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RESEARCH ARTICLE

ANOMALY OF BIRTH OF CORONARY ARTERIES FROM THE PULMONARY ARTERY: EXPERIENCE OF THE CARDIOPEDIATRICS DEPARTMENT OF THE LOUIS PRADEL HOSPITAL OVER THE LAST 20 YEARS

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Abstract

Submit: Abnormal coronary artery birth from the pulmonary artery (PA) is a rare disease, characterized by age-dependent clinical forms. We will describe the clinical, electrical and echocardiography features and the management strategy of this disease in adults.

Materials and methods: We enrolled 9 patients, 6 with the diagnosis of ALCAPA and 3 of ARCAPA. The mean age was 32 years with a sex ratio of 1.5/1. The presentations consisted of exertion symptoms and incidental findings. LVEF was reduced in 33% of patients with ALCAPA. Moderate mitral regurgitation (MR) was found in 44% of cases, particularly in cases of ALCAPA. Coronary CT scans confirmed that all our patients had coronary artery anomalies from the PA. The surgical treatment of coronary reimplantation was the therapeutic method of choice and was performed in 88% of our patients.

Conclusion: Coronary birth defect from PA is a very rare congenital pathology. The clinical symptomatology that leads to the discovery of the disease is diverse. Doppler imaging and CT scann is a key tool in the positive diagnosis. Early re-implantation surgery can avoid complicated forms.

Introduction:-

Abnormal birth of the coronary arteries from the pulmonary artery is a rare congenital heart disease. These anomalies are known by the Anglo-Saxon name: ALCAPA: anomalous left coronary artery from the pulmonary artery, when it is the left coronary artery that is affected; ARCAPA for the right coronary artery: anomalous right coronary artery from the pulmonary artery.

The incidence of ALCAPA is 1/300000 births (1), ARCAPA is much less frequently encountered, with an incidence of 0.002% (1). The clinical manifestations are diverse, ranging from sudden death due to myocardial ischemia or ventricular arrhythmia to the incidental discovery of a mitral regurgitation murmur or cardiac imaging.

Our work consists in describing and analyzing the epidemiological, clinical, electrical and echocardiography aspects as well as the therapeutic strategy of these diseases in order to bring out a profile comparable to the data in the literature.

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Materials and Methods:-

-Type and location of study:

This is a retrospective study over the last 20 years, carried out in the pediatric cardiology department of the Louis Pradel Hospital in Lyon (France).

-Inclusion and exclusion criteria:

Patients with a diagnosis of ALCAPA or ARCAPA hospitalized during the last 20 years in the congenital cardiology department of Louis Pradel in Lyon (France) were included. Any patient with another associated congenital heart anomaly was excluded from the study.

-Study population:

The trial enrolled 09 patients who met the inclusion criteria. Six patients were admitted for ALCAPA and three for ARCAPA.

Results:-

-Age and sex:

