Annals of Case Reports & Reviews

Case Report

doi: 10.39127/2574-5747/ACRR:1000322

Abdelaoui H, et al. Annal Cas Rep Rev: ACRR-322

Aortic Arches' Abnormalities: About 8 Cases

H. Abdelaoui[1*], FZ. Alaoui-Inboui[2], S. Salimi[3], B. Slaoui[4]

[1]Intern in the Pediatric Pulmonology and Cardiology Department, Harrouchi Children's Hospital, Morocco
[2]Pediatrician in Pediatric Pulmonology and Cardiology Department Harrouchi Children's Hospital, Morocco
[3]Professor in the Pulmonology and Pediatric Cardiology Department Harrouchi Children's Hospital, Morocco
[4]Professor In the Pediatric Pneumology and Cardiology Department, Harrouchi Children's Hospital, Morocco

[*]**Corresponding author:** Abdelaoui Hanae, Intern in The Pediatric Pulmonology and Cardiology Department, Harrouchi Children's Hospital, Morocco.

Citation: Abdelaoui H, Alaoui-Inboui FZ, Salimi S, Slaoui B (2022) Aortic Arches' Abnormalities: About 8 Cases. Annal Cas Rep Rev: ACRR-322.

Received Date: April 28th, 2022; **Accepted Date:** May 02nd, 2022; **Published Date:** May 05th, 2022

Abstract

The aortic arches abnormalities include many malformations affecting the aortic arch, its branches or the pulmonary artery. They can lead to complete or incomplete vascular rings development, responsible for tracheo-esophageal compression. It's symptomatology is very varied. We report a series of 8 cases with an average age of 7 months and 7 days. The main symptomatology was wheezing dyspnea and early onset of symptoms, their chronicity and the absence of a free interval between these episodes. Congenital stridor found in five cases in this series. Dysphagia and vomiting are described in two and three cases respectively. The chest angioscan confirmed the aortic arcs abnormalities diagnosis in all cases. Aortic arch Congenital abnormalities are rare and varied, sometimes asymptomatic. This study reminds us that in front of any recurrent respiratory or digestive symptoms in infants, we must also think about this malformative entity.

Introduction

Anomalous aortic arches have been known since the 18th century. Their anatomical description was made in 1737 by Hommel. Their radiological aspects in 1926 by Arkin and those of the right retro-esophageal subclavian in 1936 by Kommerell. On 1939, double arc clinical description by Wolman was underlined [1]. Aortic arcs abnormalities are rare and are for 1% of cardiovascular congenital abnormalities [1,2]. They are the result of 4th arc abnormal development brought together by a common embryological origin. They are a complex group with some forms that are relatively common; others are rare [3,4]. Double aortic arch anomaly is the most common and accounts for 40 to 50% of the symptomatic vascular strips, these abnormalities are responsible for severe functional signs from birth to esophageal compression [1-5]. Other signs are incidental findings during a radiological checkup.

Results

We report a retrospective study of eight cases of aortic arch anomalies. Infants age was between 1 and 20 months with an average age of 7 months and 7 days. The male sex was the most represented in 75% of the cases (6 boys and 3 girls). The clinical symptomatology was mainly respiratory symptoms. All children had multiple wheezing dyspnea episodes without a clear interval. The wheezing appeared early in the first month of life. Congenital stridor was found in five cases while chronic vomiting was found in three cases and dysphagia to solids in two cases. One child had been treated for infant asthma without any improvement. Only one infant had down syndrome. The clinical checkup underlined the presence of respiratory distress and stridor in five cases and hypotrophy in six cases.

Chest X-ray performed in all infants was normal in 5 cases. In three cases it highlighted a bronchial syndrome, an alveolar focus of the middle lobe and upper and middle right lobe alveolar focus. Echocardiography performed on five infants revealed no abnormalities. Esogastroduodenal transit was held in 3 infants and was abnormal in all three cases Figure (1-2) (Table1). Thoracic CT angiography was performed in all infants to confirm abnormalities of the aortic arches diagnosis Figure (3-4-5).

Only one infant had had an esophageal fibroscopy, and it was normal. This digestive fibroscopy was performed because of wheezing as well as chronic vomiting and dysphagia. It aimed to rule out peptic esophagitis complicating gastroesophageal reflux. Six infants have undergone surgery and two others are awaiting treatment. The outcome was favorable with progressive symptomatology improvement in all operated cases.

