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CASE REPORT

Anomalous Single Coronary Artery and Wolff Parkinson White Syndrome

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ABSTRACT
The coronary anomalies are very uncommon to be seen in clinical practice. These anomalies include; a right coronary artery arising from the left coronary cusp; a left coronary artery originating from the right coronary cusp; a circumflex artery having a separate origin from either the left or right coronary cusp or from right coronary artery and the presence of a single coronary artery either originating from the left or right coronary cusp. The presence of a single coronary system is rare.The presence of a single coronary system originating from right coronary cusp is extremely rare .The association of a single coronary system origination from right coronary cusp and Wolff- Parkinson-White is strikingly rare and may not be reported before.
We are reporting a case of a 38-year-old female patient who presented to the hospital with recurrent typical chest pain and palpitations. Coronary angiography revealed a single coronary circulation arising from the right coronary cusp with co-existing Wolff -Parkinson -White Syndrome (WPW). The patient's electrocardiography, computed tomography and coronary angiography results are discussed. We would like to highlight the rarity of this unique coexistence as well as highlighting the risk of sudden death in Wolf- Parkinson-White might not be only due to the fatal arrhythmia but also secondary to an associated adverse consequence of the anomalous coronary artery.

Key words: Anomalous coronaries, Single coronary trunk, Preexcitation syndromes, Wolff-Parkinson- White Syndrome, Sudden death.

Introduction

Single coronary artery(SCA) is a rare anomaly in which an entire coronary system arises from a single coronary sinus. As a lone discovery its incidence is 0.024% to 0.066% in the general population undergoing coronary angiography [1, 2]. Wolff –Parkinson-White syndrome is defined as a congenital condition involving abnormal conductive cardiac tissue between the atria and the ventricles that provides a pathway for a reentrant tachycardia circuit, in association with supraventricular

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tachycardia. The coexistence of both an accessory pathway and coronary artery anomalies have not been found in the literature. However, an association between an accessory pathway and coronary sinus anomalies may suggest an embryologic link [3]. The risk of sudden death in patients with WPW or Single coronary artery circulation had been highlighted in the literature. WPW is diagnosed in most patients from an ECG done in the routine work up for different reasons while anomalous coronary arteries only diagnosed after coronary or CT coronary angiography done as a work up for ischemic heart disease. We don't investigate every patient with WPW with coronary angiography and so this association could be under reported. This association needs to be studied in the future.

Case report

A 38-year-old Sudanese female presented complaining of recurrent attacks of central chest pain and palpitations. The patient had strong family history of Ischemic Heart Disease (IHD) and myocardial infarction.

On clinical examination, the patient appeared afebrile with a pulse of 90bpm and a blood pressure of 130/70 mmhg. The remaining systemic examination was unremarkable.

Investigations revealed slightly raised creatinine kinase levels with normal troponins. An electrocardiogram (Figure 1) was done and revealed short PR interval, 114ms, prolonged QRS duration, 164 ms, delta waves in all pericordial leads, changes compatible with Wolff Parkinson White Syndrome, Type A, as well as T wave inversion in leads V1-V4.

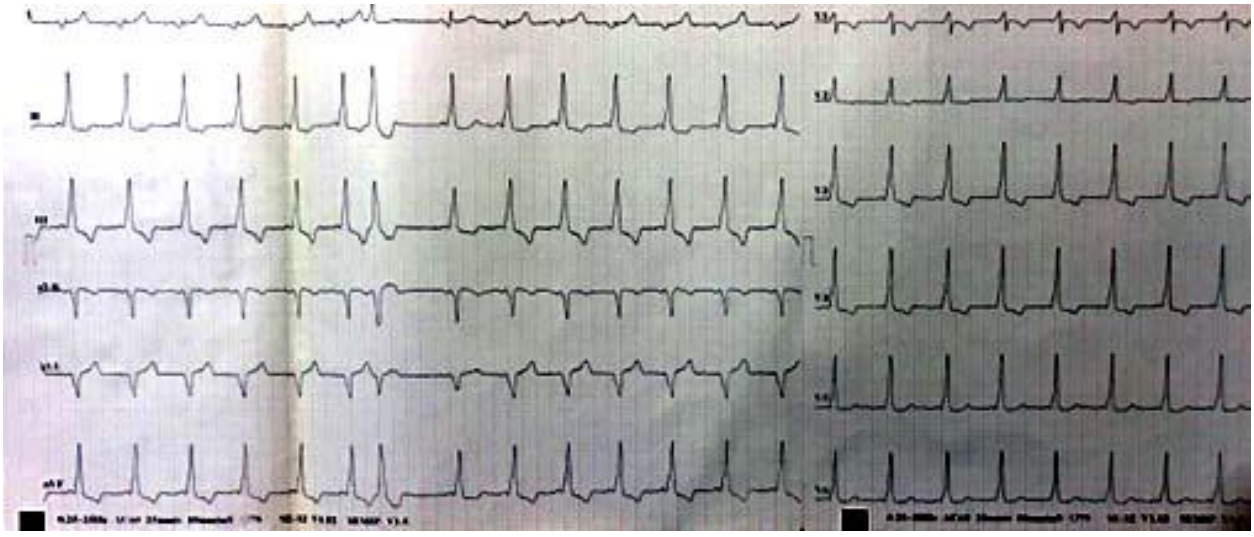

Figure 1: Showing Delta ways compatible with WPW type A.

