Myocardial bridging of coronary arteries: Sudden unexpected deaths in a 24–year–old man

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Abstract: Myocardial bridging is a congenital anomaly, mostly found incidentally in men, and primarily involved in the left anterior descending coronary artery. In some cases its association with certain cardiac complications and resultant sudden deaths has been reported. However, its role as a cause of death is rare and still controversial. The author reported the case of sudden unexpected death in a 24–year–old man with previous healthy history. Unlike previous reported cases which almost exclusively involved in a single bridge, the present case had all branches of coronary artery running deeply and lengthy through the myocardium, thus aggravating cardiac complications and rendering the death in this young man. Since no other possible causes of death could be found in the present case except for all branches of the coronary artery embedding into the myocardium with hypertrophic cardiomyopathy associated pathologic lesions, his death was attributable to fatal arrhythmia secondary to acute transient occlusion of myocardial bridging of the coronary arteries.

Key Words: coronary artery, medico-legal autopsy, myocardial bridging, sudden unexpected death.

Myocardial bridging (MB) was first illustrated in English literature in 1796 by Black [1] as a segment of a major epicardial coronary artery running through the myocardium, thus termed the “tunneled artery” or “mural artery”. MB is found in 12% to 30% of angiographic studies [2, 3] and up to 85% of autopsy studies [4-6], depending on the cases selected, the method used and the criteria defined for the studies, with most involvement in men [2-6]. MB is a congenital anomaly and originally thought as a benign variation of coronary anatomy [4-6]. It has been then recognized as an incidental phenomenon. Yet, in some cases its association with myocardial ischemia [7], acute myocardial infarction [8], hypertrophic cardiomyopathy [9], cardiac arrhythmia [10], atrioventricular block [11], atherosclerosis [12], thrombosis [13], and sudden death [14] has been reported. Also, it has been implied that MB may have the same significant clinical morbidity and mortality as coronary atherosclerosis [15]. Although, death due to MB is rare and still controversial, the author had experienced a sudden unexpected death in a young man whose cause of death was attributable to MB of the coronary arteries and reported herein.

CASE REPORT

A 24–year–old man was found lying dead on the floor of his rented accommodation by his father while waking him up to work on that morning. He was single and lived with his parents. At scene, the external examination of the body showed no signs of injury. He was fully dressed. Peripheral cyanosis was markedly present. According to his father, he had previous good healthy history, smoked 1–3 cigarettes per day for few years and occasionally drank alcohol with his friends. He had no recent history of trauma, medication, medical treatment, or drug abuse. There was no family history of acute coronary syndrome or established cardiovascular diseases.

At autopsy, the decedent revealed a muscular young man, measured 170 cm in length and 79 kg in weight.

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No external injuries or abnormalities were detected. The brain weighed 1550 g. Neither subdural nor subarachnoid hemorrhage was found. The cerebral hemispheres disclosed diffuse gyral widening and flattening consistent with cerebral edema. No spot hemorrhage or brain herniation was detected in cerebrum and cerebellum. The heart weighed 410 g and appeared slightly enlarged with mild dilatation of left lower chamber. The left ventricular wall was a little bit thinner than normal ones, measured 1.4 cm thick. All valves were normal appearance. The apertures of the right and left coronary arteries were located within their respective sinuses of Valsalva. Near their origins, cross sections of coronary arteries displayed approximately a 6.5 cm segment of the right coronary artery, a 6 cm segment of the left anterior descending coronary artery, and a 1.5 cm segment of the left circumflex coronary artery embedded into the myocardium 5 mm, 4 mm and 2 mm deep at maximum thickness, respectively (Figs. 1-3).

The coronary arteries were patent without atherosclerotic lesions or thrombi. Few dark mottling patches were detected in the ventricular septum and the anterior wall of left ventricle. Right lung weighed 500 g, left lung 460 g, with smooth surfaces and mild degree of congestion and edema. Gastric content was composed of approximately 200 ml of food material and gastric mucosa showed few areas of congestion. There was no evidence of pulmonary thromboembolism. The remainder of the autopsy was otherwise unremarkable.

Microscopic examination of the brain displayed generalized edema and congestion. The lungs showed focal congestion and edema. The heart demonstrated some areas of congestion, mild hypertrophy of myocytes, and focal myocardial and interstitial fibrosis with mild fatty infiltration around the arteries (Fig. 4). The gastric mucosa revealed some areas of congestion. The remaining visceral organs were otherwise unremarkable.

**Figure 1.** A gross section of the right coronary artery showing a segment of the artery embedded into the myocardium 5 mm deep at maximum thickness.

**Figure 2.** A gross section of the left anterior descending coronary artery showing a segment of the artery embedded into the myocardium 4 mm deep at maximum thickness.

**Figure 3.** A gross section of the left circumflex coronary artery showing a segment of the artery embedded into the myocardium 2 mm deep at maximum thickness.

**Figure 4.** A microscopic section of the heart demonstrating some areas of congestion, mild hypertrophy of myocytes and focal myocardial and interstitial fibrosis. (H&E staining, original magnification X 40).
The panel of toxicologic analysis was negative. Fatal arrhythmia triggered by acute transient coronary occlusion of myocardial bridging of the coronary arteries was strongly suggestive of being the mechanism and the cause of death.