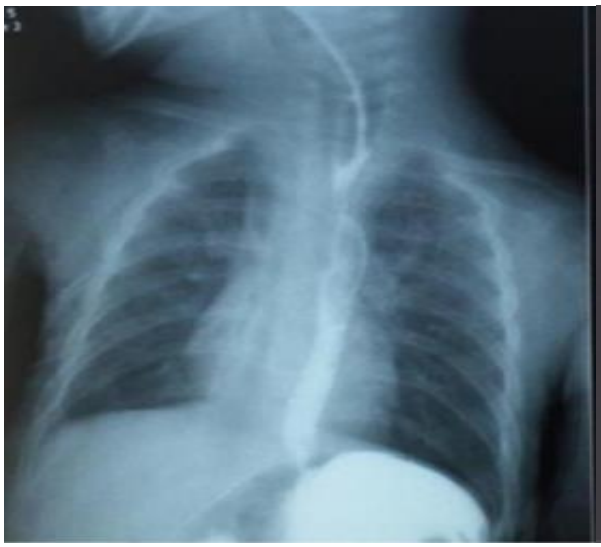


Figure 1: Posterior imprint on the thoracic esophagus in a 9-month-old infant with recurrent wheezing dyspnea highly suggestive of an anomaly of the vascular arches.

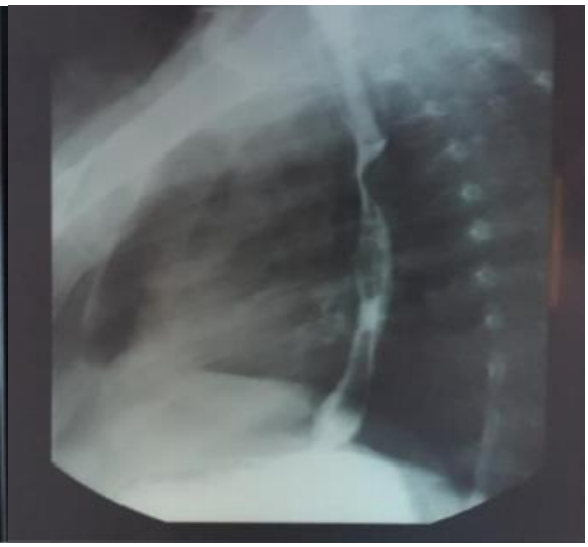


Figure 2: Posterior inprint on the thoracic esophagus of a 13-month-old wheezing infant with stridor and dysphagia.

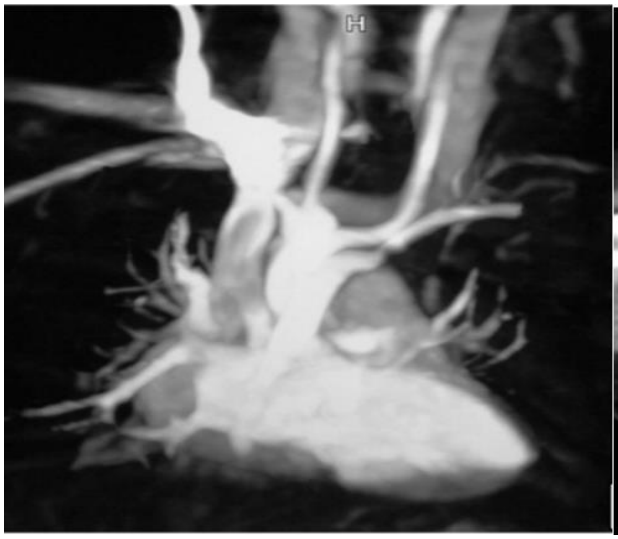


Figure 3: Thoracic CT angiogram: Double aortic arch in 3D.

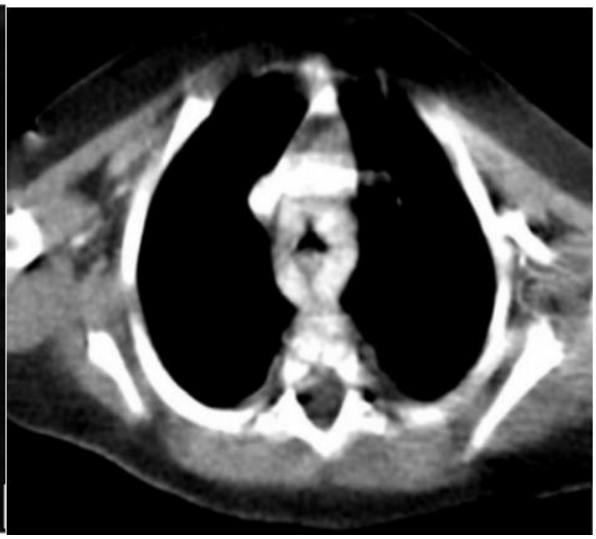


Figure 4: Appearance of double aortic arch encircling the aero-digestive axis.

