



## Ascending Aorta Hydatid Cyst Presenting as Pseudoaneurysm

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### ABSTRACT

Aortic wall with its poor blood supply is a very rare location of Hydatid Cyst (HC) infestation and mediastinal HC is a rare presentation of human hydatidosis. Here, we reported a case of primary mediastinal HC invading the ascending aorta, which initially presented as an aortic aneurysm. The patient underwent open cardiac surgery with cardiopulmonary bypass for resection of the aortic wall HC. During surgical exploration, it was found that the HC had ruptured into blood stream and residual cyst cavity contained a large thrombosis. Thus, the patient underwent ascending aorta replacement and removal of the cyst with thrombotic material. It seems that the signs and symptoms of stroke were related to emboli of cyst contents or thrombotic material into cerebral circulation. Invasion of aortic wall by hydatidosis is an exceedingly rare presentation of HC in the anterior mediastinum that may be wrongly diagnosed as a pseudoaneurysm of ascending aorta. Our study patient had an uneventful recovery and one-year follow-up revealed no recurrence of HC in mediastinum.

### ► Implication for health policy/practice/research/medical education:

It was intended for the patients who have HC rupture into blood stream and residual cyst cavity contained a large thrombosis during the surgery.

### 1. Introduction

Hydatid disease, which is caused by *Echinococcus granulosus* tapeworm, is endemic in some cattle-raising provinces of Iran (1). Careful literature review revealed that only 21 cases of ascending and descending aorta involvement by Hydatid Cyst (HC) were reported among which, only three cases were reported as ascending aorta involvement. Generally, ascending aorta involvement is caused by direct infestation of larva or by secondary invasion from another mediastinal focus. On the other hand, descending aorta is the most common segment of aorta that is involved by HC. The most common cause of descending aorta infestation by HC is secondary to presence of a focus of HC in posterior mediastinum or abdomen (2). Herein, we report a unique case of ascending aorta HC in a man who presented with an ascending aorta pseudoaneurysm

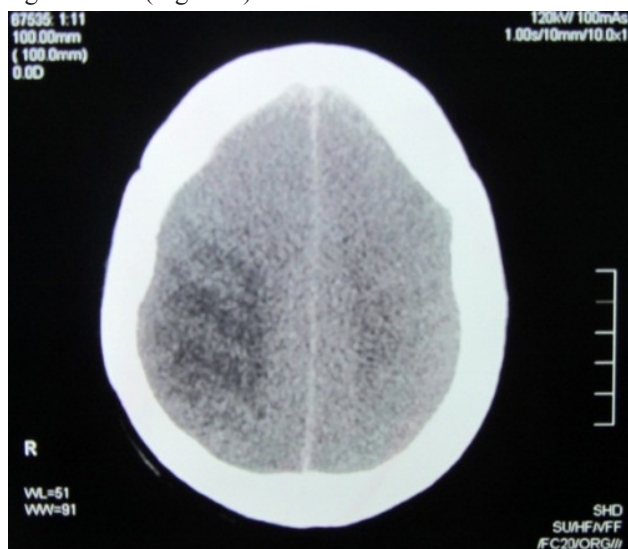
with intra-cavitary thrombosis. The patient presented with neurological symptoms and was first admitted in a neurology center.

