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## Os desafios da publicação científica no campo da saúde em Angola

### *The challenges of scientific publication in the health field in Angola*

Hermenegildo Osvaldo Chitumba<sup>✉ 1</sup> Humberto Morais<sup>2</sup>

**Palavras – Chave:** Publicações; Ciência; Angola

**Keywords:** Publications; Science; Angola

Pode-se observar que ao longo da história da humanidade, a vida dos indivíduos e das sociedades foram transformadas pelos resultados da pesquisa.

Nos países desenvolvidos são amplos os ganhos com a pesquisa científica do ponto de vista financeiro e contribuições elevadas no número de publicações<sup>1</sup>.

Objectivando examinar a produção científica global de artigos indexados à base de dados Scopus – Elsevier, do ano 1970 - 2016, Salmerón-Manzano e Manzano-Agugliaro<sup>2</sup>, concluíram que os países com maior produção científica global foram os Estados Unidos seguido pelo Reino Unido.

Apesar de actualmente os Países Africanos constituírem o berço dos principais problemas de saúde, fundamentalmente do fórum infeccioso, o contributo destes para a produção científica global ainda é marcadamente insuficiente e com alguma limitação.

Até 1996 a produção científica africana em termos globais não passava de 1.5%<sup>3</sup>. A base de dados AJOL (African Journals Online), até Fevereiro do ano de 2022 apontava a Nigéria, África do Sul, Etiópia, Kenya, Gana e Egito, como sendo os países Africanos com maior número de revistas científicas.

África do Sul, Egito e Nigéria compõem 60% do número total de artigos indexados na base de dados PubMed<sup>4</sup>.

Nos últimos anos, em Angola, tem estado a crescer o número de investigadores que de uma forma tímida e com iniciativas isoladas vêm evidendo esforços para alavancar a pesquisa e investigação clínica na área da saúde no país.

Há 5 anos, era uma miragem falar de revistas científicas electrónicas. Actualmente o país está dando passos significativos, com o nascimento de revistas científicas em vários domínios do saber. Contudo, mais do que o aumento no número de revistas científicas que por sinal são de algum modo o maior mecanismo para a difusão rápida do conhecimento, torna-se imprescindível a capacitação dos profissionais envolvidos na produção do conhecimento.

Ao analizarmos o número de artigos indexados à base de dados PubMed, usando como estratégia de busca avançada o termo *Angola[Affiliation]*, ou seja, artigos que possuem pelo menos um autor contendo a palavra Angola na afiliação institucional entre o ano de 1800 - 2022, verificamos que o sistema a 04 de Fevereiro de 2022,

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retorna 614 artigos, dos quais 554 (89,8%) foram publicados nos últimos 10 anos (2012-2021). Observando-se assim, um aumento significativo de 9 artigos publicados em 2012 para 97 artigos publicados em 2021 (Fig. 1).