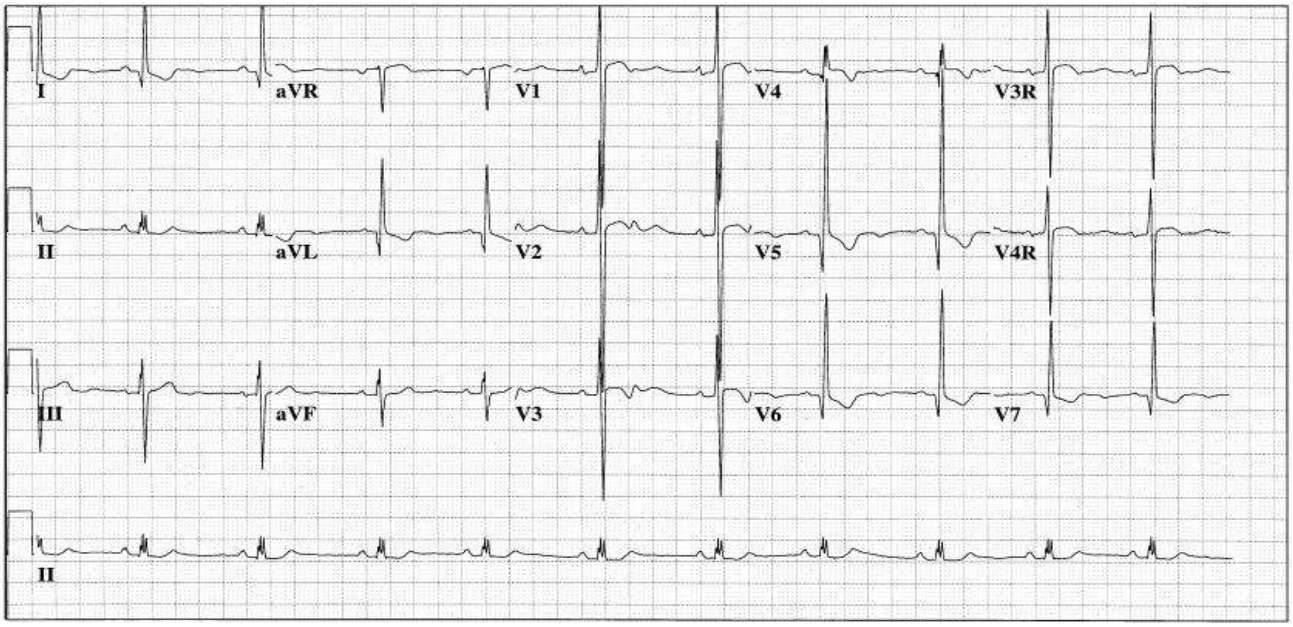
The average age was 32 years with a maximum of 65 years and a minimum of 16 years. We found a light predominance of women over men with a sex ratio of 1.5/1.

-Circumstance of discovery:

	Percentage	Form of coronary birth defect
Symptoms on exertion	**33%**	ARCAPA/ALCAPA
left ventricular failure	**22%**	ALCAPA
Incidentalum	**33%**	ARCAPA/ALCAPA
Murmur investigation	**11%**	ALCAPA

