Percutaneous coil embolisation of congenital hepatic arteriovenous malformations in two patients with congenital heart disease and review of the literature

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ABSTRACT

Congenital hepatic arterio-venous malformations (AVM) are rare vascular anomalies and have rarely been reported in the presence of congenital heart disease. The reported cases are mostly hemangiomas fed either by the hepatic artery itself or by one of its branches. We present two unique hepatic AVM cases in the presence of congenital heart defects in which the AVM was not fed by the hepatic arterial system. Transcatheter coil embolisation was successfully carried out in both of them by using non-detachable Gianturco coils. Complete occlusion was achieved without any sequel.

Keywords: Arterio-venous Malformation; Trans-Catheter Embolisation; Congenital Heart Disease

1. INTRODUCTION

Hepatic vascular malformations comprise about 10% of all cases of abnormal vascular connections within the hepatic parenchyma [1]. These are further classified into fast-flow (AVMs, arterio-portal fistulas), slow-flow (porto-systemic shunts, venous lymphatic malformations) and combined forms [2]. Hepatic AVMs should be differentiated from the benign hepatic vascular tumors (HVT) when an AVM can occur within the tumor mass itself. Unfortunately, the terms hemangioma and arterio-venous malformation (AVM) have been confused or used interchangeably in the literature [3]. Histologically, HVT are derived from mesenchymal tissue elements and are classified as hemangioendothelioma (HE) or cavernous hemangioma (HC) [4,5]. More recently, these tumors are classified into infantile hemangioendothelioma (IHE) and arteriovenous malformations (AVM)

[6,7]. Hemangioendothelioma progresses in the first year of life and then regresses spontaneously. On the other hand hepatic AVMs (in the absence of HVT) result from abnormalities in the formation of blood vessels that shunt blood through direct arterio-venous connections without abnormal neoplastic tissue between the anomalous vessels. So AVMs are neither tumors with growth potential, nor capable of regression [1,6] but have a higher and not well documented mortality rate [8]. On the contrary the HVT has an age related tendency of malignancy [5].

Hepatic AVMs are diagnosed either incidentally on abdominal sonography for other reasons or may result in congestive heart failure (CHF) due to significant intra-hepatic shortcircuiting of blood. Ultrasonography (US) and magnetic resonance imaging (MRI) are useful diagnostic tools [9,10] but selected angiography is an optimum diagnostic modality. Hepatic AVMs have been reported in association with hemangiomas, and hereditary hemorrhagic telangiectasia but rarely been reported in association with congenital heart defects. We encountered two such cases which are briefly described.

2. CASE 1

This young infant presented to our services at two months of age (weight = 5 kg) with complaints of bluish discoloration of lips and nails since early life. Pertinent physical examination features included significant oxygen desaturation to 58% on room air, normal SI, single SII and systolic ejection murmur III/VI at the left upper sternal border. He was finally diagnosed as a case of complex congenital heart disease in the form of double inlet left ventricle, hypoplastic right ventricle, transposed great arteries, large ventricular septal defect, and severe sub-pulmonary/pulmonary stenosis. Palliative balloon atrial septostomy was carried out for adequate inter-atrial mixing, which improved the oxygen saturation to 80%

on room air. He was then discharged in a stable condition.

At the age of two years (weight = 12.5 kg) he underwent diagnostic cardiac catheterization for hemodynamic assessment and delineation of pulmonary artery anatomy as a pre-Glen assessment. Descending aortograpgy, done to rule out aorto-pulmonary collaterals, revealed incidentally a significant hepatic AVM. The
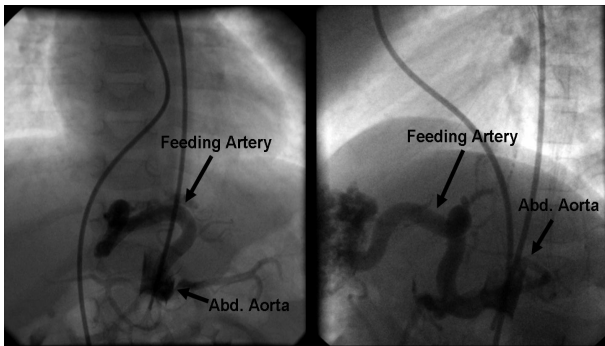

