

Headache as an Unusual Presentation of Ischemic Heart Disease: A Case Report

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Abstract

The clinical features of coronary artery disease vary, and patients may present with symptoms other than chest pain, such as headache. Rarely, the headache may be the only presenting feature without any chest discomfort, and may be confused with migraine. Failure to distinguish such headache, caused by CAD, from migraine may result in wrong treatment with disastrous fate. Elderly patient with the presence of cardiovascular risk factors having recent onset exertional headache should be evaluated for the presence of cardiac cephalgia. We intend to report a 60-year-old hypertensive, diabetic patient with a 6-months history of episodic exertional headaches, who turned out to be a case of headache angina (cardiac cephalgia). [*Journal of National Institute of Neurosciences Bangladesh, 2019;5(1): 81-86*]

Keywords: Headache angina; Cardiac cephalgia; Cardiac cephalgia

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Introduction

The clinical features of coronary artery disease (CAD) vary, and patients may present with symptoms other than chest pain. Patients with CAD who present without chest pain is frequently misdiagnosed and undertreated. These patients may have pain at other sites such as arm, shoulder, back, jaw, or epigastrium. Pain at each of these sites may occur alone or in different combinations^{1,2}. Headache as the only symptom of myocardial ischemia is quite rare and there are only few case reports in the literature on cardiac cephalgia or cephalgia^{3,4}.

Case Presentation

A 60-year-old hypertensive, diabetic patient presented in September 2018 with a 6-months history of episodic headaches. All attacks occurred only on exertion,

initially after walking for about 10 to 15 min and later on after walking for just 1 to 2 min. The headache was also provoked by activities such as lifting heavy objects. The headache was intense, excruciating in quality, and rated as 9 in severity on the visual analog scale. This intense and excruciating pain used to last for 10 to 60 min in each attack. The headache used to subside with complete rest. His routine and social activities were seriously hampered. The pain used to start posteriorly, becoming holocephalic within seconds. On a few occasions, the headache was associated with nausea. However, there was no vomiting, photophobia, phonophobia, or any aura. He never had any abdominal pain, chest discomfort or atypical chest pain, jaw claudication, or breathlessness associated with exertion or headache. He was diabetic for 10 years and

hypertensive for 12 years. Neurological examinations were normal; blood pressure was 170/90mm-Hg. Routine hematological and biochemical investigations were normal. A resting electrocardiogram (ECG) was within normal limits (Figure I). Echocardiogram showed no regional wall motion abnormality with good ejection fraction (63.0%). He had undergone magnetic resonance imaging (MRI) of the brain and cervical spine, which did not reveal any abnormality. Prior treatments with various drugs such as, amitriptyline, valproates, topiramate, flunarizine, paracetamol, indomethacin, and ibuprofen provided no benefit.

In view of exertional-only headache, a possibility of cardiac cephalgia or headache angina was considered. The patient got complete relief from headache within minutes of taking single tablet of sublingual nitroglycerin which he described as the best response with any drug. Improvement with nitroglycerine strongly suggested cardiac cephalgia. During stress test, the patient had similar headache which was associated with 2 mm depression of ST segment in inferolateral leads. The patient did not feel any chest symptom during stress testing. Headache

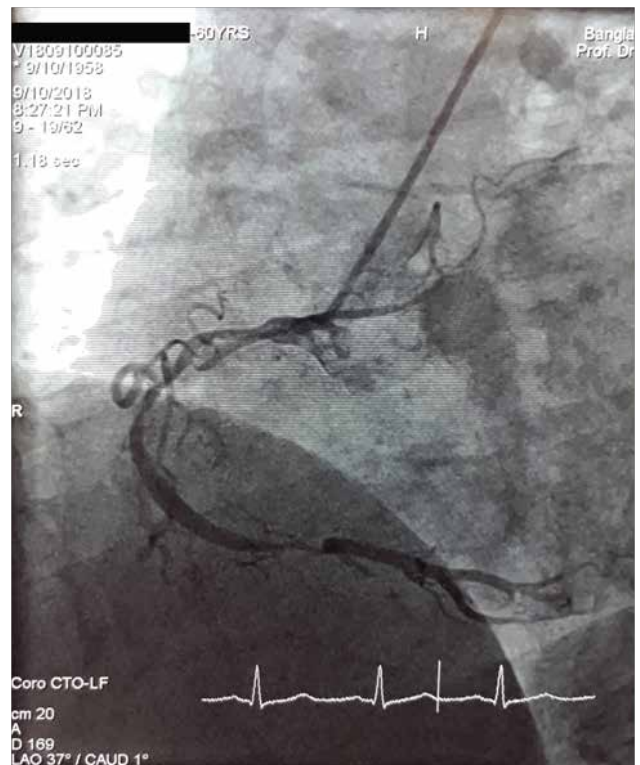


Figure II: Coronary angiogram showing 60 to 70% stenosis in proximal to mid RCA and another 90% stenosis in distal RCA