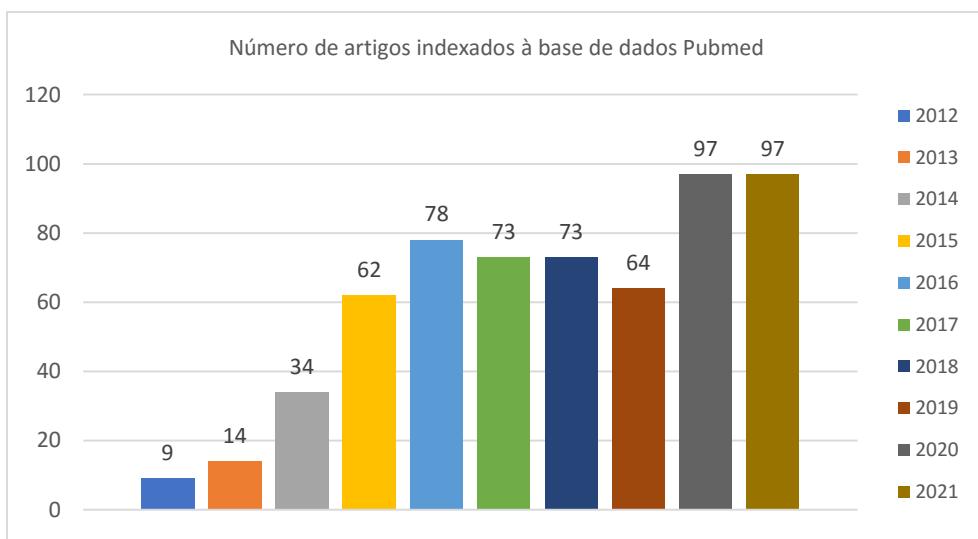


Figura 1 – Número de artigos indexados à base de dados PubMed no período de 2012 a 2021, usando como estratégia de busca o termo *Angola[Affiliation]*

Corroborando os pronunciamentos encontrados no manual de Frascati<sup>5</sup>, para que os países em via de desenvolvimento dos quais Angola faz parte, deiam passos largos e positivos na direcção certa, torna-se necessário dar a devida atenção por parte dos decisores políticos e gestores, na capacitação dos recursos humanos e financeiros destinados a investigação científica, encontrando assim um método para a consecução dos objectivos preconizados.

Este feito, terá como resultado a existência de uma massa crítica estável de docentes e investigadores que possam aferir com argúcia os principais problemas e lacunas no domínio das ciências médicas e da saúde, com o estabelecimento de prioridades na sua resolução.

Nestes dois anos de existência da RACSaúde, foi possível constatar, por um lado, a existência de muita substância a nível das Universidades, Institutos e Centros de Investigação Científica, Centros de Saúde e Hospitais dos mais variados níveis, que devidamente encaminhada teria como resultado um impulso na produção científica nacional; por outro lado, constatamos que, ainda existe dificuldade no manejo de ferramentas tecnológicas e no domínio do método de investigação científica (por parte dos profissionais de saúde, docentes e investigadores), que são pressupostos para produzir informação com rigor e qualidade aceitável.

Uma das formas de colmatar, estas insuficiências é a criação de mecanismo de incentivo aos actores do campo da produção científica.

Deve-se encarar a investigação científica não apenas como uma forma de gerar conhecimento novo para enriquecimento dos currículos, mas sim, como uma ferramenta necessária e imprescindível para o desenvolvimento, diversificação da economia, e uma forma de identificação de problemas que de algum modo requerem solução do ponto de vista da ciência.

Acreditamos que a RACSaúde venha a ter no futuro um papel fundamental na difusão dos trabalhos científicos realizados nas nossas universidades, instituições de investigação e de saúde. Apelamos assim, aos interessados o envio de trabalhos para a nossa revista. A equipa editorial tudo fará, para que, o sistema “recepção – revisão por pares – aceitação e/ou rejeição dos artigos – publicação na revista” seja feita com celeridade, sem prejuízo na qualidade dos trabalhos publicados. Todos os pesquisadores docentes profissionais de saúde e não só estão convidados a dar vida a revista e participar da internacionalização, visibilidade e desenvolvimento da ciência em Angola e no Mundo.

Agradecemos a todos os autores que consideraram a RACSaúde para tornar público os resultados de suas investigações nesta edição.

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## A narrative review on Rotavirus A in Mozambique

Edgar Cambaza<sup>1</sup> , Edson Mongo<sup>2</sup> , Aline Gatambire<sup>4</sup> , Edirsse Mateonane<sup>5</sup> , Raquel Chissumba<sup>2</sup> , Assucênia Chissaque<sup>5,6</sup> , Nilsa de Deus<sup>5,7</sup>

### ABSTRACT

**Introduction:** Rotavirus A (RVA) is a leading cause of acute infant gastroenteritis in Mozambique, responsible for approximately 13,000 annual infant deaths in peri-urban and other areas.

**Aim:** the present review aimed summarize the most relevant and recent literature regarding RVA infection in Mozambique.

**Methods:** the documents were obtained from electronic databases such as PubMed, Google Scholar, Scinapse, Scilit, and Microsoft Academic to find the leading scientific publications related to Mozambique's RVA.