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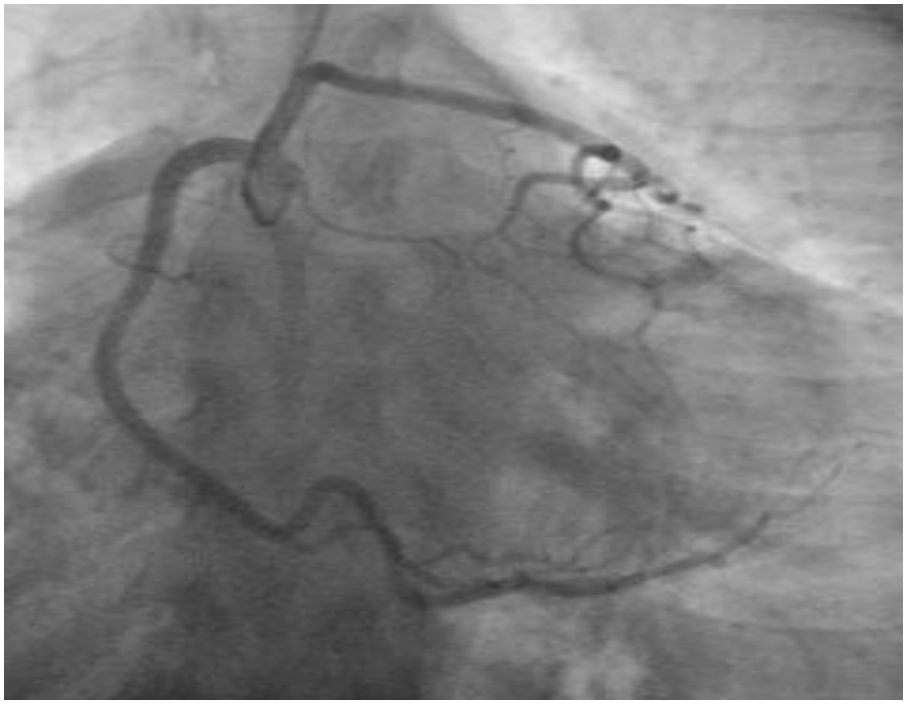

Figure 2: CAG showing single origin of all coronary arteries

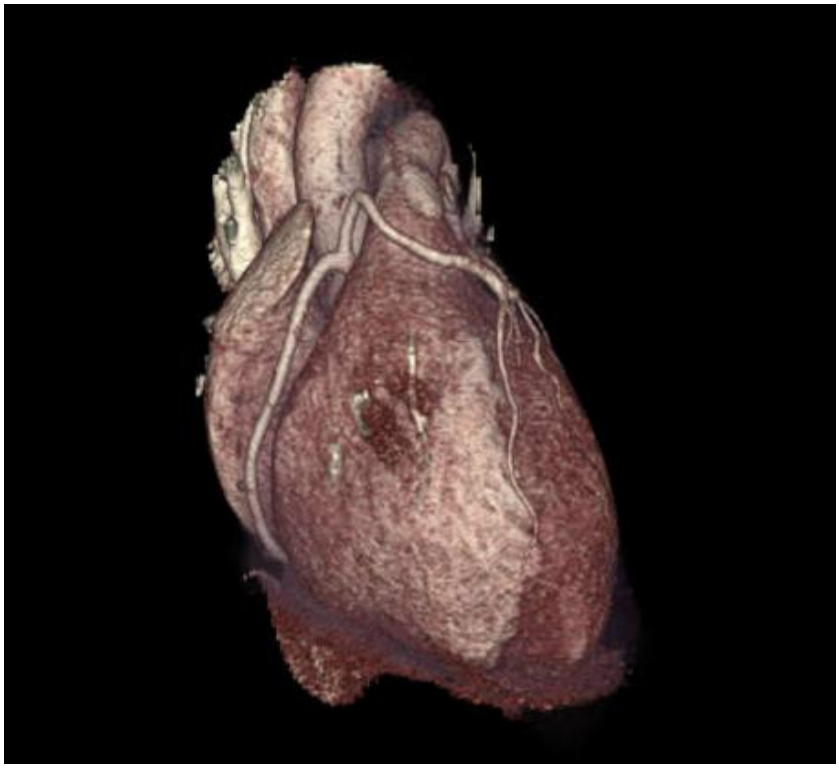

Figure 3: CCT Showing LCA Originating from Right Coronary Cusp branching in the RCA and LAD

