

World Journal of *Clinical Cases*

World J Clin Cases 2020 October 6; 8(19): 4280-4687



Contents

Semimonthly Volume 8 Number 19 October 6, 2020

OPINION REVIEW

- 4280 Role of monoclonal antibody drugs in the treatment of COVID-19
Ucciferri C, Vecchiet J, Falasca K

MINIREVIEWS

- 4286 Review of simulation model for education of point-of-care ultrasound using easy-to-make tools
Shin KC, Ha YR, Lee SJ, Ahn JH
- 4303 Liver injury in COVID-19: A minireview
Zhao JN, Fan Y, Wu SD

ORIGINAL ARTICLE

Case Control Study

- 4311 Transanal minimally invasive surgery *vs* endoscopic mucosal resection for rectal benign tumors and rectal carcinoids: A retrospective analysis
Shen JM, Zhao JY, Ye T, Gong LF, Wang HP, Chen WJ, Cai YK
- 4320 Impact of *mTOR* gene polymorphisms and gene-tea interaction on susceptibility to tuberculosis
Wang M, Ma SJ, Wu XY, Zhang X, Abesig J, Xiao ZH, Huang X, Yan HP, Wang J, Chen MS, Tan HZ

Retrospective Cohort Study

- 4331 Establishment and validation of a nomogram to predict the risk of ovarian metastasis in gastric cancer: Based on a large cohort
Li SQ, Zhang KC, Li JY, Liang WQ, Gao YH, Qiao Z, Xi HQ, Chen L

Retrospective Study

- 4342 Predictive factors for early clinical response in community-onset *Escherichia coli* urinary tract infection and effects of initial antibiotic treatment on early clinical response
Kim YJ, Lee JM, Lee JH
- 4349 Managing acute appendicitis during the COVID-19 pandemic in Jiaying, China
Zhou Y, Cen LS
- 4360 Clinical application of combined detection of SARS-CoV-2-specific antibody and nucleic acid
Meng QB, Peng JJ, Wei X, Yang JY, Li PC, Qu ZW, Xiong YF, Wu GJ, Hu ZM, Yu JC, Su W
- 4370 Prolonged prothrombin time at admission predicts poor clinical outcome in COVID-19 patients
Wang L, He WB, Yu XM, Hu DL, Jiang H

- 4380** Percutaneous radiofrequency ablation is superior to hepatic resection in patients with small hepatocellular carcinoma

Zhang YH, Su B, Sun P, Li RM, Peng XC, Cai J

- 4388** Clinical study on the surgical treatment of atypical Lisfranc joint complex injury

Li X, Jia LS, Li A, Xie X, Cui J, Li GL

- 4400** Application of medial column classification in treatment of intra-articular calcaneal fractures

Zheng G, Xia F, Yang S, Cui J

Clinical Trials Study

- 4410** Optimal hang time of enteral formula at standard room temperature and high temperature

Lakananurak N, Nalinthassanai N, Suansawang W, Panarat P

META-ANALYSIS

- 4416** Meta-analysis reveals an association between acute pancreatitis and the risk of pancreatic cancer

Liu J, Wang Y, Yu Y

SCIENTOMETRICS

- 4431** Global analysis of daily new COVID-19 cases reveals many static-phase countries including the United States potentially with unstoppable epidemic

Long C, Fu XM, Fu ZF

CASE REPORT

- 4443** Left atrial appendage aneurysm: A case report

Belov DV, Moskalev VI, Garbuzenko DV, Arefyev NO

- 4450** Twenty-year survival after iterative surgery for metastatic renal cell carcinoma: A case report and review of literature

De Raffele E, Mirarchi M, Casadei R, Ricci C, Brunocilla E, Minni F

- 4466** Primary rhabdomyosarcoma: An extremely rare and aggressive variant of male breast cancer

Satală CB, Jung I, Bara TJ, Simu P, Simu I, Vlad M, Szodorai R, Gurzu S

- 4475** Bladder stones in a closed diverticulum caused by *Schistosoma mansoni*: A case report

Alkhamees MA

- 4481** Cutaneous ciliated cyst on the anterior neck in young women: A case report

