with a reported incidence of 4-5 cases *per* million children aged < 15 years *per* year, while its incidence in adults is uncertain[3]. LCH may affect any organ or system, but it most frequently affects the bone, skin, pituitary, liver, spleen, hematopoietic system, lung, lymph nodes, and central nervous system (CNS)[4,5]. Cases of LCH involving the brain parenchyma and presenting as an isolated brain tumour have been reported, but all the reports lack complete multimodal imaging. Herein, we have reported a case of LCH involving the brain parenchyma and bilateral lungs, which was assessed using computed tomography (CT), high-resolution CT (HRCT), magnetic resonance imaging (MRI), and ¹⁸F-fluorodeoxyglucose (¹⁸F-FDG) positron emission tomography (PET)/CT. Furthermore, we have reviewed the relevant literature.

CASE PRESENTATION

Chief complaints

A 47-year-old man was referred to our hospital with a headache for one week and sudden syncope in the morning.

History of present illness

A 47-year-old man was referred to our hospital with a headache for one week and sudden syncope in the morning.

History of past illness

The patient had no history of polyuria or polydipsia. No other illnesses were observed.



Personal and family history

The patient had no known comorbidities or family history, but had a 15-year smoking history.

Physical examination

No rash or positive neurological signs were found on physical examination.

Laboratory examinations

Laboratory tests results showed increased in carcinoembryonic antigen (CEA) levels (7.03 ng/mL, reference: 0-5 ng/mL), with no other abnormal findings.

Imaging examinations

Non-contrast brain CT showed irregularly shaped nodular foci with isodensity at the left frontal corticomedullary junction. Large patches of hypodense edema were noted in the adjacent white matter (Figure 1A and B represent the lateral ventricular and basal ganglia levels, respectively). Contrast-enhanced brain CT showed significant heterogeneous enhancement in the left frontal foci (Figure 1C and D, the same level as the Figure 1A and B). Coronal and sagittal views of contrast-enhanced CT images showed irregular morphology of the lesion and poor demarcation with the adjacent skull (Figure 1E and F). Bone window CT showed no abnormalities in the adjacent skull (Figure 1G and H, the same level as the Figure 1E and F). Subsequently, the patient underwent a brain MRI. Axial T1-weighted images (T1WI) showed heterogeneous hypointense lesions in the left frontal lobe (Figure 2A). Axial T2-weighted images (T2WI) showed a heterogeneously mixed hyperintensity signals with hypointense areas in the left frontal lobe lesion (Figure 2B). After administration of gadolinium, the lesion showed heterogeneous enhancement on axial (Figure 2C), coronal (Figure 2D), and sagittal T1WI (Figure 2E). No abnormalities were found on sagittal T1WI of the sellar region (Figure 2F).

Considering the elevated CEA levels and CT and MRI manifestations, further investigation was required to rule out brain metastases. Therefore, ¹⁸F-FDG PET/CT was performed. Maximum-intensity-projection imaging showed a focal increase in ¹⁸F-FDG uptake in the right maxillary sinus and multiple foci of increased ¹⁸F-FDG uptake in the bilateral lung fields (Figure 3A). Axial (Figure 3B-D) and coronal (Figure 3E-G) views of the selected PET, non-enhanced CT (NE-CT), and fused PET/CT images showed moderately increased ¹⁸F-FDG uptake in the left frontal nodule [the maximum standardized uptake value (SUVmax) of the lesions and surrounding tissues are shown in Supplementary Figure 1]. No abnormal ¹⁸F-FDG uptake was observed in the sellar region (Supplementary Figure 2). Axial (Figure 3H-J) views of the selected PET, NE-CT, and fused PET/CT images showed multiple cysts with peripheral exudation in the upper lobes of bilateral lungs, with slightly increased ¹⁸F-FDG uptake. HRCT was performed to further evaluate the pulmonary lesions. Axial (Figure 4A), coronal (Figure 4B), and sagittal (Figure 4C and D, left and right lungs, respectively) views of HRCT images showed multiple scattered small thick-walled irregular cysts and small nodules. Sinusitis was diagnosed in the right maxillary sinus. Bilateral lung manifestations should be differentiated from pulmonary LCH, but brain nodules are more difficult to diagnose and should be differentiated from gliomas.

