Heart Surgery Forum 2024; 27(9): E990–E997
doi: 10.59958/hsf.7771

Case Report

Cardiac Hemangioma Located in the Interatrial Septum: A Case Report and Literature Review

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Submitted: 13 June 2024 Revised: 11 July 2024 Accepted: 2 August 2024 Published: 8 September 2024

Abstract

Cardiac hemangiomas are extremely rare benign tumors that can occur in any part of the heart. We report the case of an adult female with no history of structural heart disease or symptoms of discomfort. Cardiac ultrasound showed a mass on the right atrial surface of the atrial septum. Intraoperatively, the mass was identified within the atrial septum, at the junction of the inferior vena cava and the ostium of the coronary sinus. The mass did not enter the left or right atrial cavities. The mass was successfully removed, and cardiac hemangioma was confirmed by postoperative pathology. This report discusses the clinical manifestations, diagnostic approaches, and treatment options for cardiac hemangiomas based on an analysis of previously reported cases. Owing to the risk of arrhythmia in patients with hemangiomas in the atrial or ventricular septum, surgical resection is recommended.

Keywords

cardiac hemangioma; cardiac neoplasms; cardiac surgery

Introduction

Cardiac hemangioma is a rare primary cardiac benign tumor [1]. The incidence of primary cardiac tumors is 0.002% in the general population, while the incidence of cardiac hemangiomas is only 2.8% of all primary cardiac tumors. Cardiac hemangiomas can occur in any part of the heart, and different locations lead to different complications [2]. The majority of patients with hemangiomas are asymptomatic, and most hemangiomas are found during autopsies or physical examinations [3]. Here, we report a case of an asymptomatic hemangioma located inside the atrial septum.

Case Report

A 63-year-old female patient presented for evaluation of an incidental finding of a hyperechoic mass in the right atrium during a routine physical examination. The patient's current medical history and physical examination of the heart showed no cardiac obstruction, with no abnormalities evident on 12-lead electrocardiography, chest radiography, or related blood tests, including tumor markers. Echocardiography revealed a homogeneous hyperechoic mass of approximately 18 × 22 mm in the right atrium, attached to the right atrial surface of the atrial septum, near the inferior vena cava, and firmly fixed in position. There were no obvious signs of blood flow in the mass (Fig. 1). Chest computed tomography (CT) showed a nodular high-density shadow at the posterior edge of the right atrium, with a CT value of approximately 37 HU (Fig. 2). After evaluation by the surgical team, the patient underwent tumor resection. Through a median sternotomy, a double-cannula approach was used to establish cardiopulmonary bypass. After the patient had cooled to 32 °C, the heart was perfused with cardioplegic solution to induce cardiac arrest. Exploration of the right atrium revealed normal atrial structure. The tumor was located within the atrial septum near the junction of the inferior vena cava and coronary sinus ostium, without invading either the left or right atria. The adventitia of the tumor was intact and measured approximately 20 × 15 × 10 mm. The tumor was completely resected, and because of the extent of the resection, autologous pericardial tissue was used to repair the atrial septal defect. The postoperative pathological report identified the tumor as a cardiac hemangioma (Fig. 3). The patient was discharged on postoperative day 7 without any complications. The patient has remained asymptomatic after 1 year of follow-up.

Discussion and Results

Cardiac hemangiomas are rare benign cardiac tumors that account for only 2.8% of all primary cardiac tumors [3]. Relevant literature reports from 2010 to 2024 were retrieved from the PubMed database using the keyword "cardiac tumor, hemangioma". Inclusion criteria: (1) duplicate cases in different articles were regarded as single cases; (2) cases with hemangioma finally confirmed by histopathology; (3) cases with complete clinical treatment process. A total of 34 case reports were included. Among them, 14 patients were male (41.2%), 19 were female (55.89%), and 1 was of un-

