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Case Report

A Surgical Case of Lipomatous Hypertrophy of the Atrial Septum

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Abstract

We present a rare case of lipomatous hypertrophy of the atrial septum (LHAS) characterized by the abnormal accumulation of adipose tissue within the interatrial septum, forming a tumor-like mass. The reported incidence of LHAS is 2.2–8%, with advancements in imaging techniques expanding diagnostic opportunities. A 75-year-old female with a history of meningioma, right breast cancer, cholecystolithiasis, and ovarian cyst; comorbidities, including diabetes, hypertension, and hyperlipidemia; and a body mass index of 33 kg/m^2 underwent resection for an enlarging interatrial septal tumor, ultimately leading to the diagnosis of LHAS. Imaging revealed a 36 mm × 28 mm tumor extending from the atrial septum into the right atrium, with a computed tomography value of –76 HU. Histopathologically, the tumor was mainly composed of mature adipocytes along with hypertrophied myocardial fibers and scattered brown adipocytes, confirming the diagnosis of LHAS. Although LHAS is often managed with observation following diagnosis, our report on surgical intervention presents a rare and noteworthy occurrence. This case highlights the importance of recognizing and addressing the rare occurrence of LHAS, which often requires surgical intervention in cases of tumor enlargement. The successful resection and diagnosis of LHAS in this patient underscore the importance of considering this condition in the differential diagnosis of cardiac tumors.

Keywords

lipomatous hypertrophy; atrial septum; cardiac tumor; interatrial septum; imaging modality

Introduction

Lipomatous hypertrophy of the atrial septum (LHAS) is a benign lesion characterized by an abnormally massive accumulation of fat in the interatrial septum, forming a tumor-like mass. The prevalence of LHAS is estimated to be 2.2–8% [1,2]; however, with the advancement in diagnostic imaging, diagnostic opportunities for LHAS have increased. Herein, we report a case in which an enlarged atrial septal tumor was surgically removed, leading to the diagnosis of LHAS.

Case Report

A 75-year-old female was referred for evaluation of a right atrial tumor detected on chest computed tomography (CT). Four months before admission, she presented to our emergency department with palpitations and underwent a chest CT scan. She had a history of undergoing surgical procedures for meningioma, right breast cancer, cholecystolithiasis, and ovarian cysts. Additionally, she had been treated for diabetes, hypertension, hyperlipidemia, and obesity (body mass index: 33 kg/m^2) at our hospital. On admission, blood tests and chest radiography yielded unremarkable results, and an electrocardiogram showed sinus rhythm with no arrhythmia.

A contrast-enhanced CT scan displayed a 36 mm × 28 mm mass extending from the atrial septum into the right atrium, with a CT value of –76 HU (Fig. 1A,B). Upon retrospective chart review, a contrast-enhanced CT scan performed at our hospital a decade ago revealed a 23 mm × 19 mm mass in the same location, which had persisted since that time (Fig. 1C,D). Magnetic resonance imaging (MRI) revealed a tumor in the atrial septum with high signal intensity on both T1-weighted and T2-weighted images and signal suppression with fat suppression (Fig. 2). Echocardiography revealed a hyperechoic mass protruding into the right atrial side of the atrial septum (Fig. 3). Coronary angiography revealed no significant stenosis of the coronary arteries. Notably, there was no discernible blood flow within the tumor using either modality.

Despite being asymptomatic, aside from palpitations, the tumor exhibited growth on imaging, prompting our decision to proceed with surgical resection. The procedure was performed under cardiopulmonary bypass, and cardiac arrest was induced. Following a right atrial incision, the surface of the atrial septal wall was smooth, with a palpable mass (Fig. 4A,B). After incising the atrial septal wall