Figure 1. Abdominal aortogram in straight antero-posterior and lateral projections, showing feeder artery arising from abdominal aorta and supplying the arterio-venous malformation. *Abd* = abdominal.
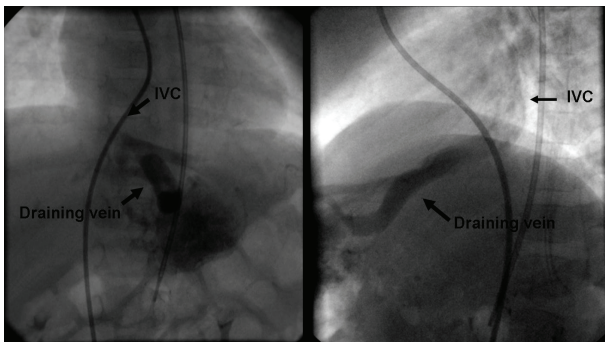

Figure 2. Abdominal aortogram in straight antero-posterior and lateral projections, showing the malformation draining through a vein (draining vein) to the hepatic portion of inferior vena cava (IVC).
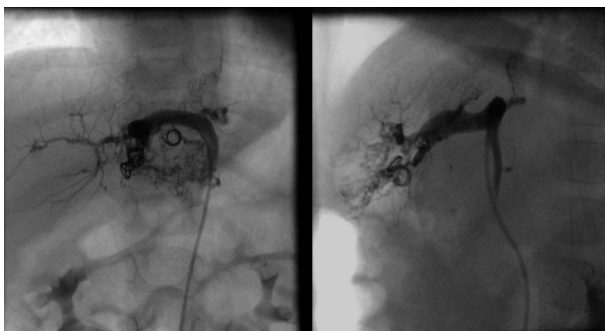

Figure 3. Selective angiogram in the arterio-venous malformation (AVM) in straight antero-posterior and lateral projections after coil embolisation, showing almost complete occlusion.

feeder artery was seen arising from the abdominal aorta **(Figure 1)** and the malformation was seen draining into the hepatic portion of inferior vena cava **(Figure 2)**. The AVM was occluded by deploying multiple (four coils of 5 mm and one coil of 3 mm diameter) non-detachable Gianturco coils (Mreye embolisation coils; Cook: William cook, Europe) retrogradely through a 4-French end-hole catheter. No complications were encountered and post occlusion angiography confirmed near complete occlusion **(Figure 3)**. At the same time he underwent lateral tunnel fenestrated Fontan procedure. Lately he was seen in the out-patient clinic at the age of 3 years (weight = 15.5 kg). He was doing fine without significant complaints and his oxygen saturation ranged between 88-90% on room air.

3. CASE 2

This was the product of 35 week gestation, born (weight = 2.1 kg) to a 35 year old mother with good Apgar score at one and five minutes. Her physical features were consistent with Down syndrome **(**confirmed by chromosome analysis). At day one of age she developed congestive heart failure and was electively intubated because of significant respiratory distress. Physical examination revealed apparent distress with retractions, heart rate of 200/min, respiratory rate of 78/min, and liver palpable 4 cm down the right costal margin. Chest roentgenogram showed cardiomegally and increased vascular markings. Echocardiography confirmed her having juxta-ductal coarctation of the aorta (gradient = 20 mmHg). Incidentally, an abnormal flow reversal was also seen at the transverse arch level in the absence of aortic insufficiency. The reason for this flow reversal could not be ascertained. She was taken to the catheterization laboratory in view of looking for the reason of this abnormal flow reversal in the aortic arch and possible balloon angioplasty of coarctation. Pull back of catheter from ascending to the descending aorta revealed a pressure drop of only 14 mmHg from ascending to descending aorta. Aortic angiogram revealed a large aneurysmal and a bit tortuous vascular channel (maximum diameter of 3 mm) arising from the right internal mammary artery, supplying the right lobe of the liver **(Figure 4)** and draining to the hepatic portion of IVC **(Figure 5)**. Small Patent ductus arteriosus (PDA) was also seen. This vascular malformation was successfully occluded by deploying multiple (4 coils of 3 mm and one coil of 2.5 mm diameter) non-detachable Gianturco coils (Mreye embolisation coils; Cook: William cook, Europe) retrogradely through an end-hole catheter. Repeat angiography confirmed complete occlusion of the AVM **(Figure 6)**. Subsequently the patient was successfully extubated and

discharged in a stable condition.