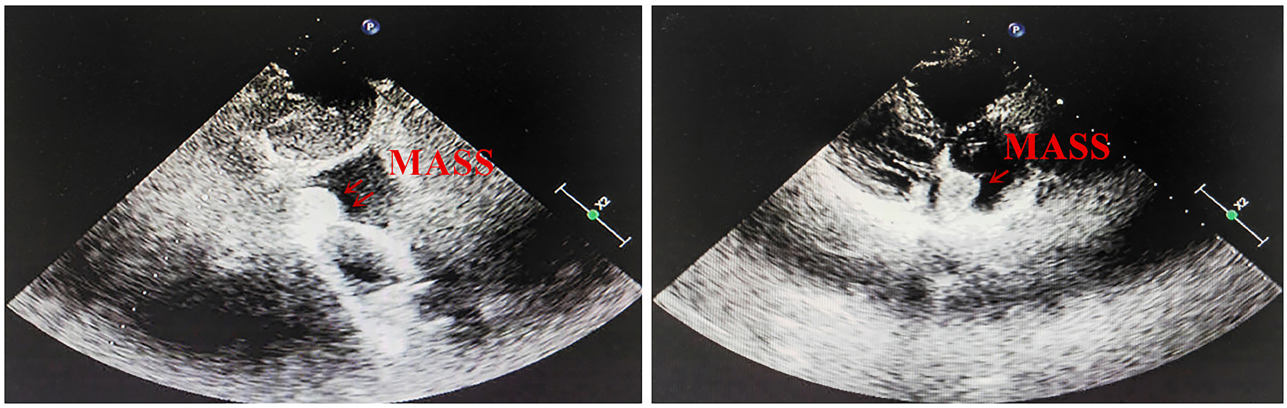

Fig. 1. Transthoracic Doppler ultrasound shows a mass in the interatrial septum, near the inferior vena cava. MASS means an abnormal mass, which is the cardiac hemangioma in this case.

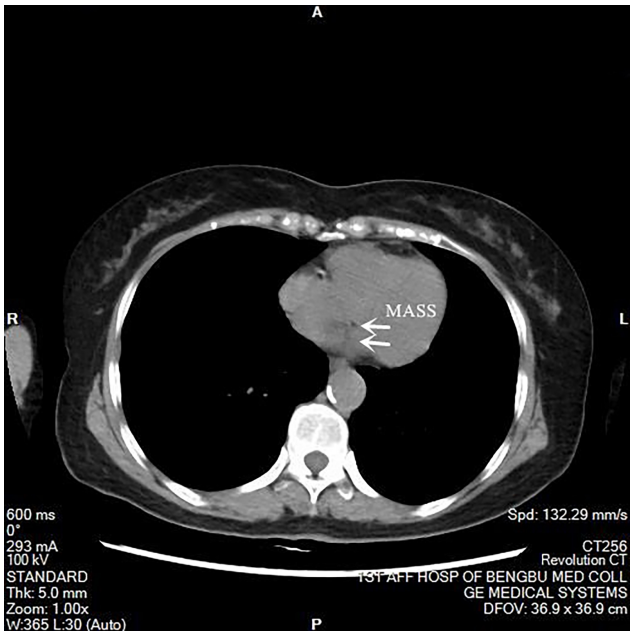

Fig. 2. Chest computed tomography shows abnormal nodular hyperdense shadows on the posterior border of the right atrium. The arrow points to MASS, which is the cardiac hemangioma described in this case report.