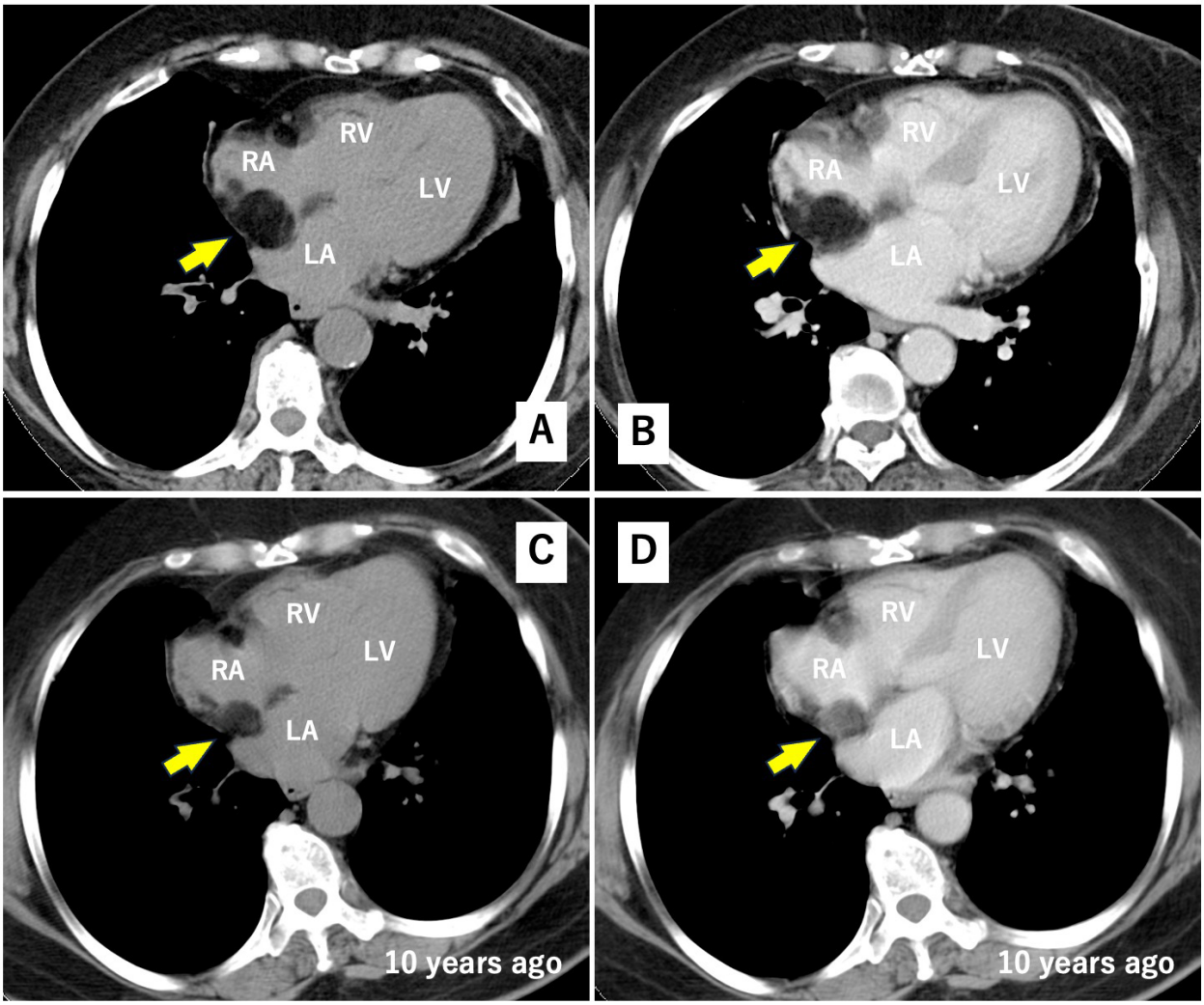

Fig. 1. LHAS on CT. CT images taken before surgery (A,B) and 10 years ago (C,D) are compared. Both show a mass in the atrial septum with low attenuation on plain CT (A,C) and no enhancement on contrast-enhanced CT (B,D) (yellow arrows). In addition, the size of the mass has increased from 23 × 19 mm 10 years ago to 36 × 28 mm. LHAS, lipomatous hypertrophy of the atrial septum; CT, computed tomography; RV, right ventricle; RA, right atrium; LV, left ventricle; LA, left atrium.

on the right atrial side, adipose tissue was discovered, and the boundary was indistinct. Whereas confirming the tumor through palpation, the surrounding area was dissected using an electrocautery scalpel, and the tumor was excised en bloc (Fig. 4C,D).

Pathological examination revealed a tumorous lesion characterized by mature adipocytes accompanied by hypertrophic myocardial fibers and occasional brown adipocytes, confirming the diagnosis of LHAS (Fig. 5). The patient's preoperative palpitations resolved postoperatively, and there were no signs of recurrence observed on contrast-enhanced CT at the 2-year postoperative assessment (Fig. 6) or on transthoracic echocardiography during the 3-year follow-up.