**Findings:** The search allowed us to find 20 peer-reviewed journal articles, three official reports, eight abstracts from national conferences, one thesis, and other documents to supplement the information. Rotavirus frequency ranged between 24 to 42.4%, 34.8% attributable to Mozambique. Most data are hospital-based from Maputo, Sofala, Zambézia, and Nampula provinces. Nampula province shows the highest number of cases. Thus, there might be some bias on the geographical distribution of the virus. The prevalence is high in children less than one year. Regarding the control, the Expanded Program on Immunization (EPI) introduced a monovalent vaccine in September 2015 (Rotarix), which positively impacted the reduction of RVA cases. After vaccine introduction, a high diversity of RVA genotypes was observed, with the predominance of G1P[8] and the emergence of G9P[4], G9P[6], G3P[4]. However, only the whole genome sequence can confirm if it is due to the natural genotypes fluctuation. RVA infection was detected in swine, and a recent analysis reported an RVA strain from children clustered with different animal strains.

**Conclusion:** The scarce yet highly reliable research resources allowed scientists to detect RVA G1P[8] and other genotypes, potential animal reservoirs, and to find that RVA is more prevalent during the transition dry-rainy season, and the virus becomes more frequent when children approach the 11<sup>th</sup> month, to then decline as they age. It is essential to develop studies providing a broad view of RVA reservoirs as part of the strategy to control its dissemination.

**Keywords:** Rotavirus infections; Children; Diarrhea; Mozambique

### INTRODUCTION

Diarrhea remains a tremendous public health problem globally and is associated with 446,000 deaths among children under five years old in 2016 <sup>1</sup>. In Mozambique, even with the strategies implemented to reduce diarrhea cases in the last two decades (improving hygiene, sanitation, and access to clean water) <sup>2</sup>, diarrhea remains the leading cause of mortality of children under five years old, with 4383 deaths annually <sup>1</sup>.

Rotavirus, particularly group A (RVA) <sup>3</sup>, is among the significant causes of acute infant gastroenteritis globally <sup>1,4</sup>. Data from the global burden of annual RVA mortality shows that in 2016, about 128,500 children under five years old died, of which 104,733 in sub-Saharan Africa <sup>1</sup>. The World Health Organization <sup>5</sup> (WHO) recommended introducing the rotavirus vaccine in countries with high prevalence and countrywide continuous surveillance before and after the vaccine's introduction.

The global enteric multicenter study (GEMS) showed RVA as the leading cause of diarrheic diseases in Mozambique with an attributable factor of 34.8% and reported a prevalence ranging from 24 - 42.4% <sup>6</sup>. Nevertheless, there is very little published literature on Mozambique's RVA and its impact on human or animal health, particularly in the northern and central regions of the country <sup>2,7-10</sup>. Thus, there is a need to expand

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surveillance and epidemiological studies to understand the impact of local strains and improve the current control measures<sup>9-11</sup>. It is also essential to identify the strains and compare them with others from different countries to understand the molecular epidemiology and design appropriate measures. Thus, this study reviews the most relevant information on RVA infection in Mozambique.

## METHODS

We used Google Scholar, Scinapse, Scilit, PubMed, Scielo, and Microsoft Academic to find the leading scientific publications related to Mozambique's RVA to write this manuscript. The leading search keywords were "rotavirus" and "Mozambique." We selected scholarly documents with keywords or related in the title. In some cases, the titles presented Mozambique's districts or provinces' names, and some of the documents were about Rotavirus and other diarrheic diseases.