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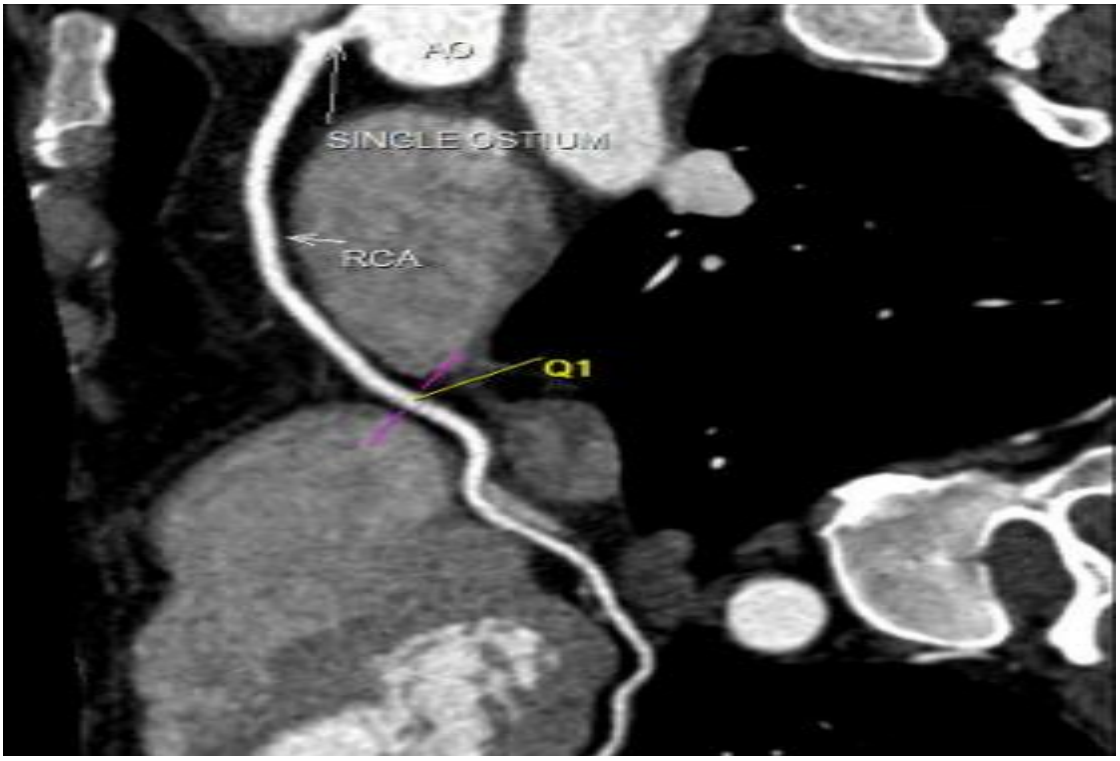

