







## “Singultus” unmasking potentially fatal vascular dissections

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### ABSTRACT

Aortic dissection (AD) is a serious condition in which the intimal layer of aorta tears and blood surges in between the intimal and medial layers of aorta causing it to separate (dissect). It usually presents with excruciating pain radiating to the back. Here we present a unique presentation of AD where an old-aged Caucasian male presented with a chronic history of intractable hiccups. His computed tomography (CAT scan) revealed the dissection of the descending thoracic aorta. He was managed conservatively and was discharged home in stable condition. The purpose of this report is to highlight this unusual presentation of AD and unmask the possible etiology of hiccups in such cases.

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Hiccup; dissection; cardiovascular medicine; cardiovascular system

## 1. Introduction

AD has been traditionally considered as an important differential of chest or back pain, especially in patients with risk factors such as smoking or hypertension. Other than chest pain it may also present with fainting, stroke, signs of heart failure, and cardiac arrest. Our case and the literature review we performed showed that it can present very unusual symptoms like hiccups due to the irritation of the phrenic nerve. Physicians should therefore keep a high index of suspicion for such presentation in the appropriate clinical settings and should take measures to exclude this fatal association of hiccups with AD.

### 1.1. Case presentation

A 62-year-old male presented with nausea, vomiting, and hiccups for two days. He mentioned that he first started feeling nauseous and has since vomited three times. Vomitus was nonbilious, non-bloody, and was followed by continuous hiccups. On further questioning, he said that he has episodic hiccups for the last two years, but it has recently got worse and has started interfering with his sleep and daily activities. He denied abdominal pain, fever, chest pain, cough, dysphagia, reflux, shortness of breath, headache, dizziness, problem with hearing or vision, swelling of legs, or weight change. He endorses being pre-hypertensive 20 years ago but has not followed his primary care physician regularly and did not know his baseline blood pressure. The patient denied having any other medical conditions in the past and has not been using any medications on a regular basis.

On presentation, His blood pressure was 191/101. On initial examination, the patient looked distressed

due to ongoing hiccups. Physical examination including chest, abdomen, head, neck, ear, throat and complete neurological examinations were normal.

### 1.2. Investigations

Pertinent investigations included hemoglobin of 12.4g/dl, platelets of 103,000, and mildly raised D-dimer levels of 309. His liver functions were significant for AST 82 U/l and ALT 144 U/l. After initial investigations, a CAT scan of the thorax was performed which showed dissection involving the descending thoracic aorta including portions of the abdominal aorta (Figure 1).

### 1.3. Differential diagnoses

On presentation, our suspicion was GERD, Peptic Ulcer disease or uremic hiccups, but absence of pain and normal kidney functions ruled out these causes.

### 1.4. Treatment

Initially, the patient was managed symptomatically with antiemetics and famotidine. The blood pressure was controlled with nicardipine and metoprolol and chlorpromazine was given for hiccups management. After definitive diagnosis of AD, treatment for blood pressure was optimized with amlodipine and metoprolol. Vascular surgery was taken aboard, but the patient denied any surgical intervention.



**Figure 1.** Computed tomography angiography (CTA) showing descending thoracic aortic dissection.

### 1.5. Outcome and follow-up

Patient's nausea and vomiting were relieved, but hiccups persisted for two more days and were finally relieved. His blood pressure was well controlled with medications, and he was discharged on the third day of his admission with plans for follow up as an outpatient.

## 2. Discussion

Watson et al. in 1956 for the first time introduced arterial dissection as a condition resulting from blood penetration into arterial wall, causing a split between the vessel coats like medial laminae, with or without a tear of the tunica intima (inner vessel layer) [1]. Many predisposing conditions have been identified since then, such as hypertension in about 70–80% patients, positive family history, aortic valve disease, aortic aneurysm, aortic surgeries, and connective tissue diseases like Marfan and Ehlers-Danlos Syndrome [2]. The most common presentation in the majority of patients (greater than 90%) is chest pain. Traditionally radiating chest pain to the back in appropriate clinical settings was thought to be the hallmark for AD. Our case was unique where the patient denied any chest pain and presented with unusual symptoms like persistent hiccups.

There is scant literature about this rare presentation and hence the mechanism remains unknown. We believe that the cause for hiccups could be

a direct mechanical effect on the phrenic nerve which lies in the proximity to the dissecting aorta. The three previously reported cases also mentioned the same mechanism [3]. Owing to the long course of phrenic nerve, it can be hypothesized that other arteries like carotid and main branches of the aorta can also impinge on the phrenic nerve and can give rise to hiccups. What caused them to present with painless hiccups is still unknown.

We did a comprehensive literature search on PubMed using different MeSH terms for hiccups and aortic dissection. All retrieved articles were read by two independent authors and only three reported cases were found where hiccups were the presenting complaint for dissection (Table 1).

All these cases were reported in male and mostly in their middle aged or old aged. Only one of the reported cases had aortic dissection as the cause of hiccups while the other two cases were of vertebral and carotid dissection. All the cases had an associated risk factor with hypertension present in two cases and one patient had a history of smoking and migraine. Diagnosis was made by CT scan, MRA, and angiography, respectively. Two of the cases had successful recovery with conservative management in the patient with aortic dissection and with possible stenting in the patient with vertebral artery dissection. There was no data available on outcome and management in the patient with carotid artery dissection. Our case marks the fourth case of an arterial dissection associated with hiccups, and the second case of aortic artery dissection presenting with hiccups. Our patient was unique in terms of presentation as he was having intractable hiccups and gastrointestinal symptoms like nausea and vomiting.

