A primary cardiac schwannoma of the right ventricle: a case report and literature review

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Abstract

Background: Primary cardiac schwannoma remains extremely rare and difficult to distinguish from other myocardial tumours. We report a case of cardiac schwannoma that occurred in the lateral wall of the right ventricle and grew in the myocardial walls. It is the third case of schwannoma that occurred in the free wall of the right ventricle. Moreover, we reviewed and summarised the literature for cases involving benign cardiac schwannomas.

Case presentation: We present a case of a 64-year-old woman who presented to our centre with syncope for 1–2 min. Echocardiogram and contrast-enhanced computed tomography subsequently revealed a 2.9 × 1.9 cm homogeneous mass originating from the anterior wall of the right ventricle. The patient underwent thoracotomy to resect the mass, which was pathologically verified as Schwann cell tumour.

Conclusions: This is a rare case added to the limited existing literature on cardiac schwannoma. Comprehensive analysis of various imaging examinations is helpful to determine the extent of the tumour. Complete surgical resection is recommended for similar cases involving cardiac schwannomas, especially when the patient has related symptoms. Patients generally have a good prognosis. The pathogenesis of cardiac schwannoma needs further research in order to prevent and manage this rare lesion.

Keywords: Cardiac tumour, Schwannoma, Case report

Background

Primary cardiac tumours are extremely rare tumours with a prevalence of 0.02%–0.056% [1], and cardiac Schwann cell tumours are even rarer. Schwannoma is a slow-growing tumour that arises from Schwann cells in the surrounding nerve sheath [2]. Primary cardiac schwannoma is believed to originate from the cardiac plexus or the cardiac branch of the vagus nerve [3]; but its pathogenesis remains unclear. We explored the pathogenesis of cardiac schwannomas that have not been explored in detail in previous literature. Cardiac schwannoma has a variety of clinical manifestations, ranging from asymptomatic findings on imaging studies to exertion, chest pain, tachypnoea and arrhythmia, which are related to tumour size and compression of adjacent structures (e.g. large vessels, cardiac chambers, mediastinal structure and coronary artery [4]). Preoperative diagnosis is difficult, but the identification of such tumours is of great value in the development of treatment strategies and prognostic assessment. In this report, we present a case of a cardiac schwannoma, which is the third case of schwannoma that occurred in the free wall of the right ventricle. In addition, we reviewed and summarised cardiac schwannomas, which have been reported in the English literature. This is the most detailed summary and discussion of cardiac schwannoma in the past 18 years (Table 1).
| No | Author et al. | Year | Age(years) | Sex(M/F) | Location | Symptoms | Size(cm) | Imaging findings | Treatment | Survival and prognosis | Follow-up time (mouth) | Comorbidities |
|----|---------------|------|------------|----------|----------|----------|----------|------------------|-----------|----------------------|------------------------|--------------|
| 1  | Hallman et al. [5] | 1966 | 12 | F | Anterior RV surface near AV groove | Easy fatigability, dyspnea on exertion | 3 | Unknown | Unknown | Unknown | Unknown | Unknown |
| 2  | Gleason et al. [5] | 1972 | 26 | F | RA near its junction with IAS, 2 cm below inlet of SVC | Heart murmur, incidental finding at operation (ASD, PS) | 1.5 × 1.5 | Unknown | Unknown | No recurrence | Unknown | Unknown |
| 3  | Stephen Factor et al. [6] | 1976 | 55 | F | Lateral border of RA, superior to AV groove | Incidental finding at autopsy | 7 × 5.5 × 5 | – | – | – | – | Crystadenocarcinoma of the ovary, hypertension, intermittent intestinal obstruction |
| 4  | Betancourt et al. [7] | 1979 | 32 | F | Intracavitary tumor attached to parietal band of crista | Chest pain, shortness of breath | 8.75 × 6.25 | Marked cardiomegaly on chest X-ray | Surgery with CPB | No recurrence | 36 | Hypertension |
| 5  | Monroe et al. [5] | 1984 | 70 | M | Anterior LV surface, below AV groove | Incidental finding at autopsy | 7 | Unknown | – | – | – | Lung cancer |
| 6  | Andrew D. Forbes et al. [8] | 1994 | 35 | M | Posterior LA between inferior PV and CS | Exertion-related paroxysmal atrial fibrillation, atypical chest pain | 4 × 7 | Unknown | Median sternotomy + CPB | No recurrence | 6 | None |
| 7  | Kodama et al. [5] | 1995 | 50 | M | Anterior RA, superior to AV groove | Exertional dyspnea | 9 × 5 × 6 | Unknown | Unknown | Unknown | Unknown | Unknown |
| 8  | Hashimoto T et al. [9] | 1998 | 46 | F | Between SVC and ascending Ao | None | 12 × 8 × 7 | Cardiomegaly on chest radiograph | Median sternotomy + CPB | No recurrence | 24 | Uterine fibroids |
| 9  | Bizzarri et al. [3] | 2001 | 72 | M | Intracavitary tumor attached to floor of RA, close to AV | Shortness of breath, chest pain | 5 × 4 × 4 | Unknown | Surgery with CPB | Recovered quickly | None | Mild hypertension, right renal adenocarcinoma |
| 10 | Mustafa Sirılık et al. [10] | 2003 | 61 | F | LA | Shortness of breath, atrial fibrillation with a 10-year duration | 95 × 8.5 × 6.5 | Heterogeneous and hypodense mass with central cystic foci | Median sternotomy + CPB | Remains well and disease-free | 2 | None |