### 2. Case Presentation

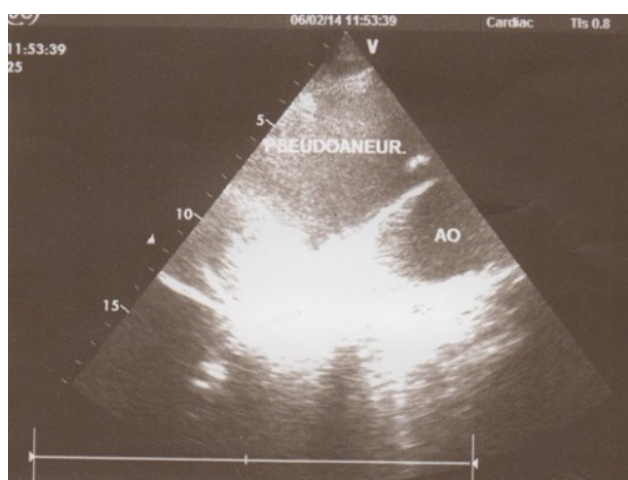
A 42-year-old cattleman with the history of close contact with dogs referred to our center for evaluation of cardiac pathology due to complains about delirium, dyspnea, and reduced consciousness. On the initial evaluation, he appeared alert. Although aphasic, he nodded appropriately to questions. The remainder of the neurological exam was non-remarkable. Aphasia slowly improved over several days. The patient was able to recount that his symptoms began with right upper-extremity weakness that progressed to right hemiparesis and inability to verbalize his feeling and pain. Then, the patient developed right-sided neurological findings. The neurological examination of the patient also revealed right hemiplegia. Thus, the patient was referred to our hospital from a neurology ward with the diagnosis of transient neurological attack. Preoperative brain Computed

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Tomography (CT) scans revealed an ischemic change in the left brain hemisphere (Figure 1). Additionally, plain chest X-ray revealed a large mass in the right border of the cardiac shadow. Transe Thoracic Echocardiography (TTE) also revealed a 15-cm ascending aortic pseudoaneurysm in the right border of the ascending aorta compressing the right atrium (Figure 2).



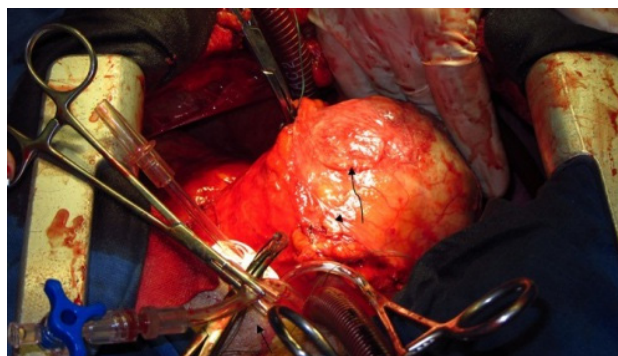
**Figure 1.** Ischemic Stroke of the Left Hemisphere



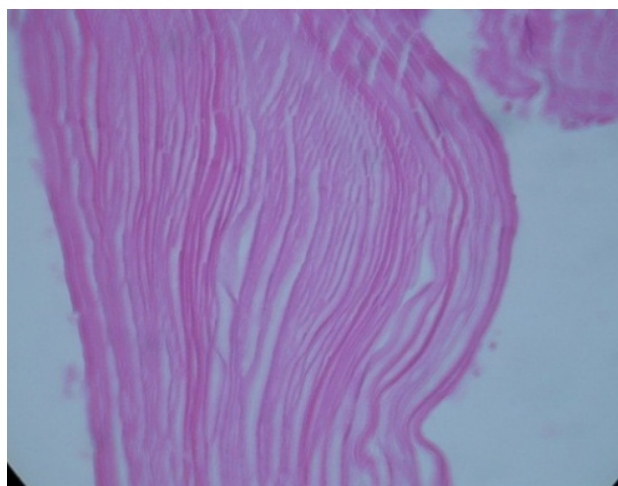
**Figure 2.** The Huge Pseudoaneurysm

Ascending aorta had a normal diameter in the upper and lower parts of the pseudoaneurysm. Since the pathological diagnosis of pseudoaneurysm was made and HC was not primarily considered in differential diagnosis, CT scan of other organs was not performed to look for other possible locations of HC. Gross pathology of the resected cyst revealed its malignant behavior by rupture of the cyst to the blood stream (3). Signs and symptoms of the ruptured cyst were retrogradely taken from the patient's family. However, they documented the presence of symptoms related to anaphylaxis when he developed systemic rash, red skin, and neurological symptoms. Cerebral emboli of the cyst contents, in this case, could be either daughter cysts or thrombosis formation in the inner layer of the cyst. Therefore, open heart surgery was performed through a median sternotomy.