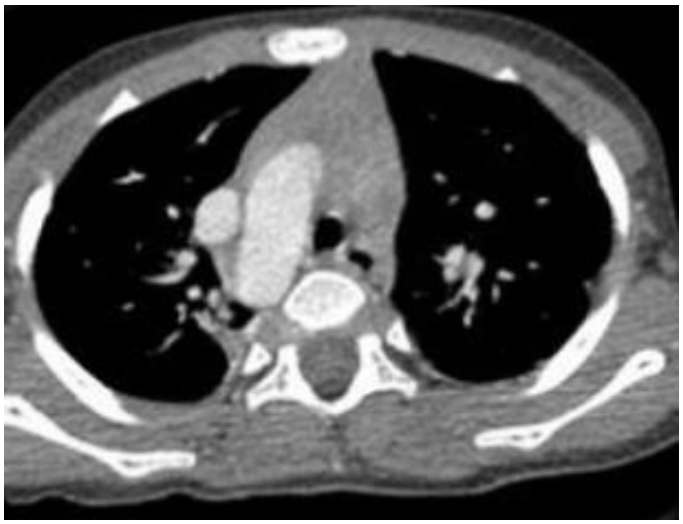


Figure 5: patient CT angiography, image of the arch of the aorta on the right.

PATIENT	Chest X-Ray	ultrasound	TOGD	ANGIOSCAN
1	Normal	Normal	Posterior imprint on the thoracic esophagus	Double aortic arc
2	upper and right para-cardiac alveolar and lobar foci	-	Extrinsic imprint on the posterior side of the proximal esophagus, esophagustight stenosis, esophagus' vascular compression by an aberrant sub-clavian artery.	Aberrant birth of the right sub-Clavian arteryon on the posterior part of the arch of the aorta + contiguous birth of the 2 primary carotid arteries.
3	Normal	NORMAL	_	Double aortic arch encircling the trachea and esophagus
4	Normal	NORMAL	Large extrinsic posterior imprint with narrowed aspect of the opposite trachea	Double aortic arch encircling the trachea and esophagus with compression
5	NORMAL	-	_	Double aortic arch encircling the aerodigestive axis
6	bronchial Syndrome	_		Double aortic arch encircling the aerodigestive axis which is narrowed at this level with a predominant hemi-arch on the right
7	Right Middle Lobe Focus	NORMAL	-	Aspect of a double aortic arch dominating on the right tying the aero-digestive axis
8	NORMAL	NORMAL	-	Double aortic arch responsible for tracheoesophageal compression

Table 1: Summary of the radiological of infants with vascular arches anomaly

Discussion

aortic arches Abnormalities bring together many malformations affecting the aortic arch, its branches or even the pulmonary artery. They can lead to the formation of complete vascular rings (double arc or straight arc accompanied by a left arterial ligament) or incomplete (aberrant left pulmonary artery, birth defect of the brachiocephalic arterial trunk or of the retro-oesophageal subclavian artery); its symptomatology is very variable [6].

The date's Symptoms onset varies, from the neonatal period, if the vascular ring is tight, to the incidental discovery in adulthood in forms with little compression [10]. In an Irish study, the diagnosis mean age was 13 months [8]. While in this series it is 7 months 7 days. Indeed, these abnormalities are most often highlighted when they are symptomatic. It can also be a discovery during the assessment of a congenital heart disease or during a systematic X-ray [7].

Clinically, the symptomatology is characterized by stridor, respiratory distress episodes with sometimes asphyxia. Dysphagia signs are often overlooked and are considered on the second rate [1,2].

In the Irish series of Gormley's 16 cases stridor was found in all cases [8], persistent cough and chronic dyspnea with a proportion of 75% each, apnea in 60% of them, recurrent wheezing, and dysphagia in 56% and 25% of the patients respectively [8]. In this series the clinical signs were mainly wheezing dyspnea in all patients and stridor in five infants. This joins Sarbeji's study which noted the predominance of stridor and wheezing dyspnea in all the patients [9]. This functional symptomatology can simulate other pathologies including infant asthma [4]. This is the case of a patient who has been treated for infant asthma without any clinical improvement.