Kim YH, Lee J

- 4488** Extremely rare case of successful treatment of metastatic ovarian undifferentiated carcinoma with high-dose combination cytotoxic chemotherapy: A case report

Kim HB, Lee HJ, Hong R, Park SG

- 4494** Acute amnesia during pregnancy due to bilateral fornix infarction: A case report
Cho MJ, Shin DI, Han MK, Yum KS
- 4499** Ascaris-mimicking common bile duct stone: A case report
Choi SY, Jo HE, Lee YN, Lee JE, Lee MH, Lim S, Yi BH
- 4505** Eight-year follow-up of locally advanced lymphoepithelioma-like carcinoma at upper urinary tract: A case report
Yang CH, Weng WC, Lin YS, Huang LH, Lu CH, Hsu CY, Ou YC, Tung MC
- 4512** Spontaneous resolution of idiopathic intestinal obstruction after pneumonia: A case report
Zhang BQ, Dai XY, Ye QY, Chang L, Wang ZW, Li XQ, Li YN
- 4521** Successful pregnancy after protective hemodialysis for chronic kidney disease: A case report
Wang ML, He YD, Yang HX, Chen Q
- 4527** Rapid remission of refractory synovitis, acne, pustulosis, hyperostosis, and osteitis syndrome in response to the Janus kinase inhibitor tofacitinib: A case report
Li B, Li GW, Xue L, Chen YY
- 4535** Percutaneous fixation of neonatal humeral physeal fracture: A case report and review of the literature
Tan W, Wang FH, Yao JH, Wu WP, Li YB, Ji YL, Qian YP
- 4544** Severe fundus lesions induced by ocular jellyfish stings: A case report
Zheng XY, Cheng DJ, Lian LH, Zhang RT, Yu XY
- 4550** Application of ozonated water for treatment of gastro-thoracic fistula after comprehensive esophageal squamous cell carcinoma therapy: A case report
Wu DD, Hao KN, Chen XJ, Li XM, He XF
- 4558** Germinomas of the basal ganglia and thalamus: Four case reports
Huang ZC, Dong Q, Song EP, Chen ZJ, Zhang JH, Hou B, Lu ZQ, Qin F
- 4565** Gastrointestinal bleeding caused by jejunal angiosarcoma: A case report
Hui YY, Zhu LP, Yang B, Zhang ZY, Zhang YJ, Chen X, Wang BM
- 4572** High expression of squamous cell carcinoma antigen in poorly differentiated adenocarcinoma of the stomach: A case report
Wang L, Huang L, Xi L, Zhang SC, Zhang JX
- 4579** Therapy-related acute promyelocytic leukemia with FMS-like tyrosine kinase 3-internal tandem duplication mutation in solitary bone plasmacytoma: A case report
Hong LL, Sheng XF, Zhuang HF
- 4588** Metastasis of esophageal squamous cell carcinoma to the thyroid gland with widespread nodal involvement: A case report
Zhang X, Gu X, Li JG, Hu XJ

- 4595** Severe hyperlipemia-induced pseudoerythrocytosis - Implication for misdiagnosis and blood transfusion: A case report and literature review
Zhao XC, Ju B, Wei N, Ding J, Meng FJ, Zhao HG
- 4603** Novel brachytherapy drainage tube loaded with double 125I strands for hilar cholangiocarcinoma: A case report
Lei QY, Jiao DC, Han XW
- 4609** Resorption of upwardly displaced lumbar disk herniation after nonsurgical treatment: A case report
Wang Y, Liao SC, Dai GG, Jiang L
- 4615** Primary hepatic myelolipoma: A case report and review of the literature
Li KY, Wei AL, Li A
- 4624** Endoscopic palliative resection of a giant 26-cm esophageal tumor: A case report
Li Y, Guo LJ, Ma YC, Ye LS, Hu B
- 4633** Solitary hepatic lymphangioma mimicking liver malignancy: A case report and literature review
Long X, Zhang L, Cheng Q, Chen Q, Chen XP
- 4644** Intraosseous venous malformation of the maxilla after enucleation of a hemophilic pseudotumor: A case report
Cai X, Yu JJ, Tian H, Shan ZF, Liu XY, Jia J
- 4652** Intravesically instilled gemcitabine-induced lung injury in a patient with invasive urothelial carcinoma: A case report
Zhou XM, Wu C, Gu X
- 4660** Bochdalek hernia masquerading as severe acute pancreatitis during the third trimester of pregnancy: A case report
Zou YZ, Yang JP, Zhou XJ, Li K, Li XM, Song CH
- 4667** Localized primary gastric amyloidosis: Three case reports
Liu XM, Di LJ, Zhu JX, Wu XL, Li HP, Wu HC, Tuo BG
- 4676** Displacement of peritoneal end of a shunt tube to pleural cavity: A case report
Liu J, Guo M
- 4681** Parathyroid adenoma combined with a rib tumor as the primary disease: A case report
Han L, Zhu XF