Cardiopulmonary Bypass (CPB) was instituted by aortic and bi-cava cannulation. The mass was intra pericardial, located on the lateral aspect of the ascending aorta, extended infero-laterally to the right atrium, and pushed the right atrium medially (Figure 3). After cardioplegic arrest, the ascending aorta was opened just distal to the sino-tubular junction. Aortic lumen was opened transversely to explore the distal border of the lesion and intra-cavity thrombosis. Intraoperative exploration of the lesion revealed that it was an HC. Histopathological examination also confirmed the diagnosis of hydatidosis (Figure 4).



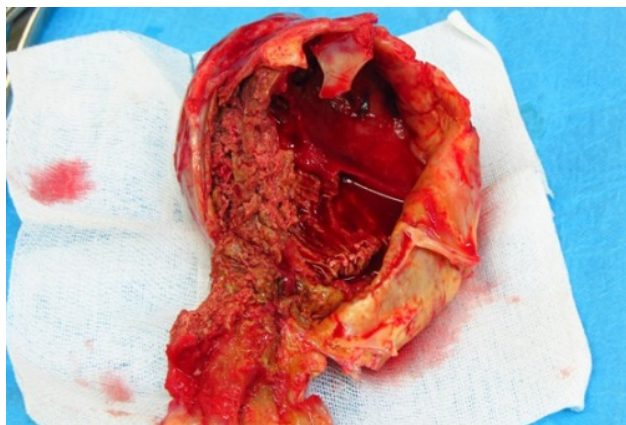
**Figure 3.** The Normal Ascending Aorta in the Cardioleg in the Cannula Site (Straight Arrow) and the Huge Hydatid Cyst (Curved Arrow)



**Figure 4.** Microscopic View of the Hydatid Cyst's Ectocyst

The ruptured HC with a narrow mouth constituted the lateral wall of the aorta causing a cul-de-sac fill of thrombosis, which was wrongly named as a pseudoaneurysm (Figure 5). After covering of the surrounding aneurysm with betadine soaked sponges, it was resected. Then, ascending aorta replacement was performed using a Dacron graft (Figure 6). Proximal anastomosis was also performed at the level of the sinotubular junction while warming started. The rest of the mass, which contained friable necrotic material, membranes, and daughter vesicles, was removed together with its pericystic wall from the surrounding tissues. After all, the patient was weaned from CPB. The postoperative period was eventful by neurological dysfunction and right hemiplegia. Hence, recovery was prolonged and the patient was weaned from the ventilator with hemiplegic sequel on



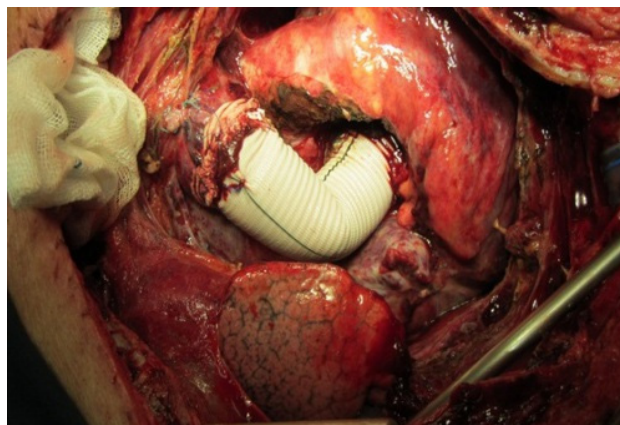


**Figure 5.** The Cul-de-sac of the Opened Hydatid Cyst with the Germinal Layer Filled With Clot

the 15th day after the operation. The patient was discharged with instructions to take albendazole for about a year and was scheduled for careful follow-ups every month.