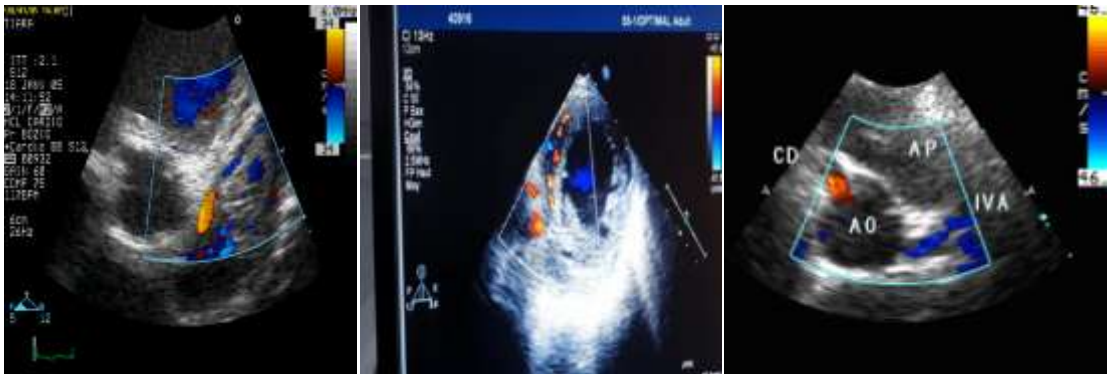
-ECG results

	ALCAPA	ARCAPA
Q wave in DI-AVL	6	0
Q wave in DII-DIII-VF	0	1
Normal	0	2

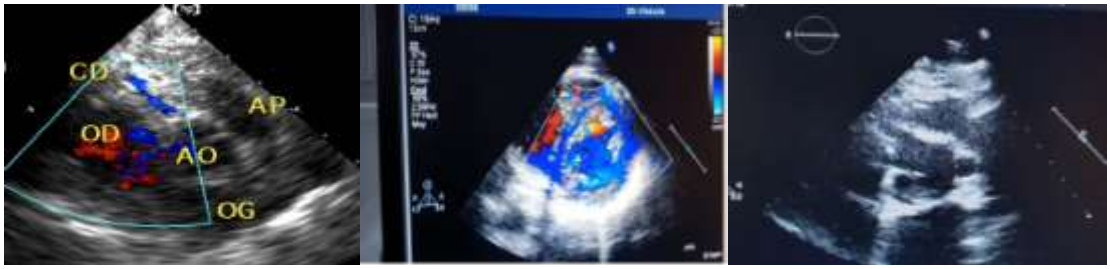


-Echocardiography findings

ALCAPA

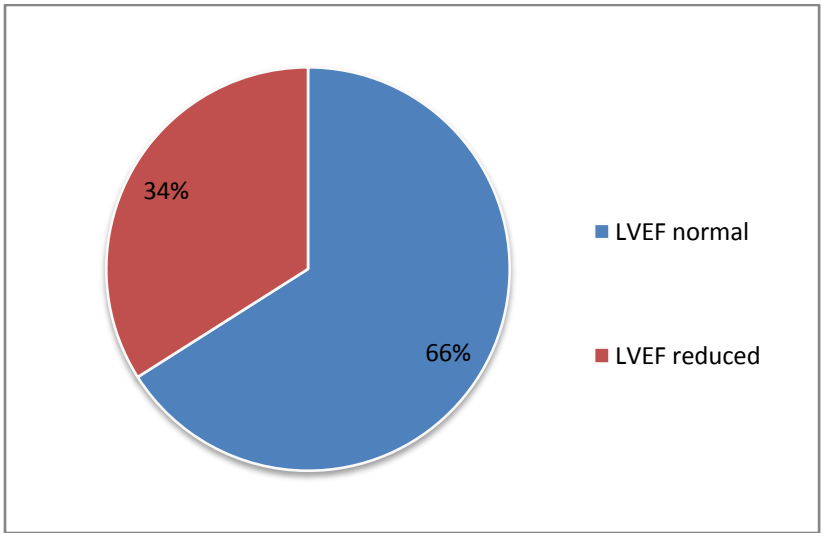


ARCAPA

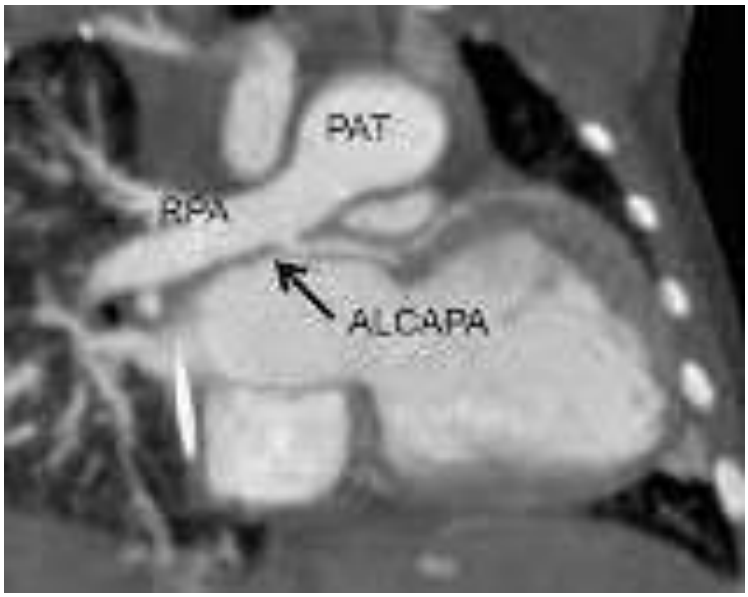


Left Cavity dilatation

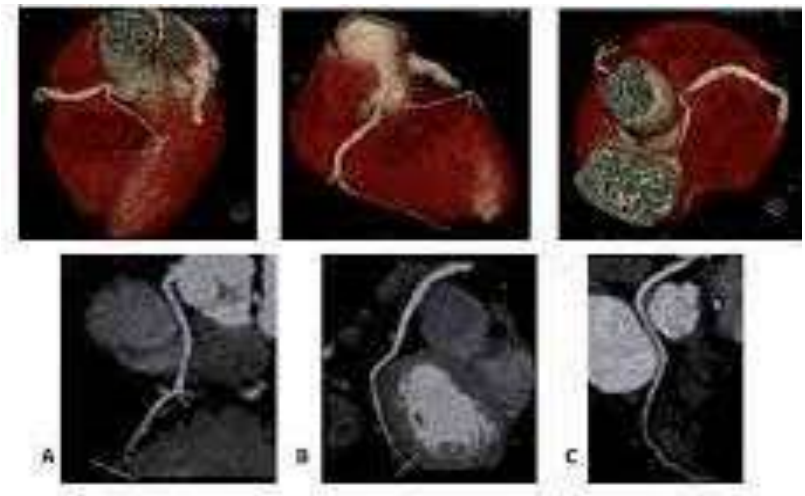
	ALCAPA	ARCAPA
Left cavity dilatation	4	0



-Coronary CT scan



ALCAPA



ARCAPA

-Treatment:

08 of the 09 patients studied had curative surgical treatment consisting of reimplantation of the coronary arteries to the aorta. Only 01 patient was treated medically.