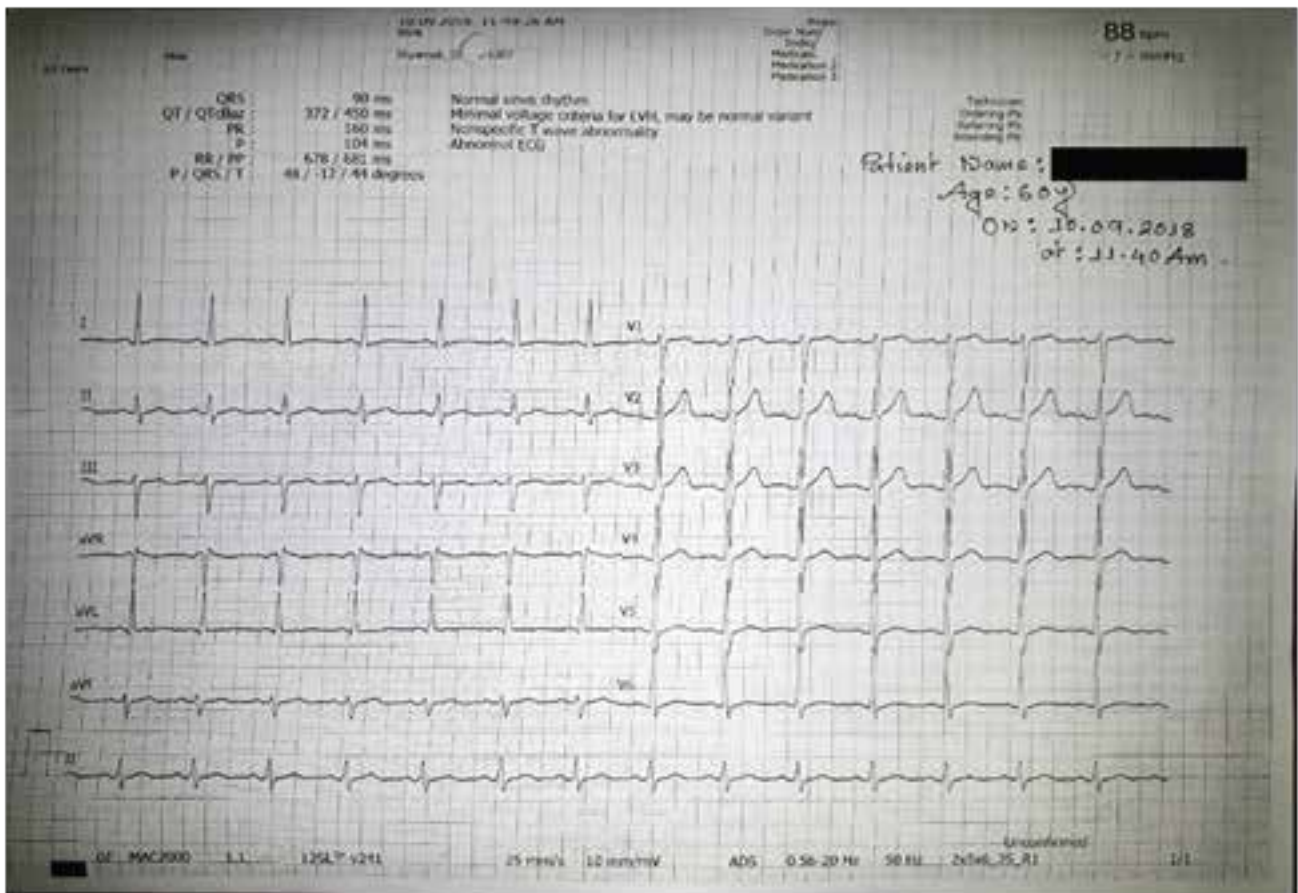


Figure I: Resting 12 lead ECG showed normal findings



Figure III: Coronary angiogram showing 95.0% stenosis in mid LAD involving origin of D1

nitroglycerine. On 10th September 2018, a coronary angiogram revealed three vessel disease with 60-70% stenosis in proximal to mid right coronary artery (RCA), another 90% stenosis in distal RCA (Figure II), 95% stenosis in the mid LAD involving D1 (Figure III) and 30 to 40% stenosis in mid LCX and 80 to 90% stenosis in principle OM (Figure IV). Coronary artery bypass surgery was done with 4 grafts. At follow up, two weeks after discharge, the patient was completely symptom free.