This diagnostic difficulty is due to symptoms variability and the similarities with other pathologies including congenital heart disease and asthma.

Hence the interest of imaging which is interesting in anomalous aortic arcs diagnosis [1]. The Thoracic Angioscan is the reference exam to specify the abnormality type and its relationship with the respiratory and esophageal structures and associated abnormalities [1]. It specifies the detailed anatomy of the double arc as was the case in six patients in this study figure (5-4). As well as a distal birth of the artery under right Clavière [12, 13], found in one case in this series.

The Neuhausen anomaly accounts for 15-20% of the symptomatic abnormalities of the aortic arcs. The trachea and/or esophagus are compressed by a vasculo-ligament ring represented by a right arch on one side and the left arterial ligament (Figure 6) [11].

Currently, magnetic resonance imaging, like the angio-scan, allows to hold on an accurate anatomical assessment using the new technology. It has the benefit of being non-irradiant and non-invasive. However, it is relatively expensive. Tracheo-bronchial endoscopy can be performed in case of congenital stridor. It rules out a laryngeal or subglottic

abnormality and affirms the vascular character of tracheal compression by the pulsatile character.

The treatment is exclusively surgical. Surgery is indicated if the esophageal compression syndrome is severe with repeated respiratory infections and asphyxiation episodes that can be complicated by cardiorespiratory arrest [14]. After surgery, complete disappearance of symptoms is noted, as was the case in our six patients. as well as complications such as chylothorax, laryngeal edema, septicemia, hemorrhagic complications, persistence of respiratory manifestations including stridor can be reported [2].

Conclusion

Congenital abnormalities of the aortic arch are rare and varied, sometimes asymptomatic. This study recalls us that before any infant's recurrent respiratory or digestive symptoms, it is necessary to know how to mention a malformation of the aortic arches. The angioscan is the reference exam to confirm the diagnosis and to specify the anatomy of the malformation to inform the surgeon. Surgery remains the main therapeutic indication.

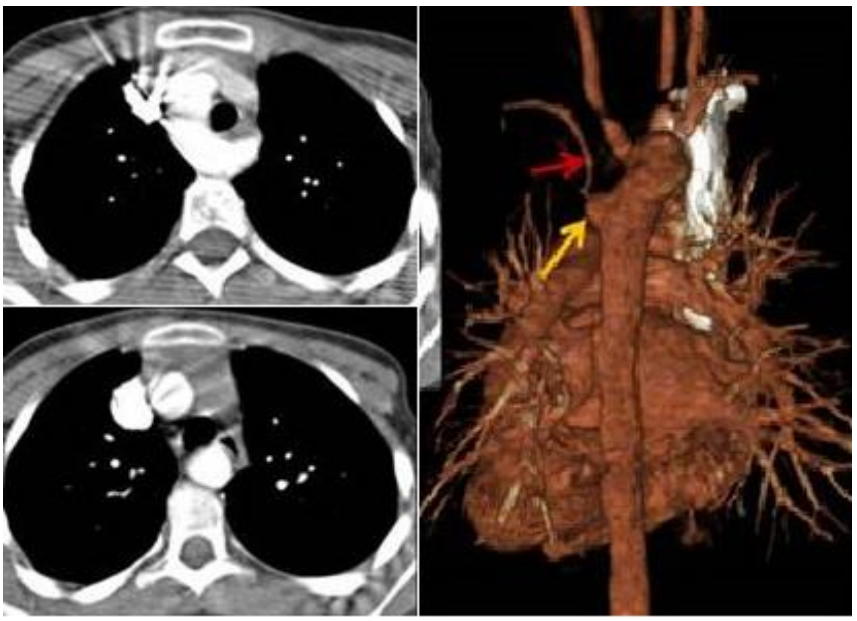
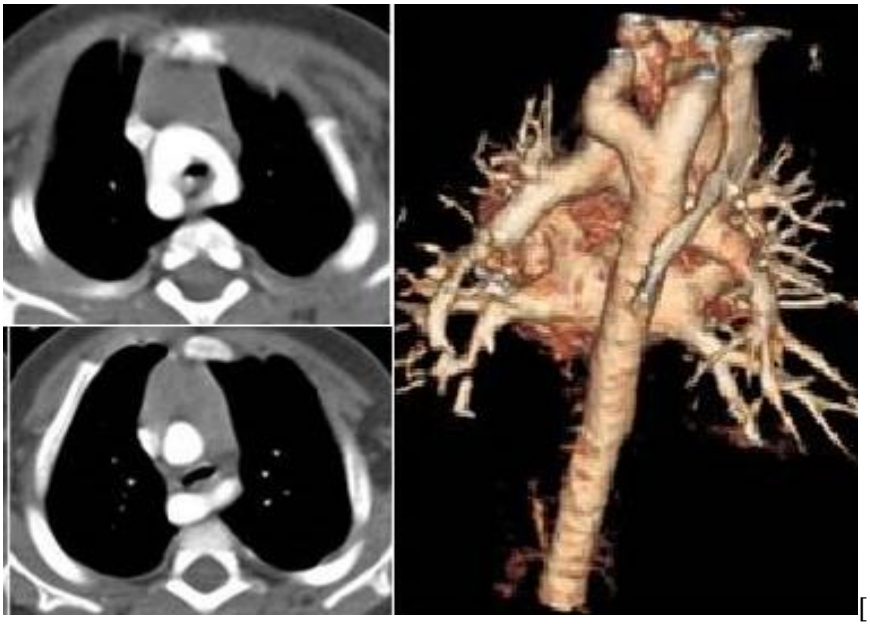