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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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RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: Yan-Xia Xing; Production Department Director: Yun-Xiaojuan Wu; Editorial Office Director: Jin-Lai Wang.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Semimonthly

EDITORS-IN-CHIEF

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

October 6, 2020

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INSTRUCTIONS TO AUTHORS

<https://www.wjgnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjgnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>



Left atrial appendage aneurysm: A case report

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Author contributions: Belov DV and Moskalev VI collected the data, reviewed the literature and contributed to manuscript drafting; Garbuzenko DV contributed to manuscript drafting and revised manuscript for important intellectual content; Arefyev NO drafted the manuscript and reviewed the literature; all authors issued final approval for the version to be submitted.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors declare that they have no conflict of interest.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was selected by an in-house editor and

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Abstract

BACKGROUND

An aneurysm of the left atrial appendage is one of the rare but potentially hazardous heart defects. The risk of lethal complications grows with its size. To date, about 150 cases of this defect have been described in the literature. We present a case of left atrial appendage aneurysm with the deformation of the mitral valve and the left main coronary and circumflex artery, which required mitral valve annuloplasty and bifurcation stenting.

CASE SUMMARY

A 58-year-old man presented to our hospital complaining of shortness of breath, general weakness, dizziness during physical exertion, and fatigue. Based on the results of echocardiography, an aneurysm of the left atrium was suspected. A free-breathing real-time cine magnetic resonance imaging with electrocardiograph synchronization confirmed the diagnosis of left atrial appendage aneurysm. The patient underwent an aneurysmectomy *via* a median sternotomy with cardiopulmonary bypass. Intraoperative transesophageal echocardiography revealed relative mitral insufficiency that was corrected with an annuloplasty ring. Intraoperative coronary angiogram showed impaired blood flow in the left main coronary and circumflex artery and 60% stenosis. For this reason, bifurcation stenting was performed. The patient had an uneventful postoperative clinical course and was discharged from the hospital on the 10th day in a satisfactory condition.

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Manuscript source: Invited manuscript

Received: May 16, 2020

Peer-review started: May 16, 2020

First decision: June 7, 2020

Revised: June 8, 2020

Accepted: September 10, 2020

Article in press: September 10, 2020

Published online: October 6, 2020

P-Reviewer: Ho CM

S-Editor: Zhang L

L-Editor: A

P-Editor: Xing YX



CONCLUSION

Left atrial appendage aneurysm is a rare and dangerous heart pathology that requires surgery to prevent related complications.

Key Words: Atrial appendage; Aneurysm; Mitral valve insufficiency; Coronary stenosis; Catheter ablation; Case report

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Core Tip: Left atrial appendage aneurysm is one of the rare, but potentially hazardous heart defects. We present a case of left atrial appendage aneurysm resection and mitral valve annuloplasty. This case highlights the fact that the disease is diagnosed either by chance or when examining the patient as a result of complications. Echocardiography in most cases will make it possible to establish the diagnosis, but magnetic resonance imaging is necessary for a more detailed assessment of changes in the heart. Surgical treatment is indicated for all patients to prevent fatal disorders.