Further diagnostic work-up

The patient underwent brain tumour resection. Gross examination showed that the specimen was a grey-brown solid tumour (Figure 5A) and the cut surface was grey-brown and grey-white (Figure 5B). Histopathological examination revealed mononucleated and multinucleated histocytes with abundant cytoplasm and slight staining (haematoxylin and eosin, magnification, × 200; Figure 5C). On immunohistochemistry, the specimen stained positive for S100, CD207 (Langerin), CD4, and CD1a, and negative for CD3 and CD20. Ki67 (MIB-1) index was slightly > 30%.

FINAL DIAGNOSIS

The final histological diagnosis was LCH.

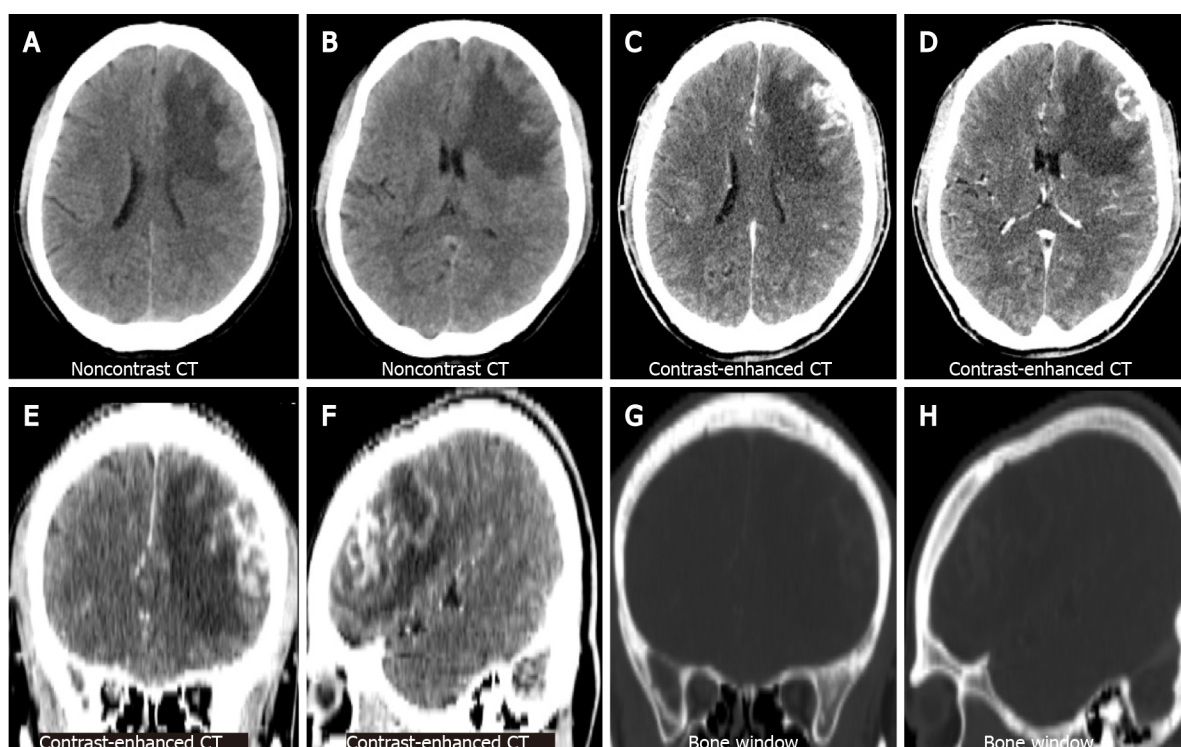


Figure 1 Brain computed tomography. A and B: Represent the lateral ventricular and basal ganglia levels on non-contrast computed tomography (CT), respectively. An irregularly shaped nodule is observed in the left frontal lobe with large perifocal low-density oedema; C and D: Represent the same level as the former on contrast-enhanced CT. The nodules are significantly enhanced heterogeneously; E and F: Represent the coronal and sagittal views of the contrast-enhanced CT; G and H: Represent the same level as the former, with no abnormalities in the adjacent skull. CT: Computed tomography.