Discussion

LHAS was initially described by Prior in 1964 [3]. The reported incidence varies based on the diagnostic modality employed, ranging from approximately 2.2% in patients who undergo multislice CT to 8% in those who undergo transesophageal echocardiography (TEE) [1,2]. Histologically, LHAS is characterized by a benign cardiac anomaly infiltrated with mature adipocytes interwoven with myocardial fibers and fetal fat cells, commonly referred to as "brown cells" [4,5]. The accumulation of adipose tissue occurs both cranially and caudally in the oval fossa, with the fossa remaining intact, resulting in the distinctive "dumbbell" appearance of LHAS [6]. Atrial septal hypertrophy typically extends more extensively on the cranial

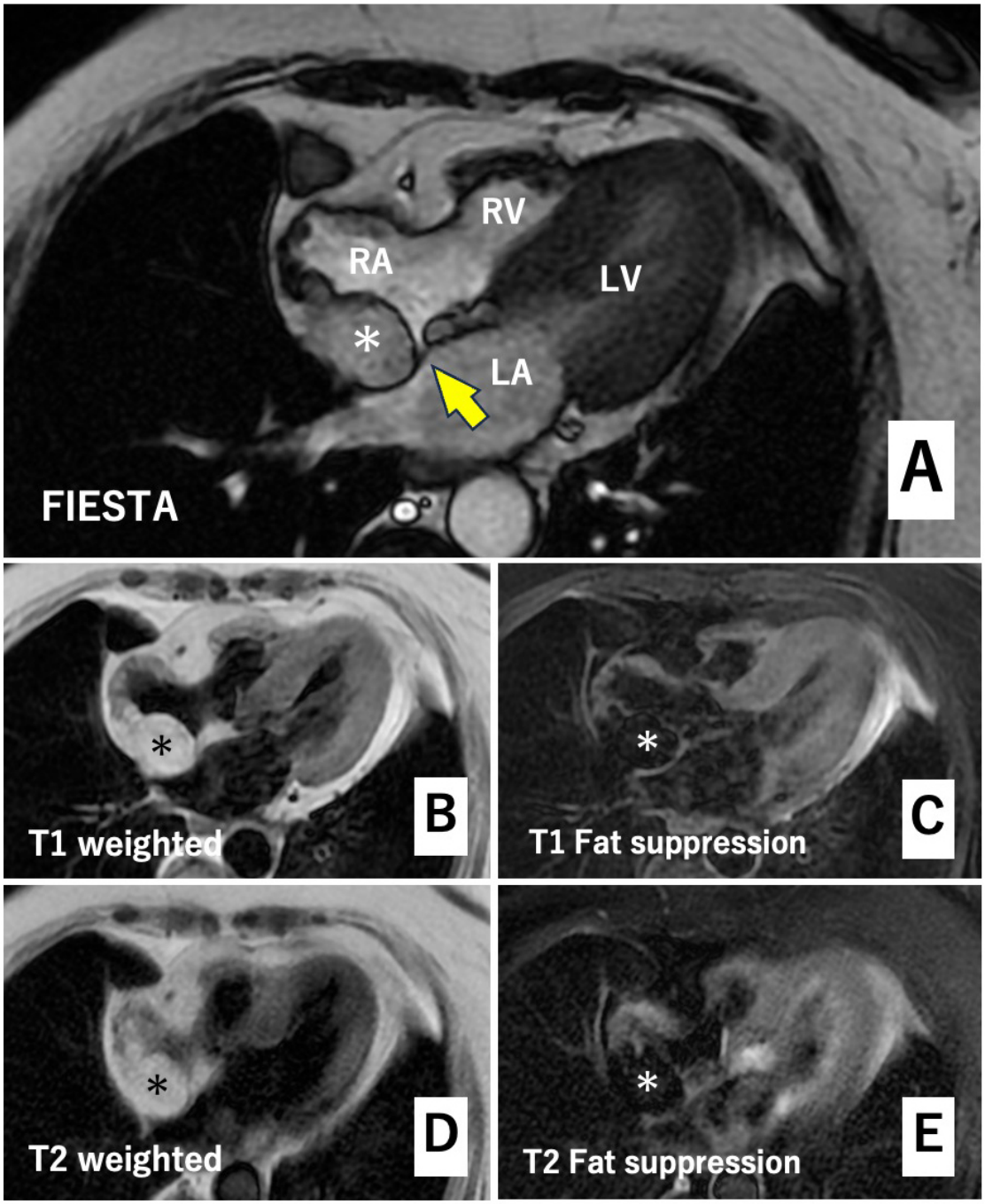

Fig. 2. LHAS on MRI. FIESTA shows a well-defined mass (*) in the atrial septum with preservation of the fossa ovalis (A; yellow arrow). The mass (*) shows a high signal (B) on T1-weighted images and a low signal (C) with fat suppression. It also shows high (D) and low (E) signals with fat suppression on T2-weighted images, consistent with the characteristics of LHAS. LHAS, lipomatous hypertrophy of the atrial septum; MRI, magnetic resonance imaging; FIESTA, fast imaging employing steady-state acquisition.