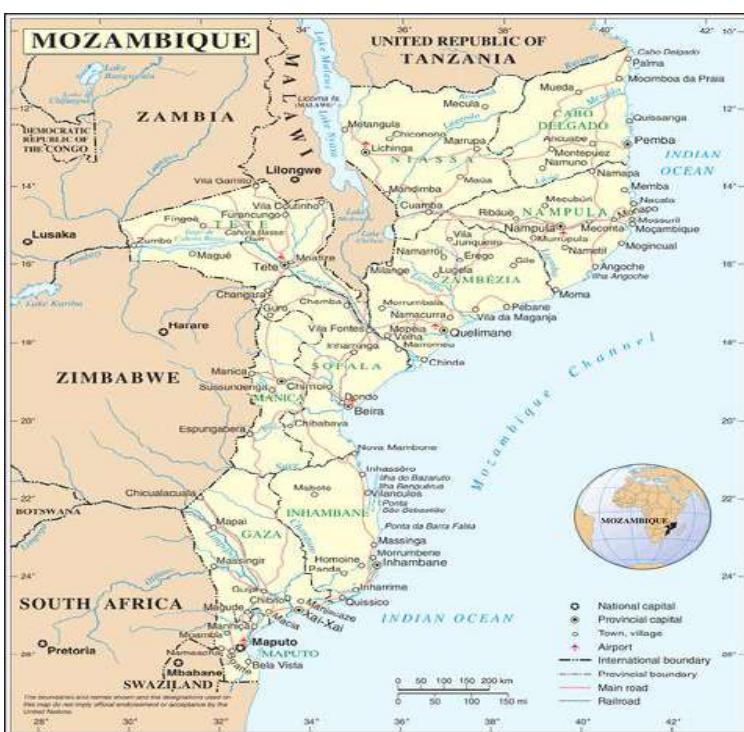
We found 20 peer-reviewed journal articles, three official reports, eight abstracts from national conferences, and one thesis through the search. We added other elements such as the map and figure as we reviewed the primary sources.

We analyzed the documents using the qualitative data analysis software Atlas.ti 9 (Atlas.ti Scientific Software Development GmbH, Berlin, 2020), using the codes "etiology," "transmission," "geographical distribution," "susceptible groups," "vaccination," "impact, surveillance, treatment," and "infrastructure." Then, we summarized the data into a coherent text, using other sources from the literature to provide more consistency to the text.

## RESULTS AND DISCUSSION

### Geographical distribution

First, it is essential to make a brief description of Mozambique (Map 1) so that anyone can easily understand how spread RVA is in Mozambique according to the current scholarly information. It is a tropical country with 801,590 km<sup>2</sup>, located in Southeastern Africa, divided into eleven provinces, with an eastern coastline (Indian Ocean) from north to south. According to the last census (2017)<sup>12</sup>, the population comprises 27,909,798 inhabitants, significantly below poverty. Diarrheic diseases are common and reported as the fourth major cause of mortality in children aged five years or less<sup>2</sup>.



Map 1 - Mozambique, the country's regions, and cities. United Nations 16, under public domain

Rotavirus is highly prevalent among < 5-year old children in Mozambique<sup>13</sup>. Thus, non-surprisingly, there have been reports from different country areas. de Deus et al.<sup>14</sup> mentioned six surveillance sites from the National Surveillance of Diarrhea (ViNaDia): Hospital Geral José Macamo, Hospital Geral de Mavalane and Hospital Central de Maputo (Maputo City), Hospital Geral de Quelimane (Zambézia Province), Hospital Central da Beira (Sofala

Province), and Hospital Central de Nampula (Nampula Province). ViNaDia surveillance system produces most of the data on the epidemiology of RVA in Mozambique. INS is the leading institution studying RVA in Mozambique<sup>10</sup>. For the sake of research, the reference mentioned above virology laboratory of INS has the country's state-of-the-art equipment for RVA analysis<sup>7,15</sup>, an integral component to achieve ViNaDia's goals. Manhiça Health Research Center (CISM) also develops studies on RVA<sup>10</sup>. Another laboratory with comparable equipment in the Directorate for Animal Sciences of the National Institute for Agricultural Research (IIAM)<sup>8,9</sup>.

Nampula has registered the highest number of cases (see details in subsection 5.3)<sup>7</sup>. There are potential explanations for why this province presented most cases. For instance, (1) Nampula is the most populated province<sup>12</sup>, and (2) there are serious hygiene issues in Nampula's urban areas impacting the spread of diarrheic diseases<sup>17</sup>. A recent analysis of the epidemiology of Rotavirus in Nampula observed that piped water was associated with rotavirus infection and suggested that this can be related to the water network that is old and damaged, while in the post-vaccine period age, year, and contact with different animals were associated with rotavirus infection<sup>18</sup>. It would be worthy of studying the epidemiology of RVA in the Novo Bairro area of Quelimane city, well-known as an urban area with very precarious sanitation and a history of cholera outbreak<sup>30</sup>.