Citation: Belov DV, Moskalev VI, Garbuzenko DV, Arefyev NO. Left atrial appendage aneurysm: A case report. *World J Clin Cases* 2020; 8(19): 4443-4449

URL: <https://www.wjgnet.com/2307-8960/full/v8/i19/4443.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v8.i19.4443>

INTRODUCTION

Left atrial appendage aneurysm is one of the rare but potentially hazardous heart defects. The bigger it grows, the higher is the risk of lethal complications^[1]. The paper presents a case of left atrial appendage aneurysm that required a left atrial appendage resection and mitral valve annuloplasty.

The PubMed database, the Google Scholar retrieval system, and the reference lists from related articles were used to search for relevant publications. Articles corresponding to the aim of the review were selected for 1962 to 2019 using the keywords: "left atrial appendage aneurysm", "left atrial appendage dilatation", "pathogenesis", "diagnosis", "treatment". The management of patients with left atrial appendage aneurysm was the inclusion criterion.

CASE PRESENTATION

Chief complaints

A 58-year old man was hospitalized to the Federal Center for Cardiovascular Surgery of the Russian Ministry of Health (Chelyabinsk) complaining of dyspnea, general weakness, dizziness during physical exertion, and fatigue.

History of present illness

Over the past five years, the patient experienced rare heart palpitations that have occurred and stopped on their own after a few minutes. Six months before hospitalization, shortness of breath, weakness, and post-exertion dizziness appeared. Heart rate increased to 120 per minute. The electrocardiogram taken in an outpatient clinic revealed an irregular form of atypical atrial flutter. Based on the results of echocardiography, an aneurysm of the left atrium was suspected. Warfarin was prescribed. The maintenance dose was 2.5 mg daily.

History of past illness

The patient had a free previous medical history.

Physical examination

On the patient's visit to the Center for Cardiovascular Surgery, his vital signs were within normal limits.

Laboratory examinations

The Quick prothrombin activity was 27.9% (reference values were 70%-120%), INR was 2.52, and the prothrombin time was 45.6 s due to warfarin treatment. Other laboratory parameters of blood and urine analyses, blood biochemistry, and arterial blood gas were within normal limits.

Imaging examinations

A free-breathing real-time cine magnetic resonance imaging (MRI) with electrocardiograph synchronization did not reveal any defects in the leaves of the pericardium and extrapericardial protrusions of the heart cavities. A giant aneurysm of the left atrial appendage (123 mm × 90 mm × 70 mm in size) had uneven and clear contours, thin walls, and turbulent blood flow in the cavity. The aneurysm had a wide communication with the cavity of the left atrium. The neck of the aneurysm was 51 mm × 55 mm in size. The cavity of the aneurysm had no pathological formations, there were no blood clots. The aneurysmically dilated left atrial appendage was adjacent to the trunk and left branch of the pulmonary artery, to the upper and lower left pulmonary veins, and to the left atrioventricular sulcus. The lower contour of the aneurysm reached the diaphragmatic surface of the pericardium. The left atrium was of normal size (Figure 1). Preoperative coronary angiography showed the absence of coronary artery stenosis.

FINAL DIAGNOSIS

The final diagnosis of the presented case is left atrial appendage aneurysm.

TREATMENT

The operative approach was *via* a median sternotomy with cardiopulmonary bypass. Along the lateral wall of the left atrium, in the projection of the left atrial appendage, there was a pulsating, spherical, and irregularly shaped aneurysm of 10 cm × 8 cm in size. It was not adjacent to the pericardium and involved the posterior wall, reaching the left pulmonary veins ostia.

The intraoperative transesophageal echocardiography showed that the aneurysmal cavity was divided by a short septum and had free communication with the left atrial cavity. Relative mitral insufficiency requiring correction was also revealed.

The mitral valve was accessed through the right contour of the left atrium. The aneurysm of the left atrium was opened from the outside, and its base was resected. The wall of the aneurysm was thin, flabby, and without any blood clots.