side than on the caudal side and protrudes into both the left and right atrial chambers. The differential diagnosis of LHAS encompasses both benign and malignant cardiac tumors involving the interatrial septum, including metastasis, myxoma, rhabdomyoma, fibroma, fibroblastic tumors, and mesothelioma [7].

The diagnosis of LHAS employs various imaging modalities, including echocardiography, CT, and cardiac magnetic resonance (CMR) [2]. On plain and contrast-enhanced CT, LHAS manifests as a localized, homogeneous, dumbbell-shaped area of fat attenuation confined to the interatrial septum [8]. By discerning the relative density, CT aids in distinguishing LHAS, characterized

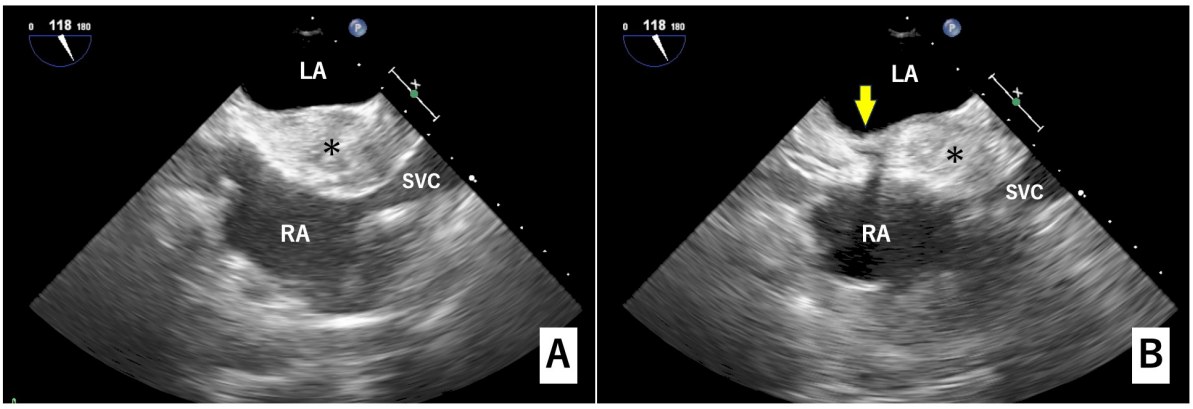

Fig. 3. LHAS on TEE. A mass protruding into the right atrium is observed in the atrial septum (*) (A). The fossa ovalis is preserved (yellow arrow) (B). LHAS, lipomatous hypertrophy of the atrial septum; TEE, transesophageal echocardiography; SVC, superior vena cava.