Mussa et al.<sup>19</sup> reported cases in the cities of Maputo and Matola, RVA infection was reported by de Deus et al.<sup>14</sup>, in children less than five years old with diarrhea in Mavalane General Hospital (Maputo City) in 41.3% and Manhiça District Hospital (Maputo Province) in 44.3%<sup>14</sup>. On the other hand, Langa et al.<sup>11</sup> reported RVA infections in Chókwè District (Gaza Province) in 24.0%<sup>11</sup>. Based on the ViNaDiA platform, RVA infection was reported in four provinces of the country: Maputo city (southern region of the country), Sofala, Zambézia (Center region of the country), and Nampula (Northern region of the country), ranging from 12.2% - 40.2% regardless to the vaccine introduction period<sup>14,20</sup>.

## Vaccination

So far, there are four licensed vaccines for Rotavirus in the world: the pentavalent RotaTeq™, monovalent Rotarix™ (GlaxoSmithKline Biologicals, Rixensart, Belgium), Rotavac® (Bharat Biotech, India), and Rotasiil® (Serum Institute of India Pvt. Ltd. India)<sup>21</sup>. Mozambique introduced Rotarix™ in September 2015 as part of the National Immunization Program. The Instituto Nacional de Saúde and partner organizations have been assessing the vaccination outcomes. After vaccine introduction, there was a significant reduction in diarrhea hospitalization and rotavirus infection, from 40.2% in 2014 to 13.5% in 2017, and reduction cases occurred in undernourished children from 42.7% to 12.2%<sup>18</sup>.

## Susceptible groups

RVA is frequent in children up to 5 years old, with a slight majority in male patients<sup>18,22</sup>. A study reported the median age of hospitalization of 9 months in 2014 and 11 in 2015 during the pre-vaccine period than ten months in 2016 and 2017 in the post-vaccine period. In Chókwè, Langa et al.<sup>11</sup> found 94.1% of children positive for Rotavirus aged up to two years old. de Deus et al.<sup>22</sup> in Manhiça and Mavalane found 52.7% of RVA cases in children from 0 to 11 months. Similar results were found in a case-control study conducted in Manhiça, where children under one year had an attributable fraction of 34.8% of RVA infection<sup>23</sup>. Thus, these findings suggest that RVA infection is higher in children less than one year and decreases as they grow up.

## Molecular characterization of RVA

Mozambique uses the WHO's algorithm for RVA detection and characterization<sup>24</sup>. Several studies on RVA uses ELISA through the commercial kits, mainly Prospect Oxoid with a specificity of 99.2% (96% - 100%) and sensitivity of 100% (95% - 100%). Reverse-transcriptase polymerase chain reaction (RT-PCR) performs the molecular characterization of RVA based on two significant proteins, VP7 and VP4, which gives the genotype combination G and P. The complete genome sequence is essential to perform the phylogenetic analysis and sequences comparison.

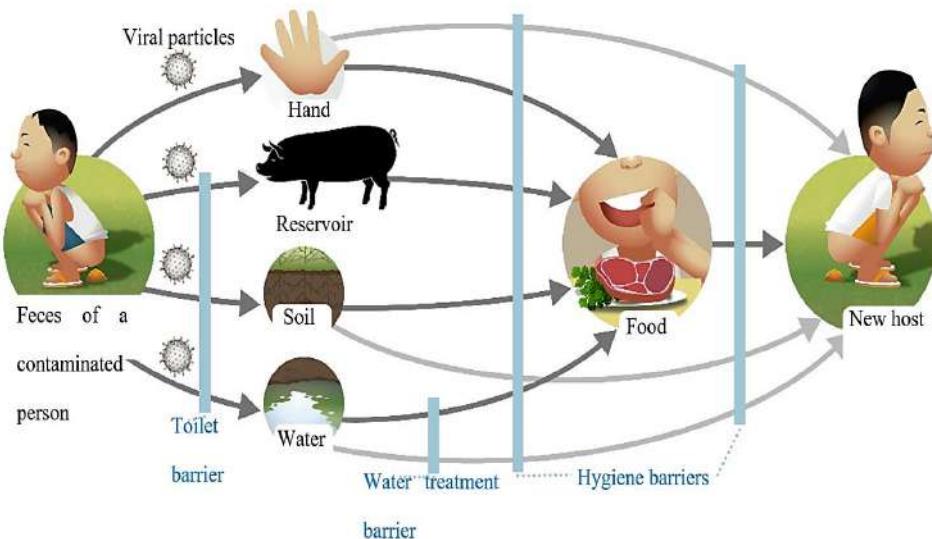
Globally the most common RVA genotypes are G1P[8], G2P[4], G3P[8], and G4P[8], being G1P[8] the most prevalent<sup>25,26</sup>. Studies conducted before the introduction of the vaccine in Mozambique (2015) reported G12P[8], G1P[8], and G12P[6] as the most common genotypes in Chokwé, a rural area in the Southern region of Mozambique<sup>27</sup>. João et al.<sup>28</sup>, in a study conducted in rural (Manhiça) and urban (Maputo city) areas, found G2P[4] and G12P[6] as the most prevalent genotypes. However, after vaccine introduction, a high diversity of RVA genotypes was