Discussion

An exertional headache has broad differential diagnoses, which includes both primary and secondary headache disorders. Vast majority of exertional headaches are benign. The secondary causes associated with exertional headaches include space-occupying lesions especially of posterior fossa, vascular abnormalities like aneurysm or arteriovenous malformation, Chiari malformation, and other obstructions of CSF flow⁵. Cardiac cephalgia is an extremely rare type of exertional headache. There are about 33 cases of cardiac cephalgia in the literature. Knowledge of cardiac cephalgia and an early diagnosis is important to prevent any catastrophe^{3,4,6}.

A diagnosis of cardiac cephalgia is made according to

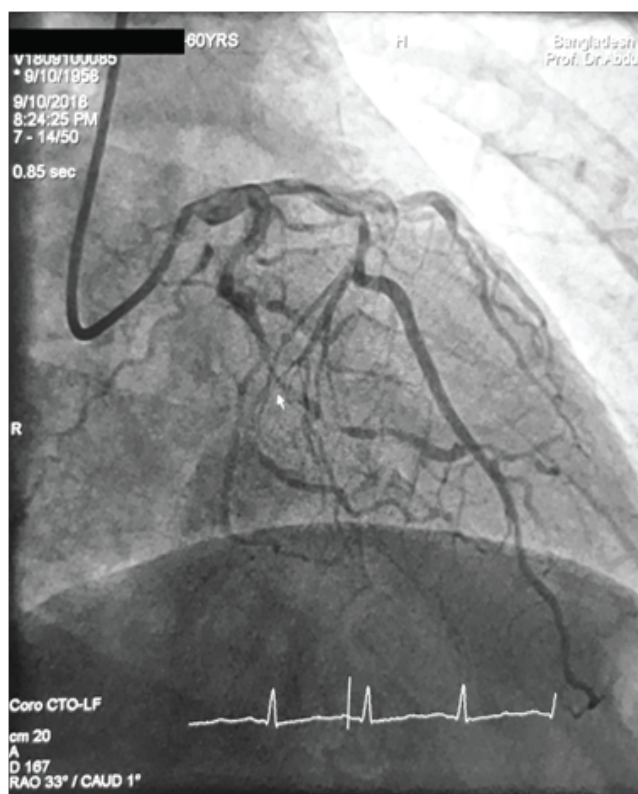


Figure IV: Coronary angiogram showing 30 to 40% stenosis in the mid LCX and 80 to 90% stenosis in principle OM

International Classification of Headache Disorders (ICHD)-3 β criteria⁷, which depend on the documentation of causation of headache by cardiac ischemia. Evidence of causation is suggested by the presence at least two of the following⁷: Headache developed in temporal relation to onset of acute myocardial ischemia; Headache worsened with worsening of the myocardial ischemia or headache improvement with improvement in the myocardial ischemia; At least two of the following four characteristics like moderate to severe intensity, accompanied by nausea, not accompanied by photophobia or phonophobia, and aggravated by exertion; and headache is relieved by nitroglycerine or its derivatives. This patient showed all four features.

Approximately 50.0% of cases of CAD are recognized because of the chest pain⁸. However, chest pain may arise in or radiate to the neck, jaws, tongue, teeth, throat, occiput, cheeks, tip of the nose, ears, shoulders, arms, hands, and to the epigastrium. An exertional headache has broad differential diagnoses, which includes both primary and secondary headache disorders. Vast majority of exertional headaches are benign. The secondary causes associated with exertional headaches include space-occupying lesions especially of posterior fossa, vascular abnormalities

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Our case presented with exertional headaches. Structural intracranial pathology was ruled out by neuroimaging. This case fulfilled the ICHD-3 criteria of cardiac cephalgia⁹. Headache was always exertional and the patient had immediate symptomatic relief with sublingual nitrate. Moreover, he had complete relief by coronary bypass surgery confirming that the pain was cardiac in origin. To the best of our knowledge, this is the second case of cardiac cephalgia reported from Bangladesh. The first reported case of headache angina in Bangladesh was by Chowdhury et al⁶ in 2015.

Most reported cases of cardiac cephalgia had headache as the main presenting feature^{3,4}. However, only 27% patients reported headache as the only manifestation of cardiac ischemia⁴. About 50%, patients also had mild

nonspecific pain in chest, epigastrium, arm or mandible⁴. Cardiac cephalgia may closely mimic migraine. Both cardiac cephalgia and migraine produce severe exertional headache with autonomic features particularly nausea. Up to 30% patients with cardiac cephalgia may have either nausea or vomiting^{3,4}. This patient felt nausea on a few occasions, but he denied the presence of any other symptoms. It is extremely important to differentiate cardiac cephalgia with migraine as erroneous use of triptans may progress cardiac ischemia. Interestingly, nitroglycerine which induces migraine provides relief in patients with cardiac cephalgia^{4,11}.

Majority of these patients have one or more risk factors for cardiovascular events, such as hypertension, diabetes, smoking, dyslipidemia and positive family history^{3,4}. This patient was diabetic and hypertensive.