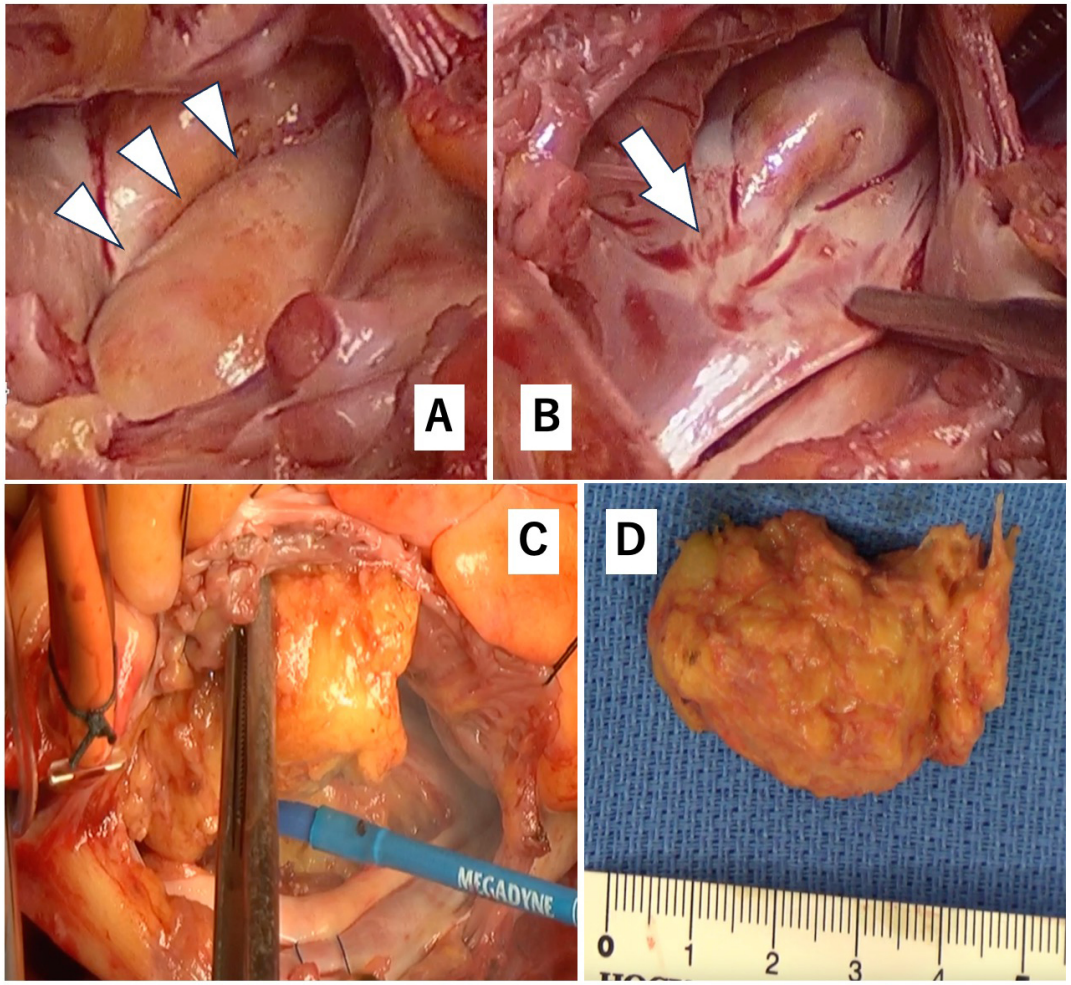

Fig. 4. Intraoperative findings. The right atrial incision reveals a smooth surface of the atrial septal wall (A, arrowhead) and preservation of the fossa ovalis (B, arrow). The atrial septal wall is incised, and the tumor is removed as a single mass (C). The removed mass is 3 cm × 4.5 cm in size (D).