observed, including the emergency of G9P[4], G9P[6], G3P[4], and G3P[8] in Mozambique<sup>29</sup>. Other authors also analyzed the most frequent genotypes in children with diarrhea but different areas<sup>11</sup>. The vaccine seems to

contribute to the increased diversity and uncommon strains circulating as G3P[4] and G3P[8], G4P[6]. However, only the complete genome sequences can confirm if it is due to the fluctuation of natural genotypes or the pressure of the vaccine.

### RVA transmission pathways

RVA is a zoonotic virus able to be transmitted between different species. Swine is a significant reservoir<sup>30</sup>, and P[6] strain has been reported in animals and children with diarrhea<sup>11</sup>. In Chòkwé, this strain was detected in children and clustered with three porcine prototype strains<sup>11</sup>. This finding shows the possible zoonotic RV transmission in Mozambique. There is a long way to build a concise body of knowledge on RVA in animals in Mozambique, but the currently known studies are a good starting point.



**Figure 1** - Rotavirus transmission pathway. The vertical blue lines represent barriers to transmission.  
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The reported data of Rotavirus in animals suggest the implantation of surveillance of Rotavirus in animals<sup>31</sup>. However, additional studies are needed to understand better the role of animals in the transmission of RVA in the country and the emergency of new strains.

RVA is a public health concern due to poor hygiene and environmental contamination (Fig. 1)<sup>8</sup>. Whole-genome sequencing of a mixed stool containing G12 and GXP[14] strains showed that the last one clustered with sequences from a diverse group of animal strains, highlighting the possible interspecies transmission of RVA in the country<sup>32</sup>. It was also observed that swine could carry the RVA without symptoms<sup>9</sup>. However, it is necessary to deepen the current knowledge on RVA reservoirs in Mozambique.

Most of the studies that analyzed the monthly distribution of RVA cases showed that the highest prevalence occurs during the cold/dry season<sup>11,14,18</sup>. The authors reported that RVA is more frequent during July, August, and September, with relatively low temperatures and low rainfall<sup>11,14,18</sup>. Furthermore, D'Souza et al.<sup>33</sup> performed a similar study in three Australian cities (Brisbane, Melbourne, and Canberra) and found similar results, suggesting that this trend is worldwide. However, since the vaccine was introduced, cases were shifted to the wet season (October and November) in Nampula province Northern of Mozambique<sup>18</sup>. It would be helpful to study how exactly weather affects the prevalence of RVA.

### Health and commercial impact

In Mozambique, RVA is a significant cause of acute gastroenteritis and is associated with symptoms such as diarrhea<sup>13,15</sup>, vomiting<sup>38</sup>, resulting in dehydration<sup>39</sup>, and undernutrition<sup>22</sup>. RVA and other diarrheic diseases often

demand high costs of medical assistance and hospitalization<sup>7,15</sup>. Swine presents similar symptoms as humans, varying in severity, resulting in significant losses in the commercial and familiar sectors<sup>31</sup>.

