

Case-report**Subarachnoid Hemorrhage due to Ruptured Anterior Communicating Artery Aneurysm in a Child with Coarctation of Aorta**

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Article Received: 2nd February, 2022; Accepted: 18th April, 2022; Published: 30th June, 2022DOI: <https://doi.org/10.3126/jonmc.v11i1.46166>**Abstract**

Intracranial aneurysms are sometimes associated with other vascular anomalies in extracranial location. Coarctation of aorta, a congenital vascular lesion can be associated with intracranial aneurysms. In patients with coarctation, evaluation of intracranial vasculature is essential. We encountered a 12-year kid with subarachnoid hemorrhage. On further evaluation, she had anterior communicating artery aneurysm. On further evaluation for secondary causes of aneurysm, she had coarctation of aorta. She was surgically managed successfully by clipping of the aneurysm.

Keywords: *Aneurysm, Coarctation of aorta, Subarachnoid hemorrhage***Introduction**


Intracranial aneurysms (IAs) are present in 1-2% of the population [1]. They are sometimes associated with some inherited disorders like autosomal dominant polycystic kidney disease, Ehler-Danlos Syndrome, Coarctation of aorta (CoA) etc. Intracranial aneurysm is seen in 10.5% of cases of coarctation of aorta [2]. Such association leads to increased morbidity such as an increased risk of rupture of the aneurysm. There have been many reported cases of ruptured IA with CoA. Here we present one such case managed in our hospital in addition to reviewing the pertinent literature.

Case report

A 12-year-old female child presented with acute onset headache and dizziness for

threedays associated with vomiting. She had no history of fever, loss of consciousness or seizure. Her antenatal and perinatal period were uneventful. She had no significant past or family history. On examination, the Glasgow Coma Scale was 15; both pupils were round regular and reactive to light. Higher mental function was intact and neurological examination was unremarkable. Nuchal rigidity was present. Cardiovascular examination showed blood pressure of 170/100 mm of Hg in both upper limbs and 90/60 in both lower limbs. First and second heart sounds were normal and systolic murmur was heard in the interscapular region on the back. Other organ system evaluation was normal.

Plain computed tomography (CT) scan of head (Figure 1) revealed subarachnoid hemorrhage

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(SAH) in the anterior interhemispheric region. Head CT angiography (Figure 2) showed a saccular aneurysm in the anterior communicating artery (ACoM) measuring 0.34 cm X 0.37 cm with the neck diameter of 6 mm. An echocardiography showed tight coarctation of aorta with normal left ventricular ejection fraction. Stenosis in the descending aorta measuring 0.4 cm X 0.3 cm at the level of T5/T6 with at least three saccular aneurysms adjacent to the site of co-arcuated descending aorta measuring 0.9 cm X 0.4 cm (neck 0.15 cm), 1 cm X 1 cm (0.5 cm) and 1.5 cm X 1.2 cm (0.7 cm) was detected on subsequent CT aortography (Fig. 3).

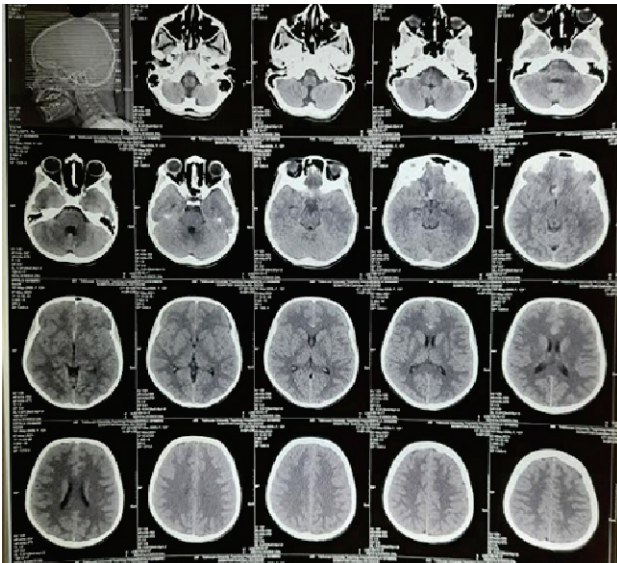


Figure 1: Computed tomography scan of head showing diffuse subarachnoid hemorrhage mainly localized to anterior interhemispheric fissure (arrow).