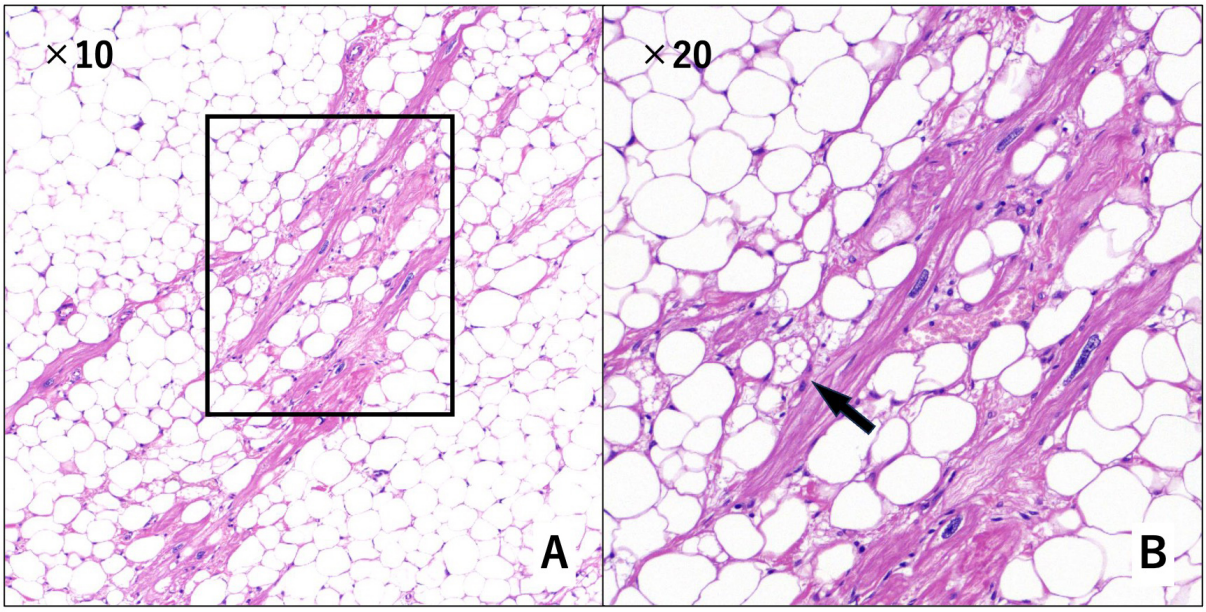

Fig. 5. Pathological findings. Hematoxylin and eosin staining; at 10×, the tumor is largely composed of mature adipocytes (A); magnification of the square in A reveals brown adipocytes with foamy spores (B; black arrow).

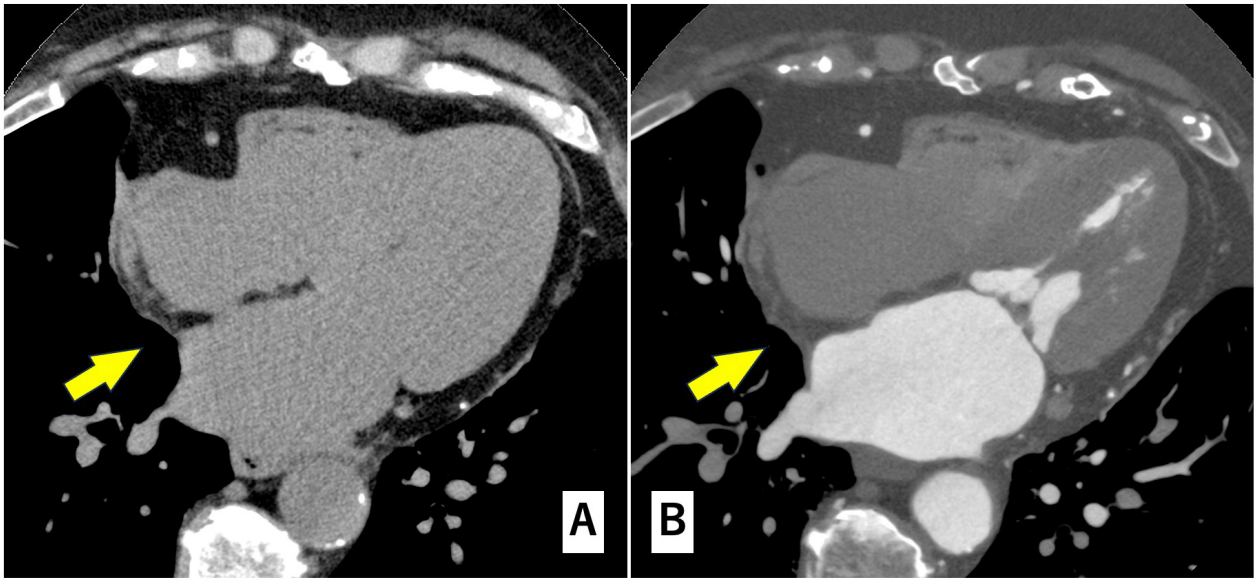

Fig. 6. CT findings at 2 years postoperatively. The CT scan taken during the second postoperative year showed no obvious recurrence of LHAS (Yellow arrow, A: plane, B: contrast-enhanced).

by its specific fat attenuation coefficient, from neoplasms or other tissues [9]. In this case, a low-attenuation area was observed in the interatrial septum on plain CT, and no enhancement was noted on contrast-enhanced CT, consistent with the characteristic imaging findings of LHAS. CMR imaging is pivotal for determining the extent of LHAS involvement in the interventricular septum and free wall. However, owing to their similar signal characteristics, LHAS may be mistaken for lipomas. The differential diagnosis of lipoma is based on specific morphological features of LHAS, such as fat infiltration exceeding 2 cm in diameter, infiltration of the rim of the oval fossa, and preservation of the oval fossa membrane [4,7]. Additionally, CMR is valuable not only for distinguishing malignant tumors but also for identifying features suggestive of malignancy, such as infiltration into extracardiac structures, involvement of two or more ventricles, infiltration into the right heart, tissue heterogeneity, indistinct borders, lesion