known sex. A review of the disease was performed by summarizing the clinical data of these published cases (Table 1, Ref. [4–37]). Cardiac vascularization can occur at any age, ranging from 6 months to 83 years. It can originate in any part of the heart, such as the pericardium, myocardium, and endocardium, but rarely in the atrial and ventricular septa. Among the 34 cases reported, 8 masses each were located in the right atrium and in the left atrium (23.5%), 9 in the right ventricle (26.5%), 6 in the left ventricle (17.6%), 1 in both the right atrium and ventricle (2.9%), and 1 each inside the atrial septum and in the pericardial cavity, originating from the left atrial appendage (2.9%). More masses were found in the right heart system compared to the left. In the cases reviewed, 8.8% of the masses were composed of multiple lesions (n = 3) while the remaining 91.2% were single tumors (n = 31). The pathological manifestation of the disease is the presence of benign proliferative endothelial cells in blood vessels, resulting in increased vascularization. Proliferating blood vessels can be classified into spongy, capillary, and arteriovenous types, of which the spongy type is the most common. In our data, 76.5% of the cases were cavernous hemangiomas (n = 26), 5.9% were arteriovenous hemangiomas (n = 2), 14.7% were capillary-type (n = 5), and 2.9% were spongy-capillary mixed type (n = 1). Unfortunately, molecular genetic research in cardiac hemangiomas is scarce. Studies have shown that vascular endothelial growth factors (VEGFs) may be associated with the formation of fetal cardiac hemangiomas. A crucial role of VEGFs during gestation is to promote the formation of myocardial blood vessels and improve blood flow, thereby maintaining the normal function of the placenta. However, genetic mutations of VEGFs or their receptors may lead to abnormally high expression, promoting the formation of hemangiomas and arteriovenous malformations. Some doctors have used β-receptor blockers to reduce the mechanism of VEGFs expression and treat cardiac hemangiomas, obtaining certain therapeutic effects. However, these approaches need to be further validated by additional research [38].

Most patients with cardiac hemangiomas have no clinical symptoms, and their clinical manifestations depend on the location, size, growth rate, and other tumor factors. Often, significant symptoms vary and occur only when the tumor invades or compresses the surrounding tissues. When a hemangioma is located in the pericardium and increases in size, a large pericardial effusion or cardiac tamponade may develop. When a hemangioma is located in the atrial or ventricular septum, the cardiac conduction system may be invaded and affected, causing arrhythmia. Cardiac hemangiomas are relatively fixed and, generally not easy to fall off. Unlike more common cardiac tumors, such as car-