In the area of the circumflex artery, the left ventricle was significantly deformed and had an indentation of 3.0 cm × 2.0 cm in size. The lower edge of the resected aneurysm was located near the fibrous annulus of the mitral valve, in the area of anterolateral commissure (A1 and P1 segments). The flaps of the mitral valve and chords were not damaged. Mitral valve annuloplasty was performed by using a PROFILE 3D® 680R annuloplasty ring (Medtronic, United States). Considering that the defect, which formed after excision of the aneurysm, was close to the fibrous annulus and the circumflex artery, we decided to suture it from the inside. In the area of the fibrous annulus, the suture was fixed to the outer layer of the annuloplasty ring (Figure 2). Intraoperative transesophageal echocardiography showed grade 0-1 mitral regurgitation and insignificant mitral regurgitant volume.

Considering the anatomical location of the aneurysm and the absence of visualization of the circumflex artery, we performed coronary angiography. Signs of impaired blood flow in the left main coronary and circumflex artery and 60% stenosis were found. Bifurcation stenting was performed.

A histological examination of the resected left atrial appendage revealed a 1 mm thin wall of the left atrium with hypertrophied cardiomyocytes with dystrophic changes and vacuolated cytoplasm, and slight fibrosis of the endo- and epicardium (Figure 3).

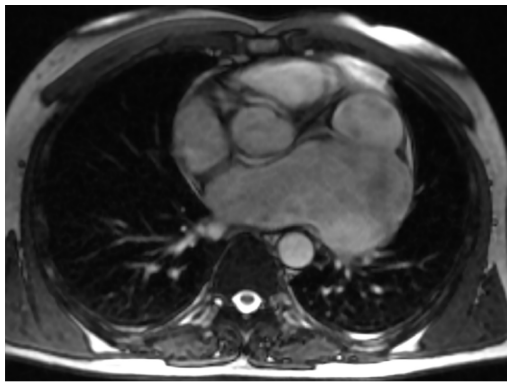


Figure 1 Magnetic resonance imaging. A white arrow shows a sharply dilated cavity of the left atrial appendage.

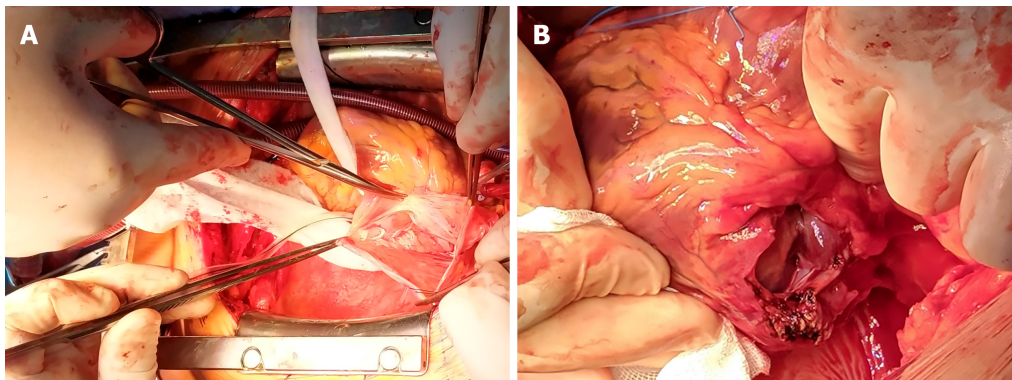


Figure 2 Aneurysmectomy. A: The aneurysm of the left atrial appendage is opened; B: The aneurysm of the left atrial appendage is excised and sutured from the inside.