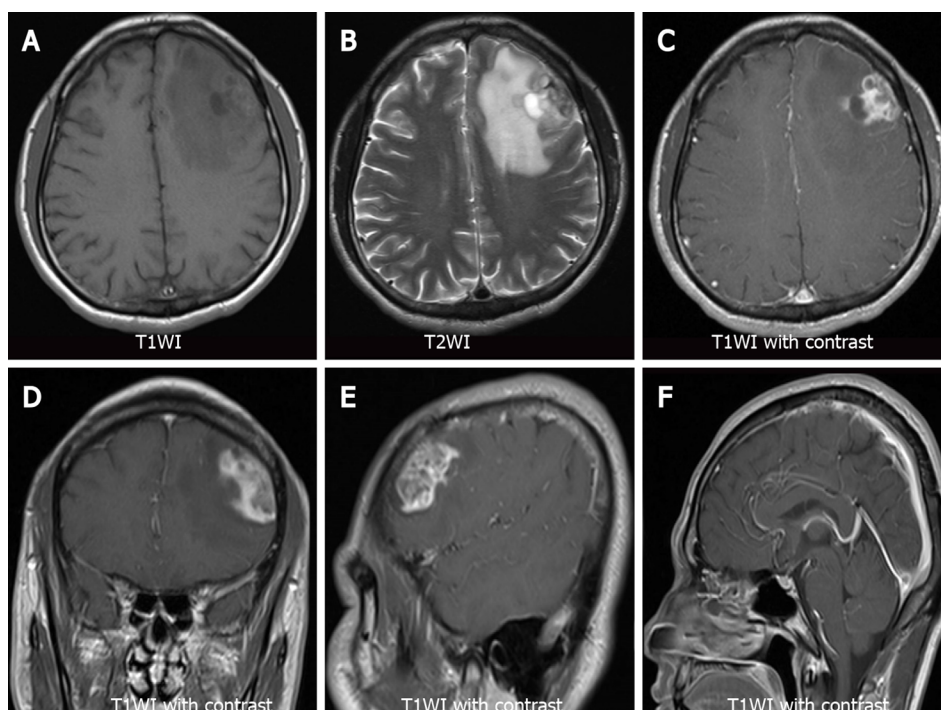


Figure 2 Brain magnetic resonance imaging. A: Axial T1-weighted images (T1WI) show heterogeneous hypo-intensity of the left frontal lobe lesion; B: Axial T2-weighted images (T2WI) show heterogeneously mixed signal of hyperintensity with hypointense areas of the left frontal lobe lesion; C-E: Axial, coronal, and sagittal views of T1WI with contrast agent administration show heterogeneous enhancement of the lesion; F: Sagittal T1WI show no abnormality in the sellar region. T1WI: T1-weighted images; T2WI: T2-weighted images.