Discussion:-

This rare pathology has clinical expressions of various severity depending on age, which is related to the degree of development of collaterals between the coronary arteries.

If the neonatal form is easily diagnosed by its severe clinical presentation, the adult one is more laborious to detect. The exceptional nature of this condition means that the literature is poor on this subject.

The average age of 32 years with extremes ranging from 65 to 16 years is a trend also found in the work of Emilie Laflamme et al, on the North American experience in the management of this disease in adults [4]. In the latter, the sex ratio was 1; compared to our series which showed a slight female predominance (sex ratio=1.5/1).

The incidentalum was reported in 33% of cases in the work of Emilie Laflamme et al, a percentage identical to that reported in our series [4]. In the same study [1, 4], a peripartum heart failure flare-up revealing an ALCAPA form was reported. This is comparable to our data showing such a peripartum complication in a young pregnant woman in the third trimester [6]. This period of peripartum could therefore reveal this anomaly which is subclinical outside of pregnancy. This is related to the hyper flow and increased oxygen requirements during the gravid state(4; 6). Symptoms of exertion, including chest pain and malaise, were found in 33% of cases in our study, both in ALCAPA and ARCAPA carriers. This is one of the modes of revelation also found in the series by Carlos et al [1, 3 and 4]. Silent myocardial ischemia is reported to be the cause of these clinical manifestations [3]. The murmur of mitral regurgitation observed in all our patients with ALCAPA is correlated to chronic ischaemia of the anterolateral pillar leading to its dysfunction [1,4]. In Emilie Laflamme's series, a similar proportion of mitral leakage was found on examination [4].

Patients with ALCAPA had a particular electrical presentation marked by the presence of Q wave in DI-AVL. This ECG appearance was reported in 58% of cases in the review by Alper et al [7]. This is described as classic in the pattern of ALCAPA [1]. It is also noted that the presence of an mitral regurgitation murmur combined with this ECG finding should raise the diagnosis of ALCAPA. [1].

In the reports of Emilie Laflamme et al.(4) and Alper Guzeltas et al (7), the same echocardiographic data suggestive of ALCAPA and ARCAPA described in our series are cited. The signs of damage found mainly in half of the patients with ALCAPA are systolic left ventricular dysfunction secondary to chronic myocardial ischaemia and mitral insufficiency of minimal to moderate severity. The later has a dual mechanism recognised on echocardiography: an ischaemic mechanism seen in the image of a hyperechogenous pillar and a functional mechanism such as left ventricular dilatation. These echocardiographic results of this congenital condition have also been reported (4) and (7). Additional coronary CT scan in all patients in our study confirmed the diagnosis of ALCAPA and ARCAPA.

The only curative treatment is surgical, the aim being to restore dual coronary circulation once the diagnosis has been made. Two surgical methods are often used. The commonly used procedure is anatomical correction by direct reimplantation of the coronary artery ostia into the aorta. In difficult forms, the Takeuchi procedure is recommended. This technique consists of tunneling the coronary artery from the pulmonary artery into the aorta. Complications may range from tunnel stenosis in the Takeuchi procedure to torsion or stenosis of the coronary ostia in the direct coronary reimplantation procedure. Follow-up of our patients showed initial clinical improvement and secondary echocardiographic parameters, notably regression of mitral regurgitation.

Conclusion:-

The birth anomaly of the coronary arteries from the pulmonary artery is a rare congenital pathology with multiple clinical expressions depending on the age and the underlying cardiac anomaly.

The electrocardiogram and multimodal imaging, namely echodoppler coupled with coroscanner, help to establish a positive diagnosis. Coronary reimplantation surgery is the recommended gold standard curative treatment.

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