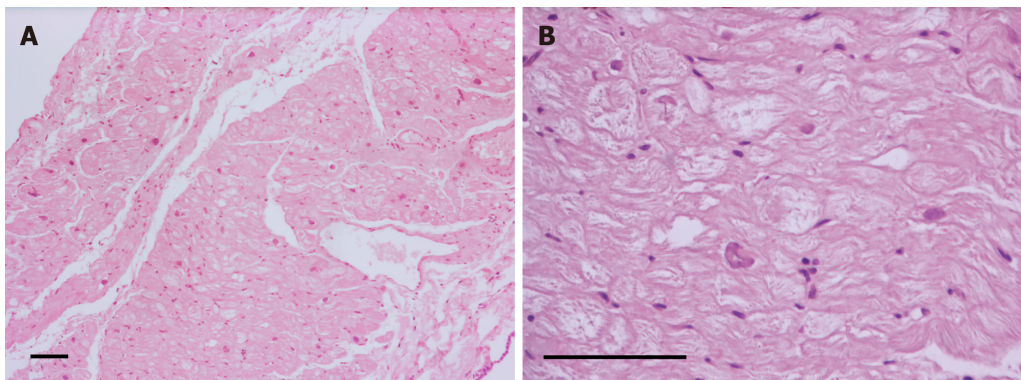


Figure 3 Hematoxylin and eosin staining of the resected left atrial appendage aneurysm. A: The wall of the aneurysm is thin, with slight fibrosis of the endo- and epicardium (scale bar 100 μ m, 100 \times magnification); B: Hypertrophied cardiomyocytes with dystrophic changes and vacuolated cytoplasm (scale bar 100 μ m, 400 \times magnification).

OUTCOME AND FOLLOW-UP

The patient had an uneventful postoperative clinical course and was discharged from the hospital on the 10th d in a satisfactory condition. Sinus rhythm restored in the early postoperative period. Atrial flutter reappeared 2 mo after discharge from the hospital and required surgical intervention; so, radiofrequency catheter ablation of the cavotricuspid isthmus was performed. Transthoracic echocardiography performed 3 mo after discharge showed a left ventricular systolic pressure of 29 mmHg and an ejection fraction of 60%, grade 0-1 mitral regurgitation, and insignificant mitral regurgitant volume. The left atrium was 43 mm \times 53 mm in size.

DISCUSSION

In 1938, Semans and Taussig reported an isolated expansion of the cavity of the left atrium in a 5-year-old girl^[2]. In 1962, Parmley described aneurysmal dilatation of the left atrium appendage in two children^[3]. The first child of 11 years old had supraventricular cardiac arrhythmia and two episodes of systemic embolism and underwent resection of the left atrial appendage and removal of the atrial septal defect at the Mayo Clinic. The second, 7-year-old patient had an aneurysm of the left atrial appendage and a concomitant congenital anomaly of the left renal artery and left kidney causing arterial hypertension. In 1963, Williams published a report on an increase in the thin-walled appendage of the left atrium in a 27-year-old man^[4]. An attempt to clamp the appendage during surgery was accompanied by cardiac arrhythmia; therefore, appendage resection had to be abandoned. However, due to embolic complications that occurred two years later, the removal of the left atrial appendage with blood clots was performed. To date, about 150 cases of this defect have been described in the literature^[5,6].

The defect has a congenital and acquired form. In the congenital form, the formation of the aneurysm occurs due to dysplasia of the scallop muscles and associated atrial muscle fibers^[5]. Poor contractile function leads to a gradual expansion and extension of the left atrial appendage as a result of increased internal pressure^[7].

Foale *et al*^[8] proposed the following diagnostic criteria for the congenital form: (1) Origin from an otherwise normal atrial chamber; (2) Well-defined communication with the atrial cavity; (3) Location within the pericardium; and (4) Distortion of the left ventricle by an aneurysm.

At the same time, Williams divided congenital aneurysms of the left atrium into extra- and intrapericardial types^[4]. In the extrapericardial type, the left atrium or appendage is prolapsed through the pericardial defect and compressed in it, which leads to the aneurysmal expansion of the extrapericardial part. The intrapericardial type is due to the underdeveloped wall of the left atrium or its appendage^[9].

The process of development of acquired aneurysms is the result of damage to the mitral valve or other conditions leading to an increase in pressure in the left atrium^[10]. At the same time, it is often difficult to differentiate the acquired and congenital forms, as in our case, since moderate changes of the mitral valve can be caused by the presence of the appendage aneurysm itself^[11].

Clinical signs of the disease occur in the third decade of life^[1]. The most common manifestations are heart palpitations (43%), shortness of breath (22%), heart rhythm disturbances (15%), embolic disorders of cerebral circulation (11%), and chest pain and discomfort (7%)^[12].

Heart rhythm disturbances occur as a result of structural remodeling of the left atrial appendage, leading to electrical dissociation of cardiomyocytes and local disturbances in conduction that favor the emergence and maintenance of arrhythmia^[13]. Left atrial appendage dilatation, atrial fibrillation, blood stasis, and thrombosis of the aneurysmal cavity are responsible for subsequent thromboembolic complications^[5].