She was followed regularly in the out-patient clinic without much complaint till the age of 16 months, when she started having shortness of breath on feeding and on regular activities. Physical examination revealed absent femoral pulses and a non-invasive blood pressure difference of 45 mmHg between the upper and the lower limbs. Echocardiography confirmed the development of significant coarctation with a gradient of 40 mmHg
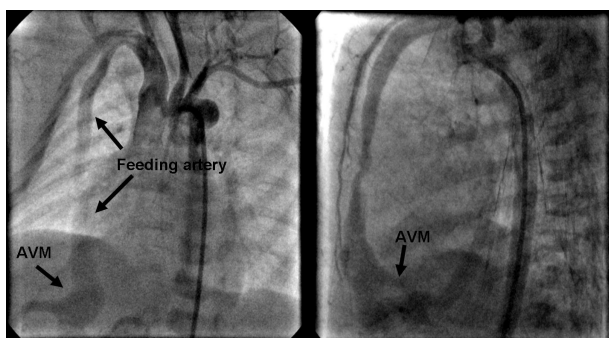

Figure 4. Aortic angiogram in LAO 15/Cr 20 and lateral projections showing dilated and tortuous internal mammary artery, feeding the arterio-venous malformation (AVM) to the right lobe of the liver.
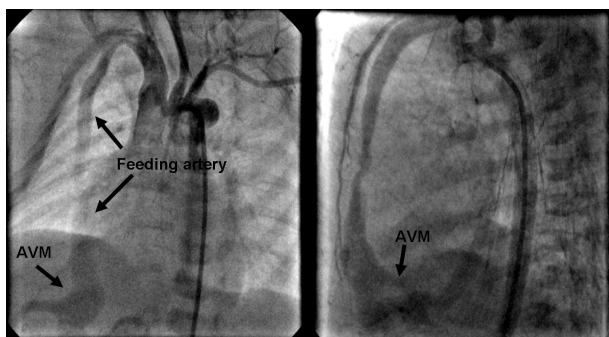

Figure 5. Selective angiogram in the internal mammary artery in straight antero-posterior and lateral projections showing the malformation draining through the hepatic portion of the inferior vena cava (IVC). A portion of the right atrium (RA) is also opacified.

Figure 6. Selective angiogram in the internal mammary artery in straight antero-posterior and lateral projections after the coil embolisation of the malformation, showing complete occlusion.

across it. Successful balloon angioplasty of the coarctation was carried out at that time. The PDA had already been occluded spontaneously.

Lately she was seen in the out-patient clinic at the age of 34 months (weight = 17 kg). She was doing fine without any complaints and there was no significant blood pressure difference between the upper and the lower limbs.

4. DISCUSSION

Hepatic AVM can be of three types: a direct communication between a systemic artery and a hepatic vein (hepatic AV fistula); a communication between the hepatic artery and the portal venous system (hepatoportal fistula); multiple arterio-venous miocrofistulae as part of the hereditary hemorrhagic talengiectasia or hemangioma. There have been many reports in the literature discussing all aspects ranging from diagnosis to treatment options for hepatic AVMs, but most of the reports have included hemangiomas and other proliferative disorders under this condition. Ours are true AVMs, (hepatic AV fistula) in the sense that there is a solitary feeder artery supplying the AVM and then draining through a venous channel to the hepatic portion of inferior vena cava without significant capillary phase, resulting in high cardiac output state. Also an association of hepatic AVM with cyanotic congenital heart disease has never been reported before. Keeping in view the importance of the condition and its new association with the congenital heart disease, some aspects are discussed below with a brief review of the literature **(Table 1)**.

5. DEVELOPMENT OF AVM

Beside hepatic artery, the liver can be supplied by a number of extra-hepatic arterial sources including, right or left inferior mesenteric arteries, intercostal arteries, superior mesenteric artery, celiac axis, left gastric artery and the adrenal arteries. The extra-hepatic arterial supply has been demonstrated in about 96% of patients studied by balloon occlusion of the main hepatic artery [11]. These extra-hepatic arteries remain dormant under normal conditions. However, one or more of these can become enlarged in patients with hepatic vascular malformations or tumors. This was true in our first case as the course of the abnormal feeding artery to the AVM represented most probably one of the minor twigs arsing from the abdominal aorta. But in our second case the feeding artery represented a dilated and tortuous right internal mammary artery arising from the innominate artery. This was an unusual finding as the left or the right internal mammary arteries do not normally contribute to the extra-hepatic arterial supply. To our knowledge internal

mammary artery feeding a hepatic AVM has not been reported so far. Moreover, though the innominate artery is derived from the neural crest, we did not encounter any other neural crest anomaly in our case. Finally an association of neural crest anomaly with Down syndrome is not documented and needs further elaboration.

Table 1. Review of the literature.