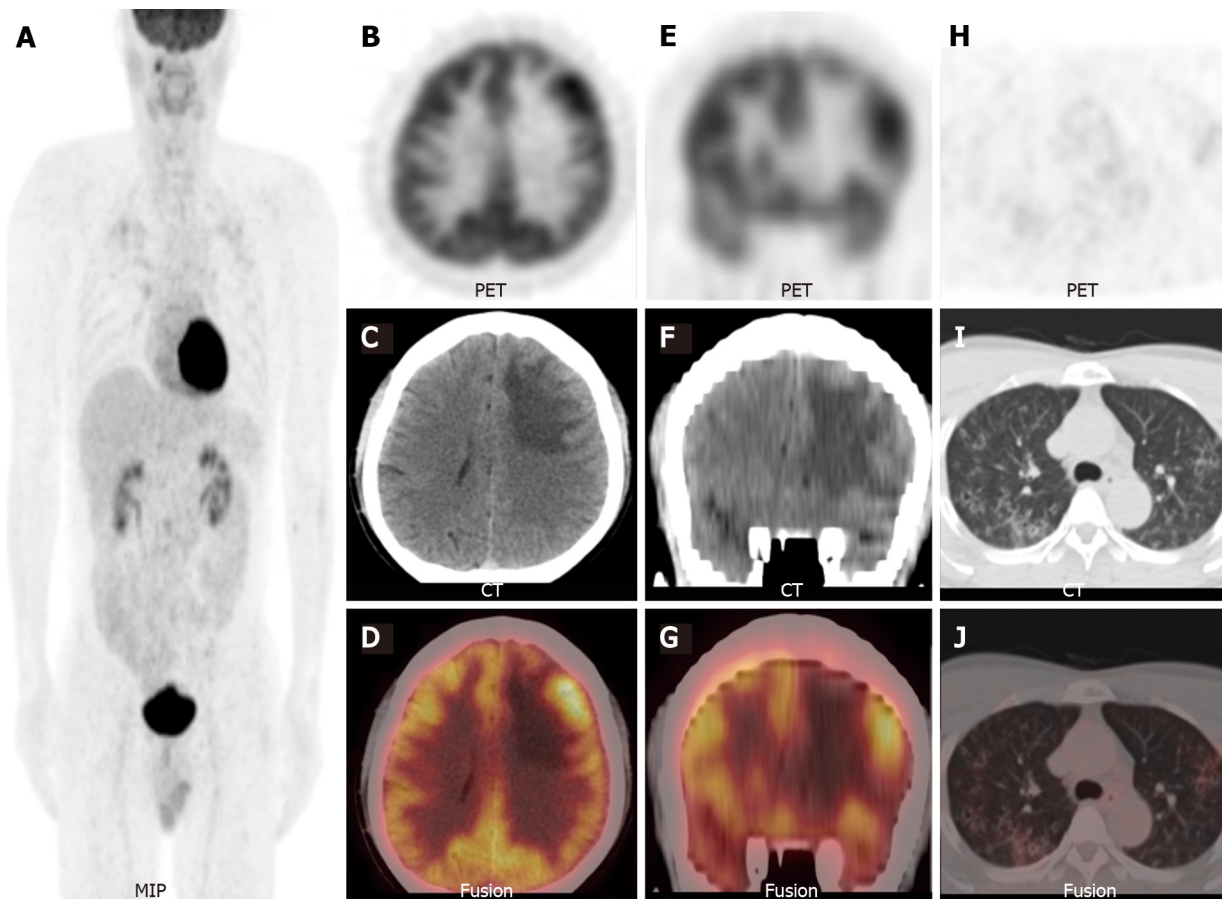


Figure 3 ^{18}F -fluorodeoxyglucose positron emission tomography/computed tomography. A: Maximum-intensity-projection show a focal ^{18}F -fluorodeoxyglucose (^{18}F -FDG) uptake lesion in the right maxillary sinus and multiple foci with ^{18}F -FDG uptake in the bilateral lung field; B-G: Axial and coronal views of the selected positron emission tomography (PET), non-enhanced computed tomography (NE-CT), and fused PET/CT images show the left frontal lesion with ^{18}F -FDG uptake (SUVmax 9.5, arrowheads); H-J: Axial views of the selected PET, NE-CT, and fused PET/CT images show multiple cysts with peripheral exudation in the upper lobes of bilateral lungs, with slightly increased ^{18}F -FDG uptake (SUVmax 3.2). MIP: Maximum-intensity-projection; PET: Positron emission tomography; CT: Computed tomography.

TREATMENT

The patient received chemotherapy (vindesine and prednisone acetate) after surgery.

OUTCOME AND FOLLOW-UP

No recurrence was observed on brain MRI at the 12-mo follow-up ([Supplementary Figure 3](#)).