## CONCLUSION

Mozambique has data on RVA from surveillance and studies. Methods such as ELISA and RT-PCR provide highly reliable results, but they might be costly for routine analyzes. It would be valuable to promote more basic epidemiological and social research on RVA and build a more robust body of knowledge.

RVA affects Mozambique's infant health, particularly in children less than one-year-old. The prevalence has declined since the introduction of the vaccine.

The predominant genotypes change over time, and the most recent data point to the G1P[8] as the most common. The current epidemiological information might not reflect the country's current situation because most data came from Maputo City and province, Sofala, Zambézia, and Nampula provinces. There shall also be surveys in the community, rather than data only from people seeking medical care.

RVA also occurs in swine in Mozambique, but the virus is likely to occur in other carriers. It is essential to develop studies providing a broad view of RVA reservoirs as part of the strategy to control its dissemination.

### Data confidentiality

The authors declare having followed the protocols in use at their working center regarding patients' data publication.

### Competing interests

The authors declare that there are no conflicts of interest nor any form of support.

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## Multiple cerebral aneurysms originating from previously resected cardiac myxoma

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### ABSTRACT

Primary cardiac tumors are very rare, accounting for 0,001 – 0,03% in autopsy series, cardiac myxoma (CM) account for approximately 50% of them. Association between CM and cerebral aneurysm is well documented in this condition but the occurrence of cerebral aneurysm after resection of CM is very rare, with only 40 reports in medical literature. We present a case of a 45 years old female patient with multiple cerebral aneurysms 2 years after a successful resection of cardiac myxoma, submitted to radiosurgery for 2 bigger aneurysms with good results. This report adds to literature new clinical findings

and a hypothesis of radiosurgery as an effective option of treatment for cerebral aneurysms with origin in CM. Keywords: Cardiac Myxoma; Cerebral aneurysm; Radiosurgery; Neurosurgery.

**Keywords:** Cardiac Myxoma; Cerebral aneurism; Radiosurgery; Neurosurgery

### INTRODUCTION

Cardiac tumors are rare lesions generally secondary lesions in continuity<sup>1</sup> or metastatic<sup>2</sup>. Primary cardiac tumors are very rare, accounting for 0,001 – 0,03% in autopsy series<sup>3</sup>, cardiac myxoma (CM) account for approximately 50% of them<sup>4</sup>. The World Health Organization (WHO) defines CM as a neoplasm composed by stellate to plump, cytologically bland, mesenchymal cells set in a myxoid stroma<sup>5</sup>.

Neurological complications may be the result of thrombus formation, such as intracranial aneurysms, infarcts, hemorrhages, brain metastases and cerebral vascular disorders can be associated with CM. Association between CM and cerebral aneurysm is well documented on this condition<sup>6</sup> but delayed cerebral aneurysm formation after resection of CM is very rare, due only 40 reports in medical literature being the first description in 1894 by Marchand<sup>7</sup>. Anyway, there are no prospective studies or cases with enough follow up to define the natural history, pathogenesis and earlier treatment remains unexplained.

We present a case of a 45 years old female patient with multiple cerebral aneurysms 2 years after a successful resection of cardiac myxoma, submitted to radiosurgery for 2 bigger aneurysms with good results.

### CASE REPORT

A 45 years old woman with 2 months of history of progressive dyspnea, initially for moderate efforts and at time of admission at hospital was for small efforts as daily routine activities. The medical history reveals childhood poliomyelitis with flaccid right upper limb monoplegia, no other relevant information.

The cardiac examination reveals a grade III apical murmur with no other relevant findings, neurological examination

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was normal except for the previous sequel monoplegia in the right upper limb. Vascular study was not done at that juncture, but echocardiogram reveals in left atrium a oval mass, pedunculated, adhered to the interatrial septum measuring 3.4 x 3.6 cm (Figure 1).