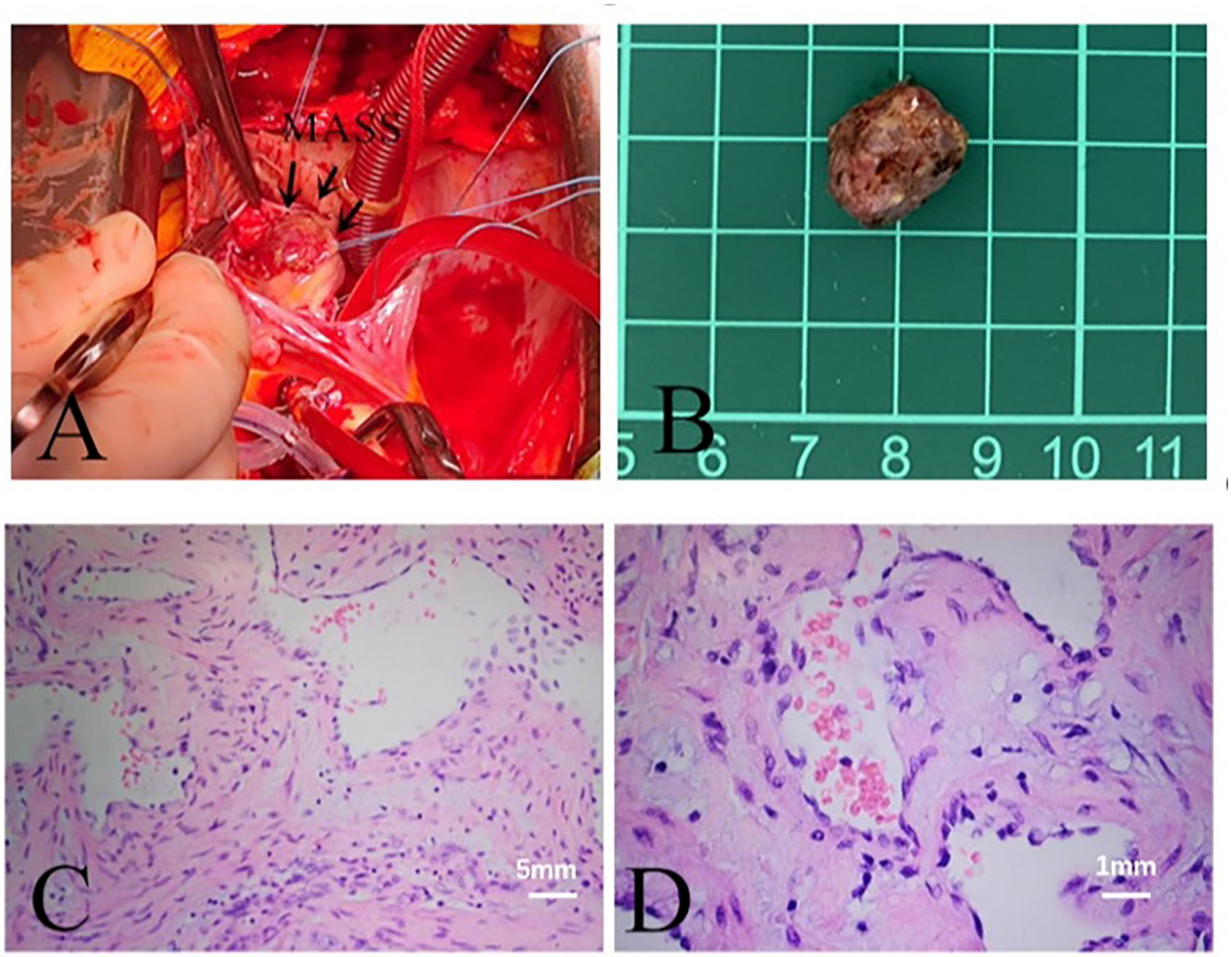

Fig. 3. Intraoperative exploration and resection of the hemangioma and postoperative pathological pictures. (A) The mass is located in the interatrial septum with an intact capsule; (B) The mass is completely removed; (C,D) Pathological examination shows small blood vessel-like tissue. The pathological type is cavernous hemangioma.

diac myxomas, whose, most common clinical manifestations involve mass loss that may cause infarction and affect the heart blood circulation, generally do not cause arrhythmia or pericardial effusion. In addition, cardiac myxomas often have a characteristic "stem-like" structure, which is connected to the tumor, and the length of the stem determines the activity of the mass. This characteristic structure can be found by cardiac ultrasound. Among the 34 cases, 8 patients were admitted to the hospital due to tumors found incidentally, without obvious clinical symptoms. Among the symptomatic patients, 1 had obvious arrhythmia, 9 had chest tightness and dyspnea, another 9 first developed palpitations, 3 each experienced syncope, chest pain, and fatigue, 1 developed arterial hypertension, and 2 each had pericardial effusion affecting circulation and edema. Some patients had more than two of the above symptoms simultaneously.

Imaging examinations can be used for the preoperative evaluation of hemangiomas. The pathological characteristic of cardiac hemangioma is the abnormal proliferation of small vessels. When abnormal blood supply or local blood flow increase is found in the mass, it often has auxiliary diagnostic value. Echocardiography has become the preferred examination method owing to its high accuracy and noninvasiveness. Echocardiography can dynamically observe the activity of hemangiomas, which is not possible with other examination methods. Among the 34 patients included, 33 underwent transthoracic or transesophageal echocardiography to evaluate the mass; 63.6% underwent transthoracic echocardiography (n = 21), 12.1% underwent transesophageal echocardiography (n = 4), and 24.2% underwent two both examinations (n = 8). In 1 case with a small mass, no hemangioma was found on transesophageal echocardiography. Although cardiac ultrasound can accurately identify cardiac tumors, there is lack of specificity regarding determining the type of cardiac tumor, and small lumps may not be found in time. Therefore, after cardiac ultrasound identifies an abnormal lump, further checks and

Table 1. Summary of the 34 case reports of cardiac hemangioma in PubMed.