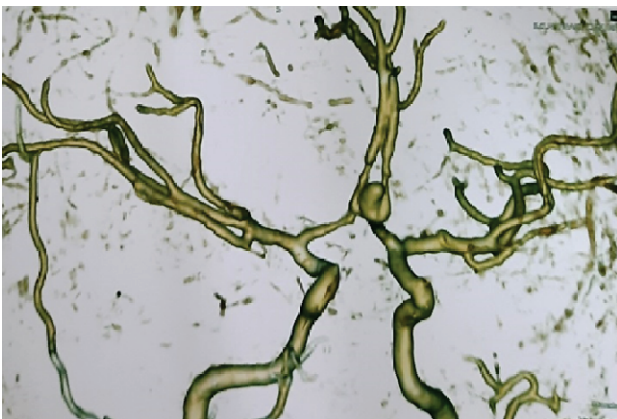


Figure 2: Three dimensional computed angiography showing anterior communicating artery aneurysm (arrow)

The patient underwent microsurgical clipping of the intracranial aneurysm via the right pterional craniotomy with application of 7 mm titanium permanent clip. Intraoperative finding was

fenestrated ACoM artery and a large saccular aneurysm at left A1 and ACom junction. In view of co-existing Coarctation special care was taken intra op to manage hemodynamics.

A post-operative non-contrast CT (Figure 4) was significant for mild pneumocephalus only. The patient made an uneventful recovery post operatively and was discharged on the 9th post operative day in a stable condition. Cardiothoracic surgery team has planned for management of the CoA once she fully recovers from the surgery.

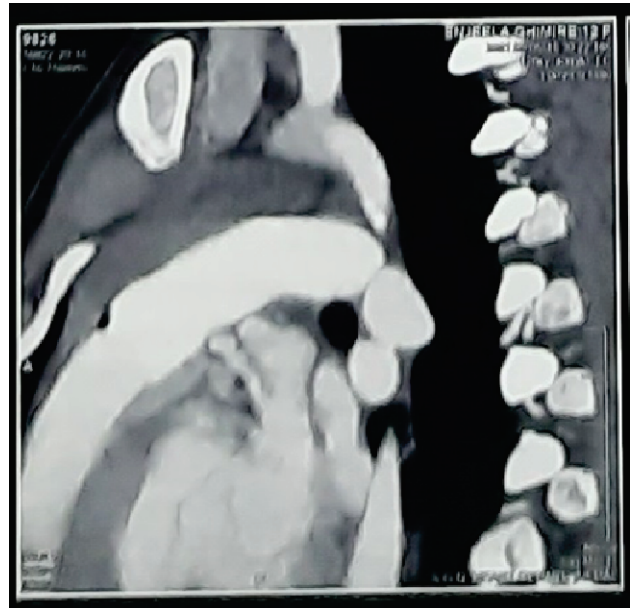


Figure 3: Computed tomography aortogram scan showing stenosis in the descending aorta (arrow)

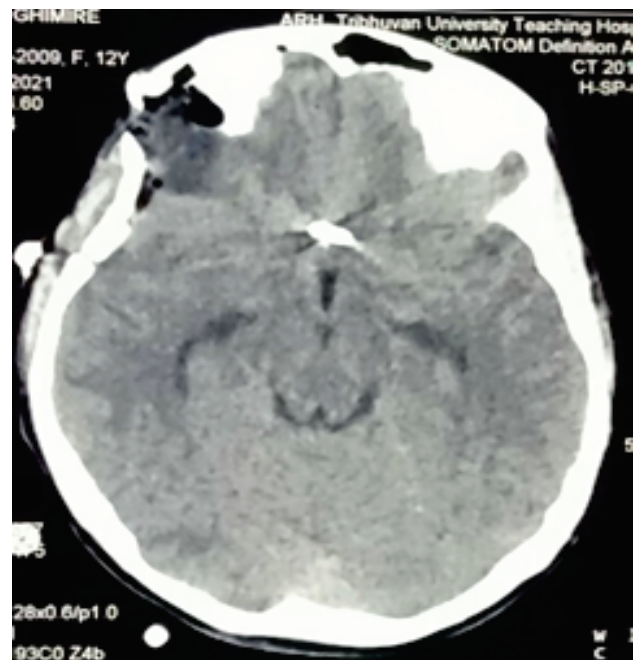


Figure 4: Computed tomography scan of the head on post operative day 1 showing post-operative changes with mild pneumocephalus and clip in position