Figure 2: Posterior inprint on the thoracic



[

Figure 7 : Neuhausen anomaly [11].

Référence

1. Brevière GM. Anomalies des arcs aortiques. Paris: Éditions Scientifiques et Médicales Elsevier SAS; 2003 [11-940-D-80; 9 p.].
2. Le Bret E, Papadatos S, Folliguet T, Laborde F. Anomalies des arcs aortiques. Paris: Elsevier; 2002 [42-767; 12 p.].
3. Gerbeaux J., Couvreur J., Tournier G. -- Pathologic respiratoire de l'enfant, pp. 130-134. Pads, Flammarion M6decine Sciences, 1979.
4. Wong P.H.C., Lee J. -- Right aortic arch and Kormnerell's diverticulum. Thorax, 1983, 38, 553-555.
5. Lincoln JCR, Deverall PB, Stark J, Aberdeen E, Waterston DJ. Vascular anomalies compressing the oesophagus and trachea. Thorax 1969; 24:295.
6. E. Le Bret, F. Roubertie, M. Coblence, M. Ly, R. Roussin, E. Belli. Anomalies des arcs aortiques . Elsevier Masson SAS [42-767].
7. A.R. Ouedraogo et al. Un cas de malformation des arcs aortiques simulant un asthme. Rev Pneumol Clin (2018).
8. Gormley PK, Colreavy MP, Patil N, Woods AE. Congenital vascular anomalies and persistent respiratory symptoms in children. Int J Pediatr Otorhinolaryngol 1999; 51:23—31.
9. Sarbeji M, Azevedo I, De Blic J, Le Bourgeois M, Scheinmann P, Paupe J. Anomalies des arcs vasculaires chez l'enfant asthmatique. Ray Tr Allergol 1991; 31:35-40.
10. Chan YT, Dkk NG, Chong ASF, Ho JCS. Double aortic arch presenting as neonatal stridor. HK J Paediatr 2003; 8:126-9 [new series].
11. Badr Slioui,1,& Achraf Zaimi,2 et Latifa Chat3 Apport du scanner multi-coupe dans les anomalies des arcs et de l'arche aortique chez l'enfant Pan Afr Med J. 2019; 33: 312 .
12. Kimura-Hayama ET, Meléndez G, Mendizábal AL, Meave-González A, Zambrana GF, Corona-Villalobos CP. Uncommon congenital and acquired aortic diseases: role of multidetector CT angiography. Radiographics. 2010 Jan;30(1):79–98.
13. BB, Hoying JA, Markwald RR. In: Molecular Development of the Heart, in Hurst's the Heart. Fuster V, Walsh RA, Harrington RA, editors. Vol. 13. New York: McGraw-Hill Book Co; 2011. pp. 172–194.
14. Sihame Lemouakni,1,& Amale Hassani,1 Hakim Elyajouri,1 Mohammed Kmari,1 Hakim Ourrai,1 Rachid Abilkacem,1 et Aomar Agadr1Double arc aortique: à propos de deux cas . Double arc aortique : a propos de deux cas Pan Afr Med J. 2017; 27: 273.