| No | Author                          | Year | Age(years) | Sex(M/F) | Location | Symptoms                                                                 | Size(cm)               | Imaging findings                                      | Treatment                                      | Survival and prognosis | Follow-up time (mouth) | Comorbidities                                                                 |
|----|--------------------------------|------|------------|----------|----------|--------------------------------------------------------------------------|------------------------|------------------------------------------------------|-----------------------------------------------|----------------------------|------------------------|--------------------------------------------------------------------------------|
| 11 | Kunihide Nakamura et al. [5]   | 2003 | 33         | F        | Anterior RA extending LA and PV | None                                      | 5 × 5.2 × 4.5         | Inhomogeneous enhancement                           | Median sternotomy + CPB                      | No recurrence              | 12                     | None                                                                            |
| 12 | Davinder S. Jaswal et al. [11] | 2003 | 49         | F        | RA adjacent to the AV groove | Mitral valve prolapse presented with pleuritic chest discomfort | 6.4 × 5.5 × 3.4       | Large heterogeneous mass                             | Surgery with CPB                             | Unknown                    | Unknown                | None                                                                            |
| 13 | Xiao-dong Chen et al. [12]     | 2005 | 51         | F        | RA       | Dizziness, tinnitus, gait instability                                      | 10.2 × 10              | Heterogeneous mass with calcifications and Cystic structure inside      | Median sternotomy + CPB                      | Unknown                    | Unknown                | Bilateral acoustic neuroma, type II neurofibromatosis                           |
| 14 | T. Rausche et al. [13]         | 2006 | 42         | F        | RV epicardium                   | Persistent coughing                       | 11 × 7                | Mass with areas of different echodensities           | Median sternotomy                          | Completely inconspicuous     | Unknown                | None                                                                            |
| 15 | Noedir A.G. Stolf et al. [14]  | 2006 | 56         | F        | RA close to the cavo-atrial junction | None                                    | 60 × 4.8               | Heterogeneous solid tumoral mass with calcifications inside | Surgery with CPB                             | No recurrence              | 36                     | Cavernous mass of the bladder                                                  |
| 16 | Sedir Sevimli et al. [15]      | 2007 | 57         | F        | The free wall of the LV         | Palpitations                              | 5.5 × 6               | Containing cystic structures                           | Surgery with CPB                             | Remained disease free        | 3                      | None                                                                            |
| 17 | Saverio La Francesca et al. [4] | 2007 | 30         | F        | Anterior and lateral surface of the superior half of the LV                | None                                    | 4 × 4 × 9               | A large multilobed cardiac mass                        | CPB + coronary reconstruction + thrombectomy + LVAD + anti-coagulation | Discharged home on postoperative day 28 | None                  | Cancer of the left chest wall                                                  |
| 18 | Sarah A Early et al. [16]      | 2007 | 57         | M        | Posterolateral wall of the RA extending to the interatrial septum          | No cardiovascular symptoms                | 4.3 × 5.2              | Heterogenous very mild enhancement on MRI             | Surgery with CPB                             | Excellent                  | Unknown                | Gastritis, normochromic normocytic anaemia                                     |
| 19 | Corey D et al. [17]            | 2011 | 67         | M        | RA involving the interatrial septum                                         | Dysspnea on exertion and syncope          | 3.1 × 2.5 × 1.7        | Intraoperative transesophageal echocardiogram revealed a cystic mass | Surgery with antegrade cardiopexia           | Do well after his surgery with no symptoms | 9                      | Severe aortic stenosis                                                          |
| 20 | Kristen Elsner et al. [18]     | 2013 | 65         | M        | Lateral wall of the LPA                                                   | Dyspnea on exertion                       | 5.2 × 4.5 × 4.1        | Heterogeneous enhancement                            | Median sternotomy + CPB + CABG             | No signs or symptoms          | Unknown                | None                                                                            |
### Table 1 (continued)