Discussion

The CoA is a common congenital aorta abnormality occurring in 4 per 10 000 live births [3]. Neurologic Complications Associated with Congenital Stenosis of the Isthmus of the Aorta was described by Woltman HW et.al. in 1927 [4]. Despite adequate relief of aortic arch obstruction, patients with CoA suffer from increased morbidity and reduced long-term survival. One of the causes for premature morbidity and death is SAH secondary to ruptured IAs, which occurs at a significantly younger age than in the general population. By middle age, 10% to 13% of patients with CoA, compared with 3% to 7% of the general population, have an IA on screening magnetic resonance angiography (MRA) [2,5,6]. The 2018 American heart Association (AHA) guidelines for the care of the adult with congenital heart disease (CHD) suggest that screening for IAs by a MR Angiography or CT angiography may be reasonable in adults with CoA [7]. In contrast, routine imaging to assess for IAs in asymptomatic patients with CoA is not recommended in the 2020 European Society of cardiology (ESC) Guidelines [8]. However, the AHA recommends screening patients with CoA for IA and suggests screening in ages 10 and 20 or in 10, 20, and 30 years would extend the life and be cost-effective [7].

The patho-physiological basis of the association between IAs and CoA is still unknown but two hypotheses are entertained. It is either hereditary or due to uncontrolled hypertension. The incidence and the risk of rupture of the IA depend on the study population and morphology of the aneurysm. Owing to their direct origin from the pre-coarctation segment of aorta, cerebro-vascular arteries are most likely to be affected by the hemodynamic changes occurring in this part of the aorta. Secondary to the increased resistance to outflow, increased aorto-cranial pressure gradients have been demonstrated by several authors correlating to the severity of stenosis [9-11]. The CoA is a connective tissue disorder accounting for 10% of the cases of congenital heart disease.¹² First, hypertension is often a constant feature in CoA patients. Second, hypertension has been considered as a major risk factor for the development of IAs even without CoA [13,14].

Despite an increased prevalence of IAs in hypertensive patients, the role of hypertension in this context has been questioned by many authors. Mc Cormick and Schmalstieg found no relationship between arterial hypertension and IAs in their review of 250 patients [15]. In a similar

study conducted in 212 cases, Andrews and Spiegel did not find any significant elevation in the blood pressure in patients with IAs compared with the general population. However, studies supporting hypertension as a risk factor for the development and rupture of IAs outnumber the studies refuting it [16]. Hypertension cannot be the sole contributor. Hudaoglu et al [17] raised a question on hypertension alone being a factor in the etiopathogenesis of IAs in CoA patients. The fact is further supported by the observation that IAs may develop or rupture even in normotensive patients, years after the repair of CoA [18]. They believed that the coexistence of these malformations with IAs is secondary to a common developmental error playing a role in their etiology. Embryologically, the heart, aorta, and cervico-cephalic arteries all share a common origin from the neural crest. After recognizing the association between a variety of congenital heart diseases and IAs, Schievink et al attributed the occurrence of IAs in CoA patients to the developmental errors of neural crest resulting in abnormal vessel wall collagen [19].

Apart from being a known risk factor for the formation of IAs, CoA has also been associated with increased incidence of IA rupture (4.8%) [18]. Clinical manifestations of patients with CoA vary with different location and degrees of lesions. The patient may present with excessive pressure difference between limbs, headache, dizziness, numbness, weakness, pain and intermittent claudication caused by hypotension in lower limbs [20].

The disease is mainly found in young and middle-aged persons and rarely above 50s thus affecting the productive period of life. Our patient presented with an intracranial bleed and was detected to have CoA on further studies. In this paper we report a rare case with CoA complicated with ACoM aneurysm and briefly describe the management of the ruptured IA. Therapies for CoA include balloon or stent angioplasty and prosthetic vessel replacement. As the risk of rupture in IAs associated with CoA is high so clipping aneurysm or interventional embolization will be an effective treatment. Our case is the first case of CoA with SAH reported from Nepal. This has been reported to highlight the fact that in young patients with SAH, a thorough search is necessary to look for any predisposing conditions for aneurysm formation

Conclusion

Coexistence of IAs and CoA is rare. A high index of suspicion is needed to look for CoA in patients



with a SAH especially in young patients. An aggressive management approach helps in dealing patients with ruptured IA with coexistent CoA successfully.

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Conflict of interest: None

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