Figure 4: Showing benign course of RCA

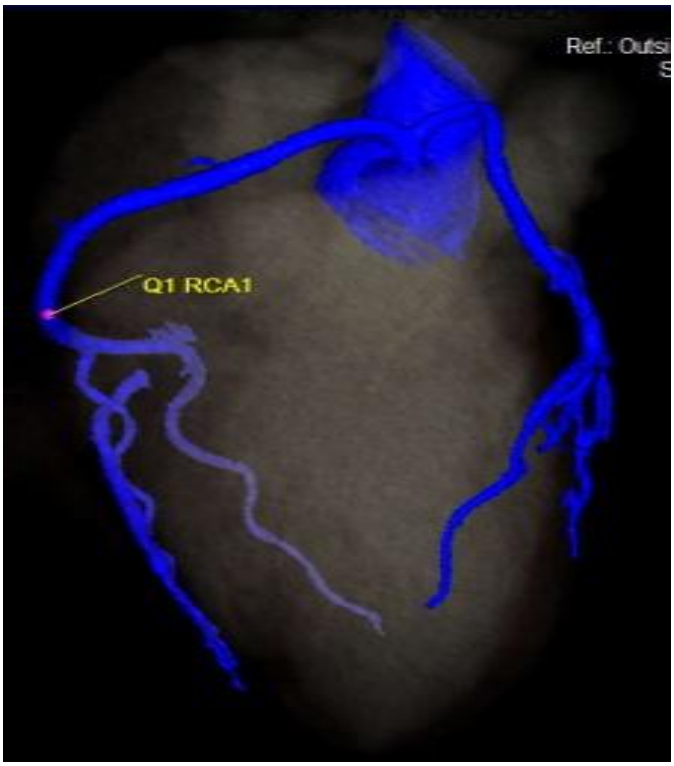

Figure 5: Showing Left main branching into RCA and LAD

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The patient was admitted to rule out Acute Coronary Syndrome. Echocardiography was done and revealed good left ventricular systolic and diastolic function as well as trivial mitral and tricuspid regurgitation. The patient was taken to Coronary angiography (CAG) Figure 2, indicated by single ectopic origin for all coronary arteries as a single coronary system arising from the right coronary cusp was identified, CT angiography and Cardiac CT (CCT) were recommended to exclude a malignant course i.e. compression of LCA between Aorta and pulmonary artery .

Figure 3, CCT revealed a benign course of the single coronary artery and its branches.

The patient continued to be symptomatic with palpitations secondary to supraventricular tachycardia and was subsequently referred for the ablation of the accessory pathway which was successfully done.

Discussion

Anomalous origin of the entire coronary system from the right sinus of Valsalva is a very rare anomaly. The course of the arteries once they originate from the right sinus is variable. Its clinical importance lies mainly in the course of the artery in relation to the great vessels, namely the aorta and pulmonary vessels. The most two common courses, anterior and posterior routes are believed to be benign [2]. Patients with an intra-arterial left main are considered to have a malignant course as sudden death is frequently observed in such patients. This is due an increased risk of coronary obstruction secondary to aortic or pulmonary artery (PA) dilation during exercise. Contrast CT Coronary angiography has become a complimentary tool in identifying the origin, characterizing the proximal course of the vessels relative to the aorta and PA and therefore can aid in the clinical decision making [4].

ongoing chest pain, ECG changes and strong family history of IHD. CAG showed a single coronary artery, the Left Main (LCA), originating from the right coronary cusp and giving rise to the Right Coronary Artery (RCA), (Figure 4,5) and Left Anterior Descending (LAD),

Reviewing such a case brings about the risk of Coronary Artery Disease (CAD) in such patients. Although coronary anomalies are rare but they are still at risk of developing coronary artery disease and they should be treated the same way as CAD in native coronaries[5]. Moreover, Hutchins et al., has advocated that the unusual take off and the tortuous course of the proximal portion of the anomalous coronary predisposes it to accelerated atherosclerosis [6].

Wolff Parkinson white Syndrome is a rare entity and mostly benign. The diagnosis is made in most cases after a routine ECG reading by expert.However sudden cardiac death had been reported in Patients with WPW and is usually caused by the propagation of the arrhythmias to ventricular fibrillation.Due to the risk of sudden death the management of WPW had been revolutionized by the introduction of Radiofrequency Catheter Ablation. Catheter ablation has been proven as very effective, safe and first line therapy for patients with symptomatic WPW syndrome [7]. However, its implication in asymptomatic patients with WPW pattern on ECG remains controversial. Asymptomatic patients may

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require invasive risk stratification and possible catheter ablation.

The association of a single coronary circulation and WPW was not found in the literature, hence this is the first case described.This case raises a few questions; could such a unique coexistence put the patient at even higher risk of sudden death than what have been postulated from only arrhythmia? Is there an embryological link between a single coronary artery circulation and an accessory pathway? Due to paucity of literature and prior research the answers to the above questions remain a mystery. Could all patients with Wolf –Parkinson-White Syndrome have this association and are not diagnosed as they are not investigated in this direction? Could sudden death, in cases of Wolf-Parkinson- White Syndrome be due to a malignant anomalous coronary course consequence as well as fatal arrhythmia? However, this case instigates an important issue; should coronary angiography and/ or CT coronary angiography be standard and paradigm investigations in patients with WPW? Early diagnosis of a coronary anomaly with a malignant course might prevent sudden death in such patients more than catheter ablation of the accessory pathway.

Conclusion

Single coronary arteries are rare but have important clinical prognostic implications putting the patient at high risk of myocardial ischemia, infarction and sudden death. The association of a coronary artery anomaly with WPW is a peculiar association and this case is the first reported in literature. A malignant Course of an anomalous coronary artery put such patients at even a higher risk of sudden death. Both WPW and malignant coronary course can be treated, once recognized, and hence dramatically curtailing the associated risk of sudden death. These facts warrant further research to identify this important association and conjoined risks. If such an association is proven to be true, then every case of wolf Parkinsonism should be investigated by at least CT coronary angiography to exclude the malignant course.

Acknowledgements

I, Dr Abdalla Eltayeb, on behalf of other authors certify that the manuscript is original. I undertake full responsibility for any ethical issue of medical activity or practice described in the manuscript.

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