Despite the large size of the aneurysm, our patient did not have any of the following symptoms associated with compression of surrounding structures: (1) Compression of the left ventricle by the aneurysm may cause an increase in filling pressure and diastolic dysfunction, leading to the appearance of shortness of breath and heart failure^[13,14]; (2) Compression of the left anterior descending artery may manifest as angina pectoris^[15]; (3) A characteristic sign of irritation of the left phrenic nerve is hiccups^[6]; and (4) The aneurysm proximity to the respiratory tract may cause a dry, unproductive cough^[16].

Cases of left atrial appendage aneurysm rupture with a lethal outcome have been described^[9].

In most cases, the disease is detected by chance because of a non-specific clinical picture. A convexity at the contour of the left atrium determined on a chest X-ray requires differential diagnosis with a pericardial cyst and heart and mediastinal tumors^[10,11].

Echocardiography is considered the main method for diagnosing left atrial appendage aneurysm and the presence of blood clots in it^[1]. While, due to the limited echo window, the transthoracic examination does not always make it possible to accurately establish the diagnosis, the transesophageal approach may provide a more detailed and clear visualization^[10].

Computed tomography and MRI are useful in unclear cases. MRI has a high resolution, which makes it optimal in assessing the surrounding structures and cardiac abnormalities. However, MRI has certain disadvantages, such as the need for regular

heart rhythm and patient's exposure to nephrotoxic contrast agents. Cardiac computed tomography makes it possible to evaluate the anatomy of the coronary arteries, if there is a suspicion of their compression, but cannot provide functional data as accurately as echocardiography or MRI^[10].

Surgical treatment is indicated for all patients to prevent the occurrence of atrial fibrillation, systemic embolism, or myocardial dysfunction^[17,18]. We performed a left atrial appendage aneurysm resection *via* median sternotomy with cardiopulmonary bypass, which is considered the operation of choice, especially in patients with thrombosis of the aneurysmal cavity and the need for concomitant interventions^[19].

Aneurysmectomy through left-sided lateral thoracotomy should be considered if isolated lesions of the left atrial appendage are present without significant violations of the heart anatomy^[20]. Endoscopic surgery is most suitable for patients with minor aneurysms in the absence of thrombosis^[21].

The issue of performing a concomitant intervention to restore sinus rhythm remains debatable. Following the most recent 2017 guidelines from the Society of Thoracic Surgeons, surgical ablation for atrial fibrillation is recommended at the time of concomitant mitral operations to restore sinus rhythm (Class I, Level A)^[22]. However, removal of the aneurysm usually results in resolution of atrial arrhythmias with no need for additional surgery^[23,24]. Moreover, concomitant atrial fibrillation surgery increases the risk for requiring a permanent pacemaker^[25]. We decided to abandon concomitant surgery for atrial fibrillation to limit the overall risks of the operation. Also, there was a possibility that atrial flutter might disappear after resection of the aneurysm. Unfortunately, arrhythmia disappeared only temporarily and required radiofrequency catheter ablation 2 mo after discharge from the hospital.

Limitations

This is a case report and the conclusions are subject to the inherent bias associated with the retrospective design. Also, this study design does not allow us to establish a cause-effect relationship between atrial appendage aneurysm and mitral regurgitation, as well as to differentiate between congenital and acquired forms of the aneurysm.

CONCLUSION

Left atrial appendage aneurysm is a rare heart defect. Given the nonspecific clinical picture, the disease is diagnosed either by chance or during the examination of a patient when complications arise. Echocardiography helps to establish the diagnosis in most of the cases. However, MRI is necessary for a more detailed assessment of changes in the heart. Surgical treatment is indicated for all patients to prevent fatal disorders. In the case when the circumflex artery is not visualized, coronary angiography may help to avoid adverse events. Antiarrhythmic surgery can decrease the risk of thromboembolic complications and may be performed either at the time of concomitant left atrial appendage resection or as a stand-alone surgical procedure, depending on the clinical situation.

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