diameter exceeding 5 cm, and the presence of pericardial or pleural effusion [7]. The present case demonstrates the above characteristics of LHAS. Two-dimensional transthoracic echocardiography and TEE play crucial roles in distinguishing LHAS from other cardiac tumors based on the characteristic appearance and location of LHAS. Cardiac echocardiography is valued for its cost-effectiveness and minimal risk of adverse effects [4]. Regardless of the imaging modality used, the characteristic dumbbell-shaped appearance of the interatrial septum, with preservation of the oval fossa, is considered a distinctive feature of LHAS [4]. In this case, the characteristic findings of LHAS were observed in all CT, MRI, and TEE examinations.

Although most patients with LHAS are asymptomatic, they can occasionally be associated with atrial arrhythmias and, even more rarely, with malignant arrhythmias or sudden cardiac death [1]. This is believed to occur because of the progressive destruction of myocardial fibers from the accumulated fat, which results in impaired contractility and electrical conduction [9]. Considering that the occurrence rate of atrial arrhythmias is related to the thickness of the septum, it has been suggested that the total amount of adipose tissue is significantly involved in the occurrence of arrhythmias [10,11]. According to case reports linking LHAS to severe arrhythmias and sudden death, the anatomical distribution of LHAS within the interatrial septum, particularly in the sinoatrial node region, may contribute to the incidence of malignant arrhythmias [10,11]. Furthermore, reports suggest an association between LHAS, obesity, and advanced age [1,2], which also aligns with the risk factors for atrial fibrillation. However, an independent relationship between LHAS and supraventricular arrhythmias has not yet been established [12], warranting further research [10].

Given its benign nature, asymptomatic LHAS is typically managed through observation. However, surgical resection may be warranted in cases where patients experience atrial or malignant arrhythmias or if there is a tendency for LHAS enlargement. Additionally, it is feasible to perform LHAS resection concurrently with other cardiac surgical procedures, such as a biopsy [13]. In this case, although LHAS was considered the most likely diagnosis due to obesity, advanced age of 75 years, and characteristic findings on various imaging examinations, the clear trend of tumor enlargement over the past 10 years raised some suspicion of a malignant neoplasm. Therefore, a tumor resection was performed. While the pathological diagnosis confirms LHAS, cases such as the present one, where CT, MRI, echo examinations, and pathological findings all align, are considered quite rare. These findings are anticipated to provide valuable information for similar cases encountered in the future.

Although this is a single case report, it is important to acknowledge the limitations in generalizing the findings from this case. Surgical resection of LHAS remains a rare occurrence, making it challenging to assess the recurrence rate and long-term prognosis after tumor resection. Consequently, future analyses involving multiple cases, including the present one, are warranted.

Conclusions

We recently encountered a case in which surgical resection was performed to treat LHAS. Aligning all preoperative, intraoperative, and pathological findings is rare, and we believe that this report will be useful in future clinical practice. The CARE checklist was used when writing this case report (**Supplementary Table 1**).

Abbreviations

LHAS, lipomatous hypertrophy of the atrial septum; CT, computed tomography; MRI, magnetic resonance imaging; FIESTA, fast imaging employing steady-state acquisition; TEE, transesophageal echocardiography; CMR, cardiac magnetic resonance.

Availability of Data and Materials

The data supporting the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions

MH designed the study and made the major contributions to the manuscript. YH, RI, SY, and KI contributed to the data analysis and discussion. AH and MO were involved in data acquisition, analysis, and interpretation. All authors contributed to the editorial changes in the manuscript. All authors have read and approved the final version of the manuscript. All authors have participated sufficiently in the work to take public responsibility for appropriate portions of the content and agree to be accountable for all aspects of the work to ensure that questions are related to its accuracy or integrity.

Ethics Approval and Consent to Participate

Institutional review board approval was obtained before the publication of this manuscript. This study was conducted according to the “Helsinki Declaration” and was approved by the Institutional Review Board of Chubu Tokushukai Hospital (IRB: No.23-19). Before deciding to participate in the trial, the patient provided written informed consent.

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Conflict of Interest

The authors declare no conflict of interest.

Supplementary Material

Supplementary material associated with this article can be found, in the online version, at https://doi.org/10.59958/hsf.7055.

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