DISCUSSION

LCH involving the hypothalamic-pituitary or skull is not uncommon, but involvement of the brain parenchyma, such as the frontotemporal lobe, is rare. As of January 2019, fewer than 30 cases have been reported in the PubMed database ([Table 1](#))[6-8]. We reviewed the relevant PubMed literature from 1990 to May 2021 and found 16 cases of brain parenchymal LCH with imaging data. The mean age was 31 years (95% confidence interval: 21.5-41.2). The male-to-female ratio was 14:2, which is consistent with that reported in previous literature reviews of intracerebral LCH, but higher than that in children with LCH[3,9,10]. The lesions were mostly located in the frontotemporal lobe (14 cases), particularly in the frontal lobe. The clinical presentation of LCH is non-characteristic and varies depending on the site. Most cases showed non-specific symptoms of mass effect such as headache, seizures, hemiparesis, and/or sensory disturbances. MRI findings without contrast were also largely non-specific.

Table 1 Summary of 16 cases with brain parenchymal langerhans cell histiocytosis with imaging data

Ref.	Age/sex	Diameter (cm) ¹	Location (Lobe)	MRI Finding			¹⁸ F-FDG PET/CT finding
				T1WI	T2WI	T1WI with contrast	
Caresio <i>et al</i> [20], 1991	29/Male	3.0	Right temporal lobe	Hypointense	Hyperintense	Ringlike enhancement	NA
Itoh <i>et al</i> [21], 1992	7/Male	NA	Right frontal lobe	Hypo-/iso-intense	Hyperintense	Intense enhancement	NA
Bogaert <i>et al</i> [22], 1994	40/Female	NA	Left parietal lobe	Hypointense	Hyperintense	Intense enhancement	NA
Vital <i>et al</i> [23], 1996	32/Female	NA	Right insula lobe	NA	Hyperintense	Intense enhancement	NA
Grant <i>et al</i> [24], 1999	20/Male	3.5	Right temporal lobe	NA	NA	Intense enhancement	NA
Katati <i>et al</i> [25], 2002	36/Male	NA	Left temporal lobe	Hypointense	NA	Intense enhancement	NA
Cagli <i>et al</i> [9], 2004	24/Male	1.5	Left temporal lobe	Hypointense	NA	Intense enhancement	NA
Yamaguchi <i>et al</i> [26], 2004	2/Male	NA	Multiple lesions/bilateral frontal and temporal lobes	NA	NA	Intense enhancement	NA
Rodríguez-Pereira <i>et al</i> [10], 2005	30/Male	5.0	Left frontal lobe	NA	NA	Gyral enhancement	NA
Rodríguez-Pereira <i>et al</i> [10], 2005	65/Male	2.5	Left parietal lobe	NA	NA	Peripheral enhancement	NA
Dieter [27], 2017	4/Male	2.0 ¹	Multiple lesions, right frontal, and parietal lobe	Iso-/hyper-intense	Hypointense	Uniform enhancement	NA
Cai <i>et al</i> [6], 2014	23/Male	4.1	Right frontal lobe	Hypo-/iso-intense	Iso-/hyper-intense	Moderate to intense homogeneous enhancement	NA
Dardis <i>et al</i> [28], 2015	64/Male	NA	Multiple lesions, left frontal and right temporal lobe, and brainstem	NA	Hyperintense	Patchy enhancement	NA
Kim <i>et al</i> [7], 2018	36/Male	3.0	Right frontal lobe	Isointense	Hyperintense	Heterogeneous enhancement	NA
Bärtschi <i>et al</i> [8], 2019	42/Male	NA	Right insular lobe	NA	Hyperintense	Intense enhancement	NA
Current case	47/Male	3.6	Left frontal lobe	Hypointense	Hyperintensity with hypointense areas	Heterogeneous enhancement	Lesion SUVmax 9.5 and contralateral SUVmax 8.1

¹The largest lesion.

NA: Not available; PET: Positron emission tomography; CT: Computed tomography; T1WI: T1-weighted images; T2WI: T2-weighted images; MRI: Magnetic resonance imaging.

Nonetheless, MRI showed hypointensity on T1WI and hyperintensity on T2WI in most cases. After administration of gadolinium, most cases showed intense homogeneous or heterogeneous enhancement. Another characteristic feature is sulcal enhancement around the lesion[6,7]. MRI images showed leptomeningeal involvement near the lesions in several cases, as reported by Kim *et al*[7]. This may be a characteristic sign of brain parenchymal LCH, but it needs to be confirmed in more cases.