| No | Author | Year | Age(years) | Sex(M/F) | Location | Symptoms | Size(cm) | Imaging findings | Treatment | Survival and prognosis | Follow-up time (mouth) | Comorbidities |
|----|--------|------|------------|----------|----------|----------|----------|------------------|-----------|------------------------|-----------------------|--------------|
| 21 | Su Kyung Hwang et al. [19] | 2014 | 55 | F | LA, attached to the left atrial appendage | Chest pain at rest | 4.3 × 4 × 3 | Mass with hemorrhagic formation and a pericardial tail | Median sternotomy + CPB | No remnant mass | 12 | None |
| 22 | Kuk Hui Son et al. [20] | 2015 | 42 | F | Atrial roof between the aorta and the SVC | Palpitations on several occasions | 10 × 9.5 | Heterogeneously enhanced | Sternotomy + CPB + 3D printing model | Discharged without relevant complication | None | None |
| 23 | Joon Chul Jung et al. [21] | 2015 | 69 | F | Interatrial septum | None | 2.8 × 2.7 × 2.5 | Cystic mass, broad base | Median sternotomy | Recovered without problems | Unknown | Sigmoid colon cancer |
| 24 | Ji-Gang Wang et al. [22] | 2018 | 59 | M | RA, attached to the underpart of interatrial septum | None | 45 × 3.5 × 3 | Unknown | Unknown | No recurrence | Unknown | None |
| 25 | Zhixiong Huang et al. [23] | 2020 | 53 | M | Behind the ascending Ao | Dyspnea on exertion, hypertension | 82 × 7.1 × 6.9 | Cystic low density mass | Median sternotomy + CPB | No recurrence | 60 | Hypertension |
| 26 | Kenji Yokoyama et al. [5] | 2021 | 46 | M | Posterior wall of the LA | None | 1.4 × 1.6 | Multiple lesions (posterior mediastinum, left pulmonary hilar area) | Median sternotomy + CPB | No recurrence | 12 | Type II neurofibromatosis |
| 27 | Wang SY et al. [24] | 2021 | 65 | F | RA, adjacent to atrial septum | Shortness of breath after activity | – | Apparent FDG uptake in the mass, SUV-max 5.2 | – | Unknown | Unknown | Unknown |
| 28 | Present case | 2021 | 64 | F | Anterior RV | None | 2.8 × 2.0 | Shallowly divided, homogeneous | Median sternotomy + CPB | No recurrence | 60 | Lung adenocarcinoma |

RV: Right ventricle, AV: Atrioventricular, RA: Right atrium, IAS: Interatrial septum, SVC: Superior vena cava, ASD: Atrial septal defect, PS: Pulmonary stenosis, CPB: Cardiopulmonary bypass, LV: Left ventricle, LA: Left atrium, PV: Pulmonary vein, CS: Coronary sinus, LVAD: CentriMag left ventricular assist device, MRI: Magnetic resonance imaging, LMA: Left main pulmonary artery, CABG: Coronary artery bypass surgery, Ao: Aorta
Case presentation

A 64-year-old woman was admitted to our institute with syncope for 1–2 min. She reported no shortness of breath, chest pain, dyspnoea or weight loss. Her medical history included lacunar infarction and ground glass nodule of the left upper lobe, which was suspected to be lung adenocarcinoma. On physical examination, the patient was afebrile and had a regular heart rate of 60 beats per minute, a blood pressure of 125/70 mmHg and a respiratory rate of 16 breaths per minute. The patient had no murmur. The results of laboratory investigations were unremarkable. Electrocardiogram revealed sinus bradycardia and a ventricular rate of 57 beats per minute.