There are certain physical and investigative findings suggestive for AD. AD can be associated with the 'pulse deficit' meaning that loss of pulse with deep inspiration, due to compression by expanding hematoma or intimal tear. Patients can have a significant difference in systolic blood pressure (>20 mm Hg) in two arms [6]. In cases of ruptured AD, patient can have hypotension and even shock can occur. The cause may be overt bleeding into the pericardial cavity (cardiac tamponade) or due to acute aortic regurgitation and rupture of the aorta. Patients can surprisingly have focal neurological signs and symptoms like stroke, altered consciousness,

**Table 1.** Previously reported cases of dissections presenting with hiccups.

Author /year/ref	Age/ sex	Presentation	Comorbidities	Artery involved	Diagnosis	Management	Outcome	Follow- up
RA Badawi [4]	38/M	Right sided weakness, word finding difficulty following hiccups	Migraine, Smoker	Carotid	MRA	N/A	N/A	N/A
StanislavHoliccek [5]	58/M	Hiccups, back pain	Hypertension	Aorta	CT scan	Conservative	Survived	None
N/A	26/M	Neck pain, hiccups	Hypertension	Vertebral	Angiography	Procedure	Survived	None
This case	62/M	Hiccups, nausea, vomiting	Hypertension	Aorta	CT scan	Conservative	Survived	N/A

Horner syndrome, hoarseness due to the extension of dissection or due to the mass effect [7]. A diastolic decrescendo murmur can be heard with the propagation of tear to the aortic valve [8]. An initial workup should include an electrocardiogram to rule out acute coronary conditions. Findings suggestive of AD are chest X-ray showing widening of aortic silhouette in 60–90% of cases and d-dimer (<500 ng/ml) having sensitivity of about 97% [9]. CT angiography, MR angiography, and transesophageal echocardiogram are imaging used to diagnose dissection [10].

AD management should be individualized. For patients who are hemodynamically stable with uncomplicated Type B dissection, medical management with controlling pain and lowering blood pressure to reduce shear stress and extension of dissection can be done [11]. Hemodynamic instability or acute Type A dissection is a surgical emergency as it can lead to life-threatening complications like cardiac tamponade and acute myocardial infarction and acute aortic regurgitation. Blood pressure control ideally below 120/80 is beneficial in these patients [11]. After stabilizing the patient, it is important to have repeat imaging to look for the resolution of AD and patients should have a regular follow up with the aim to control the inciting risk factors.

### 3. Conclusions

1. Aortic dissection can present with unusual symptoms such as hiccups, nausea, and vomiting without the usual presenting complaint of chest pain.

2. Physician should consider aortic dissection in the differential diagnosis of patients

presenting with intractable hiccups with risk factors of dissection such as smoking or blood pressure.

3. Chlorpromazine can be effective in the management of hiccups associated with aortic dissection.

### Disclosure statement

No potential conflict of interest was reported by the authors.

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### References

- [1] Watson A. Dissecting aneurysm of arteries other than aorta. *J Pathol Bacteriol.* 1956;72:439–449.
- [2] Pape LA, Awais M, Woznicki EM, et al. Presentation, diagnosis, and outcomes of acute aortic dissection: 17-year trends from the international registry of acute aortic dissection. *J Am Coll Cardiol.* 2015 Jul 28;66(4):350–358.
- [3] Hagan PG, Nienaber CA, Isselbacher EM, et al. The International Registry of Acute Aortic Dissection (IRAD): new insights into an old disease. *Jama.* 2000 Feb 16;283(7):897–903.
- [4] Holicek S. Hiccup as a major sign of aortic dissection. *Int J Cardiol.* 2014 Dec 20;177(3):e105–6.
- [5] Badawi RA, Birns J, Ramsey DJ, et al. Hiccups and bilateral carotid artery dissection. *J R Soc Med.* 2004 Jul;97(7):331–332.
- [6] Mehta RH, O’Gara PT, Bossone E, et al. Acute type A aortic dissection in the elderly: clinical characteristics, management, and outcomes in the current era. *J Am Coll Cardiol.* 2002 Aug 21;40(4):685–692.
- [7] Nienaber CA, Fattori R, Mehta RH, et al. Gender-related differences in acute aortic dissection. *Circulation.* 2004 Jun 22;109(24):3014–3021.
- [8] Nienaber CA, Eagle KA. Aortic dissection: new frontiers in diagnosis and management: part I: from etiology to diagnostic strategies. *Circulation.* 2003 Aug 5;108(5):628–635.
- [9] Suzuki T, Distanto A, Zizza A, et al. IRAD-bio investigators: diagnosis of acute aortic dissection by D-dimer: the International Registry of Acute Aortic Dissection Substudy on Biomarkers (IRAD-Bio) experience. *Circulation.* 2009 May 26;119(20):2702–2707.
- [10] Kienzl D, Prosch H, Töpker M, et al. Imaging of non-cardiac, non-traumatic causes of acute chest pain. *Eur J Radiol.* 2012 Dec 1;81(12):3669–3674.
- [11] Tsai TT, Nienaber CA, Eagle KA. Acute aortic syndromes. *Circulation.* 2005 Dec 13;112(24):3802–3813.