Endovascular Repair of the Left Sinus of Valsalva Pseudoaneurysm in a 33-Year-Old Man with Wegener’s Granulomatosis

Shokoufeh Hajsadeghi,1 Maral Hejrati,2* Mitra Chitsazan,2 Samar Moghadami,2 and Majid Dehghani3

1Department of Cardiology, Iran University of Medical Sciences, Tehran, Iran
2General Practitioner, Tehran University of Medical Sciences, Tehran, Iran
3Department of Cardiology, Lavasani Hospital, Tehran, Iran

*Corresponding author: Maral Hejrati, General Practitioner, Tehran University of Medical Sciences, Tehran, Iran. Tel: +98-9124842477, Fax: +98-2188852654, E-mail: Maral.hejrati.mh@gmail.com

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Abstract

Introduction: Pseudoaneurysm formation is a rare complication. If this condition remains untreated, it can lead to morbidity and mortality. Pseudoaneurysm can be a consequence of iatrogenic trauma, vasculitis, infection, and sometimes it forms spontaneously. This condition was treated by surgery in the past. However, nowadays an interventional procedure is mentioned as an alternative for surgical treatment.

Case Presentation: Here, we report an unusual case of Wegener’s granulomatosis complicated by pseudoaneurysm of the left sinus of Valsalva causing complete collapse of the left main coronary artery. The pseudoaneurysm was closed percutaneously using a membranous device. This study presented a patient with the rare large artery involvement in Wegener’s granulomatosis and tried to repair that without surgery. Long-term follow-up demonstrated that the procedures were successful.

Keywords: Aneurysm False, Antineutrophil Cytoplasmic Antibody Associated Vasculitis, Granulomatosis with Polyangitis, Endovascular Procedures

1. Introduction

Pseudoaneurysm is a hematoma that forms outside of the arterial wall and communicates with the artery. Pseudoaneurysm of the sinus of Valsalva (aortic sinus) is a very rare complication. It can arise from any one of the pouches of aorta artery, which are displaced behind the valves (1). Acquired pseudoaneurysm can occur as a result of chest trauma, endocarditis, arteriosclerosis and medionecrosis. Wegener’s granulomatosis as a small to medium sized vasculitis can be an underlying disorder, which leads to pseudoaneurysm formation. Aortitis can be induced by c-ANCA associated vasculitis (2). Wegener’s granulomatosis incidence was estimated to be 1 in 30000 (3). This is not a hereditary condition. Hypersensitivity reaction to an external antigen is determined by its etiology (4). Presence of two or more than the 4 following criteria is associated with a specificity of 92% for Wegener’s granulomatosis. The criteria consisted of nasal inflammation, hematuria, abnormal chest radiography and biopsy, which confirmed the diagnosis (5). Cytoplasmic antineutrophil cytoplasmic antibody (cANCA) [antiproteinase 3] is highly specific for this condition. Although surgery is the gold standard treatment for pseudoaneurysm, several less-invasive procedures have been recently taken into consideration.

2. Case Presentation

A 33-year-old man was admitted to our hospital due to an one-month lasting history of shortness of breath and chest pain radiating to his right shoulder and interscapular region. His medical history showed Wegener’s granulomatosis, which had been first noted one year previously, beginning with symptoms of fever, anorexia, weight loss, myalgia and pleuritic chest pain. Computed tomography (CT) scan of the thorax demonstrated left pleural effusion, multifocal parenchymal nodules and peripheral basilar consolidation in the left lung. CT guided biopsy of the lung lesions had revealed hemorrhage and fibrinoleukocytic exudate. Accordingly, a diagnosis of Wegener’s granulomatosis had been confirmed by the results of serologic antibody tests, which had revealed a positive (c-ANCA) level and histopathologic conclusions. Treatment had been started prednisolone and cyclosporine.

In the recent admission, his physical examination was unremarkable. Routine laboratory data showed a normocytic anemia (hemoglobin = 8.8 g/dL, hematocrit = 27%, mean corpuscular volume, MCV = 94 fl) and microscopic hematuria. Work-up for his chest pain including electro-
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Figure 1. A Round Heterogeneous Pseudoaneurysm at the Posterior Aspect of the Left Sinus of Valsalva

Figure 2. Coronary CT Scan After Pseudoaneurysm Closure Using a Membranous Device Number 12

3. Discussion

Inflammatory pseudoaneurysms are generally believed to occur only in vasculitis involving large arteries such as giant cell arteritis and Takayasu’s arteritis. In this report, we present a case of a pseudoaneurysm in Wegener’s granulomatosis as a small- to medium-sized vasculitis.

Pseudoaneurysm of the sinus of Valsalva is a rare complication. Yuan et al. accumulated the English reported cases with pseudoaneurysm of the Valsalva sinuses, until 2009, and just 12 patients were reported with this condition, 5 out of them (41.7%) were stricken spontaneously, trauma and infection were the causes of pseudoaneurysm formation in 3 (25%) and 4 (33.3%), respectively (6). According to a world literature review by Arlet and colleagues, only 11 cases with major artery involvement in patients with Wegener’s granulomatosis were reported, and aorta was involved in 3 of them (7). Heydorn et al. in 1976 reported the first surgical method for pseudoaneurysm repair (8). While surgery was the gold standard treatment in the past, several less invasive treatment options are popular today. Treatment of two pseudoaneurysms from the left ventricular outflow tract after the Ross procedure (aortic valve replacement using pulmonary autografts) using an Amplatzer and coil embolization is reported in a 13-year-old boy with right sinus of Valsalva anomalies (9).

Transcatheter closure of isolated rupture of sinus of Valsalva has been mentioned as an alternative to surgical repair according to a retrospective study of 8 patients with this condition (10). An attempt has been done for perrcuta-
neous transcatheter closure of ruptured sinus of Valsalva aneurysm in eight patients between 1995 and 2003, among them one died of progressive congestive heart failure, one needed to surgical repair finally, while the procedure was successful in other six patients (11). A case with recurrence of aortic pseudoaneurysm was successfully treated with a second Amplatzer device in 2010 (12). Successful percutaneous closure of a descending thoracic aortic graft pseudoaneurysm with a patent foramen ovale occluder device is reported in a 22-year-old female with a history of hypertension and Takayasu’s arteritis (13).

References