Study	Pts	AVM type	Presentation	Association	Treatment	Results/complications
Nielsen *et al*. [12]	3	Cerebral = 1	CHF	NA	NA	NA
		Placental = 1	CHF	NA	NA	NA
		Pulmonary = 1	CHF	NA	NA	NA
Israel *et al*. [13]	1	Retro-peritoneal	CHF	NA	Emb with Evalon	avascular mass
Favilli *et al*. [14]	4	Cerebral = 1	CHF	NA	None	Died
		Hepatic = 2	CHF	NA	Medical	Improved
		Pulmonary = 1	CHF	NA	embolisation	Died months later
Subramanyam *et al*. [15]	1	Hepatic	CHF	None	coil embolisation	Resolution
Chavan *et al*. [16]	15	Hepatic vascular	Abdominal = 5	HHT	Emb with PVA steel/platinum microcoils	Cholangitis = 1
			CHF = 11	HHT		Cholecystitis = 3
			Portal HTN = 4	HHT		One death
			Cirrhosis = 1	HHT		
Costa *et al*. [17]	1	Hepatic	CHF	NA	Coil embolisation	Resolution
Mejides *et al*. [18]	1	Hepatic Vascular	Fetal CHF	NA	Hydrocortisone in UV	Resolution
Knudson *et al*. [19]	156	Cerebral = 86	CHF (67%)		Medical and surgical	Mortality = 67%
		Hepatic = 61	CHF (48%)		Medical and surgical	Mortality = 55%
			Abd. Mass = 67%)			
		Pulmonary = 14	CHF (67%)		Medical and surgical	Mortality = 43%
			Cyanosis (43%)			
Chavan *et al*. [20]	5	Hepatic vascular	CHF, Portal HTN	HHT	Embolisation (PVA/coils)	Ischemic Cholangitis = 1
Hazebroek *et al*. [21]	2	Hepatic	Abdominal angina CHF,Abd.Distension Portal HTN	Hemangioma	Surgical ligation	Resolution
Valerie *et al*. [22]	1	hepatic	Portal HTN	Small PDA, VSD	Coil embolisation	Resolution
Whiting *et al*. [23]	2	Hepatic	Abdominal pain	HHT	Embolisation with PVA	Both died later
Chen *et al*. [24]	1	hepatic	Abdominal pain	Intrahepatic Biloma	Surgical ligation	Progression of Biloma
Alexander *et al*. [25]	1	hepatic	Persistant Pul. HTN	NA	Coil embolisation	Resolution
Hisamatsu *et al*. [26]	1	Hepatic	Epistaxis, dyspnea	Osler-Weber-Rendu	Coil embolisation	Cholangitis
						Hepatic failure/death
Prokurat *et al*. [27]	17	Hepatic vascular	CHF in most cases	Hemangiomas	Surgical lobectomy	Resolution
Akita *et al*. [28]	1	Abdominal wall	CHF	Aneurysmal PDA	Surgical ligation	Liver dysfunction

Abreviations. Abd = *abdominal*, CHF = *congestive heart failure*, Emb = *embolisation*, HHT = *hereditary hemorrhagic telangiectasia*, HTN = *hypertension*, NA = *not available*, PDA = *patent ductus arteriosus*, Pul = *pulmonary*, PVA = *pollyvinyl alcohol*, UV = *umbilical vein*, VSD = *ventricular septal defect*.

6. DIAGNOSING AN AVM

A cerebral, pulmonary or retro-peritoneal arterio-venous malformation should be suspected in newborns if a bruit or soft a continuous murmur is heard over the skull, chest or the abdomen respectively. Also, an AVM should be included in the differential diagnosis in a newborn or an infant who had unexplained CHF in the setting of normal echocardiogram. Retrospectively thinking, we believe that an AVM should be suspected in a patient who has relatively high systemic saturation for a single ventricle physiology, as in our first case. A keen review of the literature has also confirmed CHF as one of the main presenting feature in majority of the studies [12-20]. Other uncommon modes of presentation are portal hypertension [16,20,21], abdominal pain, mass, ischemia or distension [16,19-24], primary pulmonary hypertension [25] and bleeding episodes [26]. One of our cases presented in CHF and the other was diagnosed incidentally during diagnostic cardiac catheterization. Echocardiographic evaluation with colour flow mapping and pressure doppler helps further in diagnosing an AVM. Finding of an abnormal flow reversal in the aorta raises the possibility of AVM. Advanced radiological techniques including un-enhanced Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) are the useful tools in confidently distinguishing an AVM from other abnormal vascular connection including hemangiomas. Angiography during cardiac catheterization helps not only in exact delineating the feeder and draining vessels but also can help in trans-catheter occlusion, if indicated.