Echocardiogram revealed a mass with a size of $2.9 \times 1.9$ cm on the frontal wall of the right ventricle, which had a uniform internal echo and star-shaped blood flow signals (Fig. 1). Chest computed tomography (CT) scan demonstrated a $2.8 \times 2.0$ cm homogeneous mass originating from the anterior wall of the right ventricle, which has a relatively broad base. The boundary of the mass is clear and fixed. No obvious narrowing of the heart cavity was observed (Fig. 2A). The patient underwent a three-phase dynamic chest CT, which disclosed a myocardial tissue mass with slightly enhancement during the arterial phase (Fig. 2B) and a persistent moderate increase during the venous and delayed phases (Fig. 2C-D). The definite diagnosis was difficult. The patient was referred to thoracic
surgery for thoracotomy and resection of the myocardial tumour under cardiopulmonary bypass (CPB). The defect of the right ventricle was repaired. The desquamated tissue was histopathologically examined and reported as a schwannoma. Microscopic sections revealed that Cells of Antoni A tissue have modest eosinophilic cytoplasm with discernible cell borders and normochromic, elongated, tapered nuclei (Fig. 3A). Immunohistochemical studies showed that the tumour cells stained positively for S100 and SOX10 (Fig. 3B-C). Micrographs were acquired by using Nikon CI2 (Nikon) and NIS-Elements (Nikon) software. The resolution of each acquired image is 300 dots per inch.

The patient recovered and was discharged on the 10th day after surgery without complications. The patient had a ground glass nodule resection, which was subsequently confirmed as microinvasive lung adenocarcinoma, and the pathologic TNM stage was T1N0M0. The patient recovered uneventfully and had no sign of recurrence at a follow-up duration of 5 years.

Discussion and conclusion

We searched the PubMed database until July 21 2022 using the keywords “Cardiac Schwannoma” and “Cardiac tumour and Schwannoma” to identify the relevant English medical literature. The search identified 332 results. After a careful analysis of the articles, approximately 24 articles met the inclusion criteria and were included. In addition, 3 patients dates from one of the abovementioned paper [5], which could not be retrieved from PubMed, were also added to this review. The study selection process is shown in Fig. 4. Two reviewers independently appraised all included studies using the Joanna Briggs Institute (JBI) checklist for case reports and case series.

Primary cardiac tumours are very rare, and benign cardiac schwannomas are even rarer. Our review of English literature showed that 27 cases of benign cardiac schwannomas including two cases of type II neurofibromatosis [12, 25] have been reported. The age range was 12–72 years old, the mean age was 50.7 years old, and the male-to-female ratio was about 1:2. These data were consistent with previous reports [5]. Primary cardiac schwannoma is believed to originate from the cardiac plexus or the cardiac branch of the vagus nerve; therefore, it is located primarily on the right side of the heart [26]. However, we found that the right atrium is the predominant site of cardiac schwannomas (12/28), and the incidences of left atrial, bilateral ventricular and aortic outflow tracts have no remarkable differences. This finding could be attributed to the distribution of the sinoatrial and atrioventricular nodes around the right atrium because the distribution of nerve fibres around these structures is remarkably higher than that in the surrounding working myocardium. The case that we reported occurred in the lateral wall of the right ventricle. It is the third case of schwannoma that occurred in the free wall of the right ventricle. This study provides an important supplement to explore the pathogenesis of the lesion and reflect the distribution of cardiac plexus.