**DISCUSSION**

MB is commonly affected a major coronary artery and almost exclusively confined to the left anterior descending coronary artery, predominantly the middle third portion [2-6]. Most bridged arteries are superficial and short, thus being asymptomatic. However, MB can give rise to a temporary systolic narrowing of the bridged artery lumen leading to ischemic event and other cardiac complications. Some studies have shown that compression within the tunneled segment can still persist mostly throughout diastolic phase [16, 17], resulting in a complete or incomplete vessel filling, such a phenomenon termed "milking effect" [18-20]. In addition, the severity of coronary occlusion caused by MB depends upon the intensity of systolic cardiac contractility and the site, depth and length of the tunneled segment [5]. Also, if the contraction is intense and long lasting, it can cause the disruption of underlying endothelium, leading to coronary vasospasm, sclerosis, thrombosis, and resultant myocardial ischemia and infarction. Therefore, MB itself can be an independent risk factor for the development of interstitial fibrosis and myocardial ischemia [21]. The mechanism of death in the present case would have been fatal arrhythmia triggered by acute transient coronary occlusion secondary to the milking effect of the coronary bridging which was severe enough to cause such a consequence due to previously mentioned factors, i.e. a long segment of the major coronary embedded deeply into the myocardium, leading to severe perfusion impairment in the coronary artery and not only the left anterior descending coronary artery but also the right coronary artery and the left circumflex coronary artery were buried within the heart muscle, thus further aggravating their effects on cardiac complications.

Although MB is a congenital malformation, cardiac symptoms usually do not develop prior to the third decade, and the reason for this remains unclear [5]. Those who have the relevant symptoms are most often middle-aged men presenting with typical or atypical chest pain, either related or unrelated to exercise [20, 22]. Most of them also had a trigger factor initiating the cardiac symptoms or a significant cardiovascular risk factor [14]. The trigger factors including strenuous exercise [23], emotional stress [24], certain drugs [25], upper respiratory tract infection [14, 26], acute anemia [8], and full stomach [20] have been reported to initiate cardiac dysfunction, ventricular arrhythmia, conduction block, and sudden death in susceptible persons. However, in case of resultant deaths, some decedents, as in the present case, were younger aged [14, 26]. Since neither ante-mortem signs and symptoms indicating an acute coronary syndrome nor other trigger factors were established in this case, the partial full stomach might have been the only trigger factor inducing a crucial mechanism for the death, and the most likely explanation for this would be the increase of blood flow to the stomach, resulting in a decrease blood flow to the heart and compensatory increase intensity of cardiac contraction.

With regard to "Laitai", it is a sudden unexpected death in healthy young men (20 - 49 years old) who are rural residents of Northeastern Thailand, without any structural causes of death from autopsy findings to explain death [27, 28]. Since the death occurs during sleep at night, it is so called "Laitai" in Thai, or sudden unexplained nocturnal death syndrome. Usually, moaning and gurgling sounds are heard prior to the victim becoming unresponsive and expired. Such deaths can occur in the victim's male siblings for certain generations [29], indicating the genetic involvement and its mechanism of death is attributed to spontaneous ventricular fibrillation caused by idiopathic abnormal autonomic tone [30].

Unlike "Laitai", the deceased and his family were original residents of Bangkok locating in the central part of Thailand and his family did not have a history of such deaths. According to the deceased's father who slept with him that night, he insisted that he had not heard or found anything abnormal relating to the deceased's behaviors during that time. In addition, there was no family history of acute coronary syndrome or established cardiovascular diseases either. Although, the deceased was young without significant health history, there were some structural changes of his heart. The autopsy findings of the deceased showed all coronaries being embedded in the myocardium at length and depth enough to cause significant myocardial complications. The hypertrophy of myocytes, and focal myocardial and interstitial fibrosis with mild fatty infiltration around the arteries were consistent with hypertrophic cardiomyopathy which was the possible cardiac consequences of the deep MB. With these reasons and findings, the genetic testing for channelopathies was not performed in this case. Since MB of the coronaries was the only significant autopsy findings and the toxicologic testing was also negative, the author came to the conclusion that MB was the primary cause of sudden death in the present case.

In conclusion, although there have been some notions that MB may have the same significant clinical morbidity and mortality as coronary atherosclerosis, its resultant death is rare and still controversial. However, the author reported herein a sudden unexpected death in a young man whose cause of death was attributable to MB. Additionally, the present case posed several interesting issues. Firstly, the decedent was too young to suffer a myocardial infarction, having no other risk factors for coronary artery disease or thrombus, except for MB of the
coronaries. Secondly, this case together with the previously reported cases strongly suggests that MB might have been a possible cause of death, rather than just a benign condition, especially in cases of the deep and long MB variant. Thirdly, unlike previously reported such deaths which almost exclusively involved in a single bridge, the present case contained three bridges. Finally, since MB as a possible cause of death has not yet been received as a uniform notion, a complete postmortem examination in conjunction with thorough laboratory investigation must be carried out to exclude other possible causes of death as well.

Conflict of interest. The author declares that she has no conflict of interest concerning this article.

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