### 3. Discussion

HC is an endemic infestation in some provinces of Iran. Ingested parasite embryo from carnivorous fecal materials by humans crosses the intestinal wall defense mechanism easily and enters the visceral circulation where many larva are filtered in the portal venous system and the overloading larva cause Budd Chiari syndrome and portal hypertension. The remaining larva that cross the portal barrier reach the hepatic barrier system and their filtration by sinusoidal macrophages cause hepatic HC (4). Those that escape the liver defense barrier may be entrapped in pulmonary circulation and cause lung HC and pulmonary hypertension in some patients. Therefore, liver and lung are the typical locations of the cysts. Rarely, HC larva can enter the systemic circulation and directly infest great vessels, such as aorta. Larva lodging in the aortic wall transformed to a cyst may rupture into the intra-aortic lumen and echocardiographic view of the remaining hydatid sac is similar to a pseudoaneurysm. The incidence of great vessels involvement by HC is unknown and limited data are available in this regard. Descending aorta is the most common site of secondary HC invasion to the aortic wall (5). Most cases primary involve lung or abdominal HC that invades neighboring organs, such as thoracic aorta. On the other hand, ascending aortic wall involvement by HC is very rare. Harris reported a case of primary lung HC where it eroded into the thoracic aorta, causing massive hemoptysis (6). Marti also reported a descending aorta HC secondary to primary lung hydatidosis that resulted in lower extremity emboli (7). Additionally, Posacioglu mentioned a case of primary HC presenting as a large cyst involving the wall of the descending aorta and complicated by a false aneurysm (8). Biglioli also showed a rare case of ruptured pericardial HC that, as a secondary cyst, invaded the ascending aorta and caused fistula and pseudoaneurysm (9). Besides, Hadjiat demonstrated a case of a retroperitoneal HC ruptured into the abdominal aorta. The initial treatment that consisted of marsupialization of the cyst with patch angioplasty of the aorta was unsuccessful and complicated by formation of a



**Figure 6.** Replacement of the Ascending Aorta with Dacron graft

false aneurysm. The secondary operation led to placement of an aorto-bifemoral bypass, but led to a fatal outcome (10). Nisanoglu also reported a case of an abdominal aortic embolism due to rupture of a cardiac HC. They emphasized diagnostic, preventative, and treatment options for HC embolism of the abdominal aorta (11). Furthermore, Dar reported a case of HC embolism from a ruptured hydatid in a pseudoaneurysm of the descending aorta that presented with blue toe syndrome of the right lower extremity (2). After embolectomy, microscopic examination of the embolus revealed it to be blood clots breaking off to systemic circulation from attaching thrombosis to internal hydatid membranes. Escudero, too, reported a case of invading HC to ascending aorta that eroded to the chest wall and presented as a chest wall tumor (12). Moreover, Geramizadeh showed that cysts tended to appear in different and sometimes unusual body sites in various geographical areas of the world (13). In the same line, Tüzün et al. described various characteristics of HC in CT scans in different body organs (14). In a rare case report, researchers reported an HC in the wall of the descending thoracic aorta that eroded to the spine (15). Furthermore, Senturk carried out a unique imaging study of intra-pulmonary artery HC by endobronchial ultrasound. Endobronchial ultrasound clearly showed sonographic characteristics of this specific location of HC (16). Mehrabani investigated all cases of HC in East Azerbaijan province. No unusual case was reported among the 52 cases under investigation. Nevertheless, liver was the most common site of HC (17).

### 3.1. Conclusion

Although presentation of HC as an ascending aorta pseudoaneurysm is exceedingly rare, the surgical approach is the same for these two different pathologies.

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### Authors' Contribution

Study concept and design: Sabzi and Faraji. Analysis and interpretation of data: Sabzi and Faraji. Drafting of the manuscript: Sabzi and Faraji. Critical revision of the manuscript for important intellectual content: Sabzi and Faraji.

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The authors declare that there is no conflict of interests.

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