No.	First author	Year	Age/Sex	Clinical symptoms	Size (cm)	Location	Treatment	Follow up time
1	Yasir Abu-Omar [4]	2010	46 y/M	Ventricular Tachycardia	3.7 × 7.0	LV	Resection + Radiofrequency ablation	NA
2	Jayendra Sharma [5]	2011	12 y/F	Dyspnea, Fatigue	3.0 × 4.6	Right ventricular outflow tract	Resection	Asymptomatic for 2 years
3	Zain Husain [6]	2011	42 y/F	Dyspnea, Palpitation	11.0 × 6.5 × 4.5	RA	Resection	NA
4	Asako Takahashi [7]	2013	61 y/F	Dyspnea	5.3 × 6.0	LA	Resection	NA
5	Sean D. Galvin [8]	2012	83 y/M	Dyspnea	5.0 × 3.5 × 4.0	RA	Resection	Asymptomatic for 1 year
6	Satoshi Numata [9]	2013	55 y/M	Asymptomatic	1.5 × 1.2 × 1.0	RV	Resection	NA
7	Yuna Han [10]	2014	61 y/NA	Palpitation, Fatigue	1.4 × 1.1 × 0.7	LV	Resection	NA
8	Filip M. Szymanski [11]	2013	51 y/F	Asymptomatic	7.5 × 6.5	RA	Partial resection	Asymptomatic for 1 year
9	Z.S. Jonjev [12]	2013	72 y/F	Transient Cerebral Ischemia	2.5 × 1.7	LA	Resection	Asymptomatic for 6 months
10	Yihua Liu [13]	2014	53 y/M	Asymptomatic	0.74 × 0.99	LV	Resection	NA
11	Wen-Jian Jiang [14]	2014	49 y/M	Asymptomatic	6.8 × 5.0 × 2.6	RV	Resection	Asymptomatic for 6 months
12	Eric Unger [15]	2015	71 y/M	Asymptomatic	8.0 × 6.5 × 4.8	LA	Resection	NA
13	Soji Nishio [16]	2015	35 y/F	Anasarca, Pericardial Effusion	Unknow	LA	Pericardial puncture + Resection	NA
14	Yanqiu Wang [17]	2017	42 y/M	Palpitation	1.5 × 1.0 × 1.0	LV	Resection	Asymptomatic for 1 year
15	Marie L. Shaner [18]	2017	15 y/M	Chest Pain	3.0 × 2.0	RV	Resection	Asymptomatic for 6 months
16	Sara R. Vacirca [19]	2018	41 y/F	Asymptomatic	1.2 × 1.2/0.2 × 0.2	RV	Resection	Asymptomatic for 1 year
17	Jian Xu [20]	2019	56 y/F	Asymptomatic	7.0 × 4.0 × 3.0	LA	Resection	Asymptomatic for 6 months
18	Huikai Miao [21]	2019	71 y/M	Palpitation	4.3 × 3.2 × 2.6	RA	Resection + Atrial septal repair	Asymptomatic for 2 years
19	C. J. Perez Rivera [22]	2019	48 y/F	Syncopal Episodes	28 × 3.5	RA + RV	Resection	Asymptomatic for 1 year
20	Eisaku Nakamura [23]	2019	82 y/F	Dyspnea, Lower Limb Edema	8.2 × 8.0	LA	Resection + Mitral valve replacement + Tricuspid Valve Annuloplasty	NA
21	Çağdaş Topel [24]	2021	24 y/F	Palpitation, Syncopal	3.0 × 1.5 × 1.3	LA	Partial resection + Radiofrequency Ablation	Asymptomatic for 1 year
22	Zhenxing Sun [25]	2020	35 y/M	Palpitation	6.0 × 3.0	LV	Resection	Asymptomatic for 1 year
23	Keita Kamata [26]	2021	69 y/M	Asymptomatic	4.2 × 4.0 × 3.8	RV	Resection + Radiofrequency Ablation	NA
24	Takuya Fujita [27]	2022	76 y/F	Dyspnea, Pericardial Effusion	2.0 × 2.0	Left Atrial Appendage (Pericardial cavity)	Thoracoscopic Pericardial Fenestration + Resection	Asymptomatic for 6 years
25	Thu Thuy Vu [28]	2022	71 y/M	Asymptomatic	1.3 × 1.7 × 1.4	RV	Resection	NA

Table 1. Continued.