Neurilemoma originates from the peripheral nerve sheath, and its pathogenesis remains unclear. No relevant literature has proposed hypotheses regarding its cause. The National Toxicology Program and Ramazzini Institute reported that radiofrequency electromagnetic field substantially increases glioma and schwannoma in the heart of rodents [27]. Stephen Factor et al. reported that a patient with cardiac neurilemoma who received a large total amount of radiotherapy or at least one course of radiotherapy directed to the lower thoracic vertebral region for the treatment of paravertebral mass may have peripherally involved the heart [6]. The relationship between human cardiac schwannoma and radiation...
needs further research. Additionally, Das Gupta et al. studied 303 benign schwannomas and reported the interesting correlation of nerve sheath tumours with the past, concurrent or future development of a malignancy unrelated to peripheral nerves [28]. Through case review, we found that seven cases, including our case, were accompanied by other tumours, including six cases of malignancy and one unspecified case. The seven tumours included one autopsies case of ovarian cancer [6] and one autopsies cases of lung cancer [5], one case of renal cancer preceded cardiac schwannoma [3], one case of synchronous sigmoid colon cancer [21], one case of synchronous cancer of the left chest wall [4] and one case of synchronous cavernous mass of the bladder [14], one cases of synchronous lung adenocarcinoma (our case). The connection between schwannoma and other unrelated malignancy needs further experimental verification.

Primary cardiac schwannomas vary in size. The clinical symptoms are mostly caused by compression or obstruction, and some patients may have dyspnoea on exertion (5/28) [12, 17, 18, 23], chest pain (4/28) [3, 7, 8, 19], shortness of breath (4/28) [3, 7, 10, 24], palpitation (2/28) [15, 20], arrhythmia (2/28) [8, 10] and other discomfort. More than one third of patients (10/28) [4–6, 9, 14, 16, 21–23] had no related symptoms. Our case was hospitalised because of syncope, which is rarely reported in literature. The syncope may be caused by the sudden decrease or pause of cardiac output caused by the cardiac tumour.

Cardiac schwannoma can be detected by X-ray or echocardiogram, CT and magnetic resonance imaging (MRI), which can help to better determine the location and extent of the mass and the involvement of other structures [3]. Tumours are mostly heterogeneous masses with cystic changes, haemorrhages and calcifications. Uneven and mild enhancement may even occur. Some lesions have a broad base and shallow lobes, and most lesions have a clear boundary. The fibrous capsule is also one of the identification points of schwannomas from other tumours. Coronary angiography is required for patients at risk of coronary heart disease or with tumours that may involve the coronary
artery [4]. When the exact origin of the tumour cannot be obtained by CT or MRI, 3D printing and model establishment can help to clearly identify the location of the tumour and its relationship with large blood vessels [20]. The nature of the tumour is difficult to identify through imaging.

Most patients with cardiac Schwann cell tumours undergo extensive radical tumour resection and cardiac reconstruction with autologous pericardium or artificial patch under CPB [5]. The degree of involvement and reconstruction of the atrioventricular valve, coronary artery, coronary sinus or pulmonary vein are also important [4, 5, 13]. Among the 28 patients, excepting for 2 autopsy patients, 2 patients whose survival/death was not mentioned in the literature, and 1 patient whose data was not available, the survival rate of the remaining 23 patients was 100% in the follow-up period, and the postoperative prognosis is good. Our operation was also successful, and no recurrence was observed after 5 years of follow-up.

In conclusion, this is a rare case added to the limited existing literature on cardiac schwannoma. Comprehensive analysis of various imaging examinations is helpful to determine the extent of the tumour. Complete surgical resection is recommended for similar cases involving cardiac schwannomas, especially when the patient has related symptoms. Patients generally have a good prognosis. The pathogenesis of cardiac schwannoma needs further research in order to prevent and manage this rare lesion.

Abbreviations
CT: Computed tomography; CPB: Cardiopulmonary bypass; MRI: Magnetic resonance imaging.

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Authors’ contributions
WF performed data analyses and wrote the manuscript. WF, LL, MH and CXX conducted the clinical diagnosis and data collection. All authors read and approved the final manuscript.

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Availability of data and materials
All data generated or analysed during this study are included in this published article and its supplementary information files.

Declarations
Ethics approval and consent to participate
This study was approved by the Medical Ethics Committee of Yantai Yuhuangding Hospital.

Consent for publication
Written informed consent was obtained from the patient for the publication of this report and any accompanying images. A copy of the written consent is available for review by the Editor at any time.

Competing interests
The authors declare no conflict of interest.

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