7. AGE OF PRESENTATION

Antenatal diagnosis of a hepatic AVM has already been reported [18]. Postnataly a hepatic AVM can present during neo-natal period [12,14,15,17,20,25,27,28] or during infancy [6,12,13,19]. It can also present during childhood [21] especially in the setting of hereditary hemorrhagic talengiectasia [29,30] or even during adult life [20,23,24,26]. One of our patients presented during neonatal period (case #2) in respiratory distress due to high output CHF and in the other patient the hepatic AVM was incidentally discovered during cardiac catheterisation. This is in accordance with the study by Prokurat and colleagues [27], who have shown that patients presenting during neonatal period are almost always symptomatic as compared to those who present during infancy.

8. ASSOCIATION OF HEPATIC AVM AND CONGENITAL HEART DISEASE

Hepatic AVMs have been described in association with hereditary hemorrhagic talengiectasias [16,20,23] or intra-hepatic tumors [22,24]. In an adult patient [26] an association of hepatic AVM and Osler-Weber-Rendu Disease has also been described. To the best of our knowledge there has been only one report [21] describing the association of hepatic AVM with a congenital heart disease: a patent ductus arteriosus (PDA) and multiple ventricular septal defects (VSD). Though there is another report describing the presence of a PDA aneurysm in a patient who had abdominal AVM, ours is the first report describing an association of hepatic AVM and cyanotic congenital heart disease. This association is, not only of clinical significance but also has therapeutic implications, as CHD is the major cause of CHF during neonatal period and early infancy and one should keep in mind an AVM as an additional cause of high output CHF in refractory cases who do not respond to usual antifailure therapy or do not show improvement after transcatheter or surgical intervention for the primary cardiac defects.

9. TREATMENT OPTIONS

Therapeutic options for symptomatic AVMs include transcatheter embolisation or surgical ligation of hepatic artery or surgical hepatic lobectomy or liver transplantation in the advanced cases. Embolotherapy with Polyvinyl Alcohol has been described by Chavan *et al.*, and whiting and colleagues [16,20,23], while Ivalon particles were used for the same purposes by Israel *et al.* [13]. While many devices including non-detachable Gianturco coils, detachable coils, Aga vascular plugs, and AGA PDA occluders are now available for embolisation of vascular malformations and fistulas, and can be used for this purpose depending upon the location and the size of the malformations. Transcatheter coil embolisation has been described by many studies also [15,21,25,26]. We opted for Gianturco coils, because of these being available to us in multiple sizes, our expertise to use these because of frequent use and successful results in the literature [21,25,26]. Surgical ligation of hepatic artery has also been described by some [22,24]. We used transcatheter approach through femoral artery for coil embolisation of AVM, though trans-umbilical approach has also been described by Costa and colleagues [17]. All of the studies have described acceptable results with the occlusion of AVM and resolution of the symptoms.

10. COMPLICATIONS AFTER TREATMENT

Cholangitis in one and cholecystitis in another three patients has been described by Chavan *et al.* [16] out of 15 patients (one patient died due to the procedure), while sepsis has been described in 2 patients after the same

procedure by Whiting *et al.* [23]. Both of the patients died 4 and 15 months after the procedure. Intrahepatic cholangitis has been described after coil embolisation of the hepatic artery in one patient by Hisamatsu and colleagues [26]. The patient ultimately died a year later due to hepatic dysfunction. Ischemic cholangitis has also been reported after the same procedure in one out of 5 patients by Chavan and colleagues [20] while one of their patients died 7 months after the procedure due to sepsis. Apart from theses two reports none has reported any complications. Surgical ligation has generally been described as risk free, and only one report has described progression of the disease even after the procedure [24]. We did not encounter any immediate or early follow-up complications, so far.

11. CONCLUSIONS

Hepatic AVMs are rare conditions in children and a high index of suspicion is required to diagnose these especially in those cases who present with either unexplained or refractory high output CHF or in those who had relatively higher systemic saturation in the setting of complex congenital heart disease with single ventricle physiology. Many treatment options are available with acceptable results. Transcatheter coil embolisation is safe and effective for this purpose. Care should be exercised to avoid ischemic changes after either transcatheter embolisation or surgical ligation of the hepatic artery. The importance of these reports is not only the fact that such an association has not been reported before and expands our clinical differential, but also these give us clues to a deeper insight in the mechanism of cardiovascular development.

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