No.	First author	Year	Age/Sex	Clinical symptoms	Size (cm)	Location	Treatment	Follow up time
26	Jinlan Chen [29]	2022	10 m/F	Pneumonia	3.5 × 1.2 × 0.9	RV	Aspiration biopsy	Asymptomatic for 2 years
27	Yulin Wen [30]	2022	53 y/M	Dyspnea, Chest Pain	5.0 × 3.0 × 2.0	RA	Resection	Asymptomatic for 6 months
28	Mihika Shah [31]	2023	23 y/F	Postpartum Chest Pain	7.0 × 6.4	RA	Aspiration biopsy + Resection	NA
29	Shouji Zhang [32]	2023	68 y/F	Dyspnea	2.5 × 3.0 × 3.0	Atrial septum of left atrium	Resection	Asymptomatic for 3 months
30	Ting Xie [33]	2023	53 y/M	Dyspnea, History of Rheumatoid Arthritis	2.0 × 1.5/1.5 × 1.3	RA	Resection + Mitral valve replacement	Asymptomatic for 6 months
31	Jakub Batko [34]	2023	50 y/F	Palpitation	2.0 × 1.8	Atrioventricular node localization	Resection + Atrial septal repair	Asymptomatic for 1 month
32	Emeka B. Kesieme [35]	2023	49 y/F	Dizzy, Palpitation	1.2 × 1.4/0.8 × 0.6	RV	Resection + Tricuspid valvuloplasty	Asymptomatic for 1 year
33	Lilly Ilcheva [36]	2024	64 y/F	Palpitation	1.8 × 1.3	Left ventricular septum	Resection	Asymptomatic for 1 year
34	Novica Kalinic [37]	2024	52 y/F	Fatigue	3.5 × 3.1	RA	Resection	NA

F, female; M, male; y, years; m, months; NA, not available; RA, right atrium; LA, light atrium; RV, right ventricle; LV, left ventricle.

evaluation are needed by combining the cardiac ultrasound finding with other examinations. Chest CT or coronary CT angiography can also be used to effectively determine the location, size, surrounding tissues, and blood supply of the mass [39]. These diagnostic methods are especially helpful for examining small blood vessels in the mass, and have unique advantages for the diagnosis of tumor type. Fourteen patients underwent preoperative chest CT scan, with 85.7% having an abnormal mass (n = 12); meanwhile, 7 patients underwent preoperative coronary CT angiography, 71.4% of whom had abnormal masses (n = 5), including 1 case with an abnormal donor vessel and 2 cases with abnormal contrast agent aggregation. Cardiac magnetic resonance imaging (MRI) can also effectively evaluate the size, location, and surrounding tissue involvement of hemangiomas. Typically, T1-weighted images show moderate-intensity signals while T2-weighted images show high-intensity signals [40]. Ten patients underwent preoperative cardiac MRI, 60% of whom showed typical T1 isointensity and T2 hyperintensity signals (n = 6), 20% showed isointense signals (n = 2) on T1- and T2-weighted images, and 10% showed hyperintense signals (n = 1) on T1- and T2-weighted images. Coronary angiography is primarily used to evaluate tumor blood flow distribution and the presence of abnormal blood supply. "Tumor blush" is a typical manifestation of a cardiac hemangioma. Twelve patients underwent coronary angiography, 25% of whom were found to have an abnormal coronary blood supply (n = 3) while 33.3% showed the typical "tumor blush" (n = 4).