There are no previous reports of ¹⁸F-FDG PET/CT for assessing the metabolic activity of brain parenchymal LCH. To our knowledge, our case report is the first with a PET/CT description. The SUVmax of the brain lesion was approximately 9.5, which was similar to the SUVmax of LCH lesions involving other regions reported in the literature[11]. Additional bilateral lung lesions were found, and pulmonary manifestations were decisive for diagnosis[12]. As 30% patients with LCH present with multi-organ system involvement, it is important to detect involvement of other tissues (such as the bone, soft tissue, the CNS, or the lungs)[3,13]. Single or isolated brain lesions

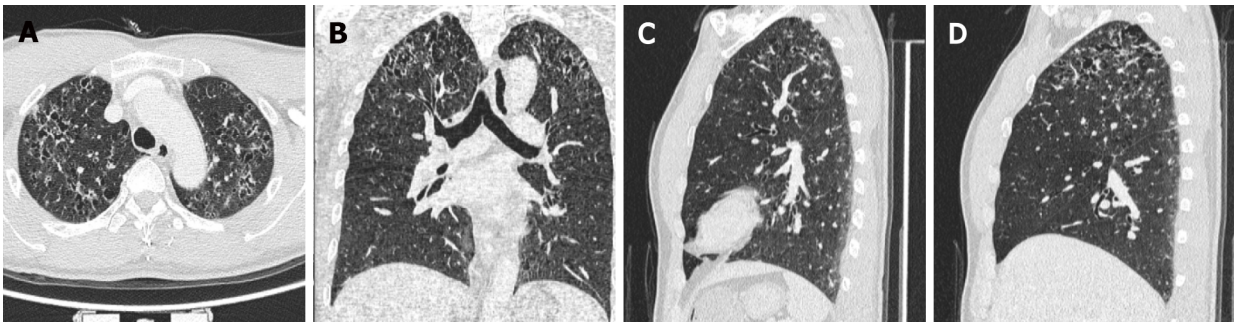


Figure 4 High-resolution computed tomography of the lung. Axial, coronal, and sagittal (the left and right lungs, respectively) views of high-resolution computed tomography images show multiple scattered small thick-walled irregular cysts as well as small nodules. A: Axial; B: Coronal; C: Left lungs; D: Right lungs.



Figure 5 Histopathological images. A: The specimen is a greyish brown and greyish dark solid tumour; B: The cut surface is greyish brown and greyish white; C: Histopathological examination reveal mononucleated and multinucleated histocytes with abundant cytoplasm and light staining (haematoxylin and eosin, magnification, $\times 400$).

have previously been reported based on only brain CT or MRI, without whole-body scans[6,7]. Without whole-body evaluation, reports of isolated brain lesions may be non-rigorous or biased. More recent studies that performed whole-body evaluations have identified a higher rate of focal LCH lesions than that previously reported[14,15]. Therefore, PET/CT or PET/MRI seems to be more appropriate for evaluating this disease[16]. This is especially true for combined bone and lung lesions as some case without obvious symptoms are incidentally detected; they may be missed by relying solely on radiography or CT[8]. Several studies have confirmed the diagnostic value of systemic scans, such as PET/CT or PET/MRI for LCH[14,15,17,18]. The diagnostic evaluation of LCH plays a crucial role in treatment planning. PET/CT or PET/MRI can be used to assess multiple foci throughout the body, guide biopsy sites, and assist with post-treatment strategies.

Based on prospective trials, the combination of vinblastine plus prednisolone is the most commonly used induction chemotherapy regimen and is administered over six weeks[19].

CONCLUSION

As a systemic disease, LCH has the potential to involve the brain parenchyma, and its diagnosis is extremely challenging. The use of multimodal imaging or whole-body imaging, combined with the manifestation of lesions at other sites, can be helpful in the diagnosis of this disease. Moreover, multimodality imaging is useful for assessing the systemic status of LCH, developing treatment plans, and evaluating post-treatment strategies.

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