Cardiac pain is mediated by sympathetic and/or parasympathetic vagal fibers. These fibers converge to the somatic fibers of the various structures of the body. Anginal pain is mediated by sympathetic fibers from C8 to T5 in 50 to 60% of cases, by vagal fibers in 10 to 20%, and through both neural fibers in 30 to 40%¹². These variations in convergence of fibers are largely responsible for the variation of cardiac pain. If parasympathetic fibers are involved, the patients would get pain in the neck and head distribution. A recent observation on 326 patients with confirmed myocardial ischemia demonstrated increased prevalence of craniofacial pain with inferior wall ischemia¹³. It is a well-known fact that symptoms of inferior wall ischemia are mediated by parasympathetic fibers of vagus nerve.

A few other hypotheses have also been suggested to explain the craniofacial pain in CAD. Elevations in intracranial pressure, release of neurochemical mediators, or spasm of cranial vasculatures are few other explanations for the headache in patients with CAD^{3,4}.

Unrecognized myocardial infarction is quite common in the elderly people. About 21.0 to 33.0% elderly men and 26.0 to 54.0% elderly female with myocardial infarction may escape clinical attention¹⁴. Therefore, it is possible that large number of patients with cardiac cephalgia remain undiagnosed. It can also be speculated that a subset of patient with myocardial ischemia who escape attention may have headache as a sole presentation.

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Conclusion

Diagnosis of cardiac cephalgia is very important. Review of the literature suggest mortality rate of about 12.0% for cardiac cephalgia. Therefore, it could be suggested that any elderly patients with the presence of cardiovascular risk factor having recent onset exertional headache should be evaluated for the presence of cardiac cephalgia.

References

1. Brieger D, Eagle KA, Goodman SG, Steg PG, Budaj A, White K, et al. Acute Coronary Syndromes Without Chest Pain, An Underdiagnosed and Undertreated High-Risk Group: Insights from The Global Registry of Acute Coronary Events. *Chest*. 2004;126(2):461–9
2. Canto JG, Goldberg RJ, Hand MM, Bonow RO, Sopko G, Pepine CJ, et al. Symptom presentation of women with acute coronary syndromes: myth vs reality. *Arch Intern Med*. 2007;167(22):2405–13
3. Wei J-H, Wang H-F. Cardiac cephalgia: case reports and review. *Cephalalgia Int J Headache*. 2008 Aug;28(8):892–6.
4. Bini A, Evangelista A, Castellini P, Lambro G, Ferrante T, Manzoni GC, et al. Cardiac cephalgia. *J Headache Pain*. 2009 Feb;10(1):3.
5. Alvarez R, Ramón C, Pascual J. Clues in the differential diagnosis of primary vs secondary cough, exercise, and sexual headaches. *Headache*. 2014 Oct;54(9):1560–2.
6. Chowdhury AW, Saleh MAD, Hasan P, Amin MG, Khan TA, Sabah KMN, et al. Cardiac cephalgia: A headache of the heart. *J Cardiol Cases*. 2015 May;11(5):139–41.
7. Headache Classification Committee of the International Headache Society (IHS). The International Classification of Headache Disorders, 3rd edition (beta version). *Cephalalgia Int J Headache*. 2013 Jul;33(9):629–808.
8. Davies SW. Clinical presentation and diagnosis of coronary artery disease: stable angina. *Br Med Bull*. 2001;59:17–27.
9. Sampson JJ, Cheitlin MD. Pathophysiology and differential diagnosis of cardiac pain. *Prog Cardiovasc Dis*. 1971 May;13(6):507–31.

10. Caskey WH, Spierings EL. Headache and heartache. *Headache*. 1978 Nov;18(5):240–3.
11. de Tommaso M, Libro G, Guido M, Difruscolo O, Losito L, Sardaro M, et al. Nitroglycerin induces migraine headache and central sensitization phenomena in patients with migraine without aura: a study of laser evoked potentials. *Neurosci Lett*. 2004 Jun 17;363(3):272–5.
12. Meller ST, Gebhart GF. A critical review of the afferent pathways and the potential chemical mediators involved in cardiac pain. *Neuroscience*. 1992;48(3):501–24.
13. Kreiner M, Alvarez R, Waldenström A, Michelis V, Muñoz R, Isberg A. Craniofacial pain of cardiac origin is associated with inferior wall ischemia. *J Oral Facial Pain Headache*. 2014;28(4):317–21.
14. Leening MJG, Elias-Smale SE, Felix JF, Kors JA, Deckers JW, Hofman A, et al. Unrecognised myocardial infarction and long-term risk of heart failure in the elderly: the Rotterdam Study. *Heart Br Card Soc*. 2010 Sep;96(18):1458–62.