Currently, the indications for surgical removal of cardiac hemangiomas remain inconclusive [3]. Owing to the possibility of sudden death, pericardial tamponade, and rupture, most researchers have suggested that active treatment should be performed. Particularly, for hemangiomas involving the septum (atrial septum and within the ventricular septum), adoption of a positive attitude toward surgical treatment is necessary, as the mass may lead to malignant arrhythmia or even sudden death. Complete resection of the tumor is the best treatment; however, intramural localization of the mass and the thickness of the surrounding myocardium pose challenges in the process, often resulting in the inability to completely remove the tumor, and ultimately, only partial resection. Among the 34 patients, 1 patient was young and was followed-up regularly after biopsy, without surgical treatment. The remaining 33 patients underwent surgical resection, of which 1 underwent partial resection due to extensive tumor invasion while the rest underwent complete tumor resection. Although the patient in this case had no obvious clinical manifestations, the hemangioma was located in the atrial septum, close to the inferior vena cava, and adjacent to the atrioventricular node. Had the hemangioma further increased in size or invaded nearby structures, it may have led to malignant arrhythmic events; therefore, surgical treatment was performed. Fortunately, the hemangioma capsule in this patient was intact and was completely removed. Postoperative pathological examination confirmed a cavernous hemangioma, and the patient recovered well without complications.

Among the 34 patients, 20 had clear postoperative follow-up records; the longest follow-up time was 6 years, with no reports of recurrence or complications. Similarly, our patient recovered well after surgery. The patient was followed up regularly at 1 month, 3 months, 6 months, and 1 year after the operation, and echocardiography and chest X-ray were performed. No recurrence or complications occurred during the follow-up period. Given the limited experience of clinicians in the diagnosis and treatment of cardiac hemangiomas, we report this case and conduct a literature review to provide a more comprehensive approach to the diagnosis and treatment of this disease.

Conclusion

Cardiac hemangiomas are rare and can occur at any age. Atrial hemangiomas show no typical clinical symptoms. The symptoms, if any, are related to the location or size of the tumor. Echocardiography, MRI, and coronary angiography are helpful in the diagnosis of this disease; however, the final diagnosis will still be based on the histopathological examination. This highlights the importance of multidisciplinary diagnosis. For patients with obvious clinical symptoms, the treatment method should be selected according to the patient's physical condition, anatomical location of the lesion, and other factors; in such cases, surgery is the primary treatment option. The optimal treatment for asymptomatic patients with atrial hemangiomas is yet to be determined. Some scholars advocate the use of corticosteroids, radiotherapy, and β-receptor blockers for the treatment of asymptomatic hemangioma; however, the efficacy of these treatment methods remains controversial, as sufficient evidence and controlled studies supporting them are lacking. In view of the potential impact of cardiac hemangiomas on heart rhythm and circulation, surgery is recommended as soon as possible, if there are no contraindications.

Availability of Data and Materials

Relevant data supporting the findings of this study are available from the authors upon request.

Author Contributions

CS and SZ designed and performed the study. GL and WD were responsible for the data analysis and writing of the manuscript. Methodological support was provided by CS. SY and CC helped gather information cases, read the

full text and provided comments. All authors read and approved the final manuscript. All authors have made a great contribution to the editing of the manuscript. All authors are fully involved in the work, the content of the appropriate part of the public responsibility, and agree to be responsible for all aspects of the work, to ensure that the problems associated with its accuracy or completeness.

Ethics Approval and Consent to Participate

The author has got the informed consent of published cases. This study was approved by the Human Experimentation Ethics Committee of Bengbu Medical University (protocol No. 2021170).

Acknowledgment

The authors wish to thank Dr. Shuai Zhao for his assistance in the transthoracic echocardiographic examination.

Funding

This work was supported by grants from Projects of Natural Science Research in Colleges and Universities of Anhui Province (2022AH051531), Scientific Research Project of Anhui Provincial Health Commission (AHWJ2023A30184), Bengbu Medical College university level Quality Engineering Project (2023fyjyxm21), Natural Science Projects of Bengbu Medical University (2023byzd078, 2023byzd093).

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.7771.

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