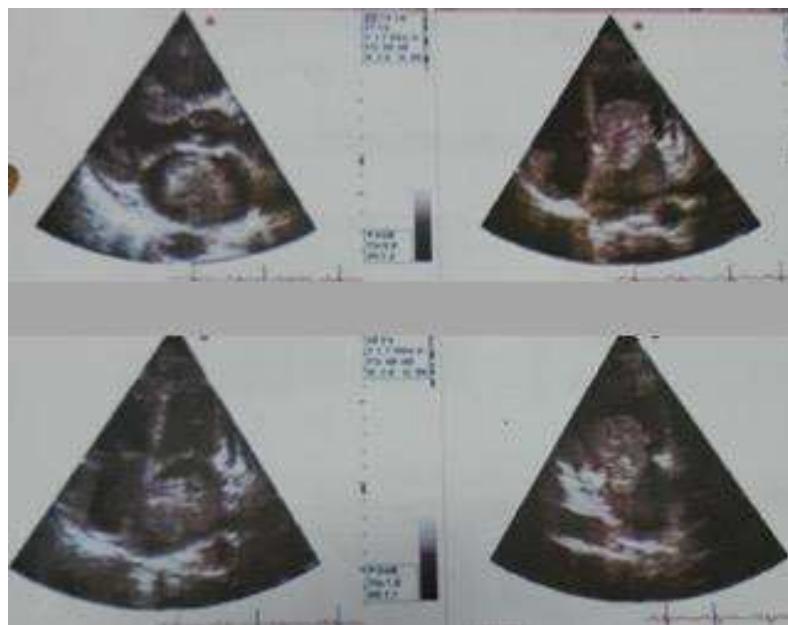


Figure 1 – Initial echocardiogram reveals in left atrium a oval mass, pedunculated, adhered to the interatrial septum measuring 3.4 x 3.6 cm.

Surgical resection was performed with complete resection and without postoperative complications, the patient was discharged from the hospital a few weeks later asymptomatic.

Two years later the patient complains of multiple episodes of tonic – clonic seizure, radiologic investigation with CT and MRI reveals multiple lesions suggestive of fusiform aneurysms (Figure 2A-2B, diagnostic was confirmed by digital subtraction angiography (DSA) showing multiple distal small fusiform aneurysms of middle cerebral artery M4 segment being the two largest lesions frontal and parietal 7 and 5 mm (figure 2C-D). The patient was submitted to radiosurgery with a single dose of 12 Gray in both lesions.

Seizures were clinically controlled using valproic acid 500 mg twice a day.

One-year DSA reveals control of the two irradiate lesions and new small and distal fusiform lesions (Fig. 2 E-F).

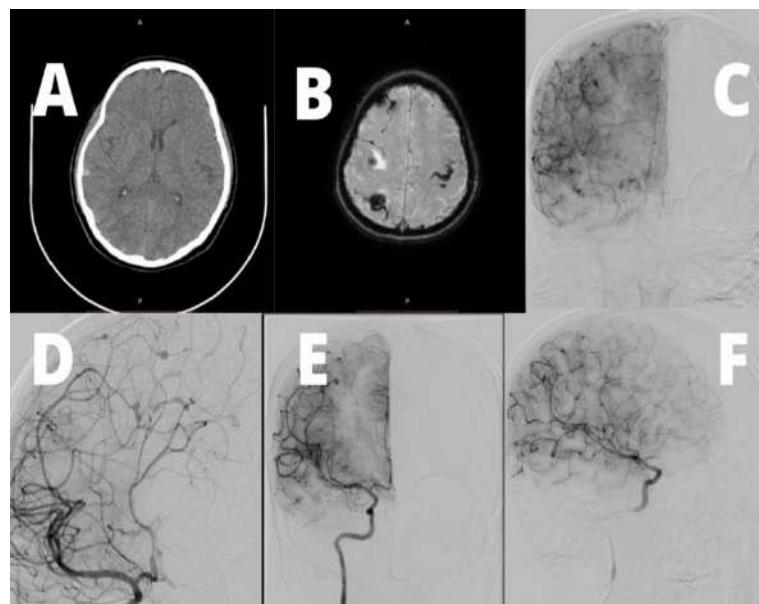


Figure 2 – CT and MRI reveals multiple lesions suggestive of fusiform aneurysms (A) and (B), respectively. Digital subtraction angiography (DSA) showing multiple distal small fusiform aneurysms of middle cerebral artery M4 segment being the two largest lesions frontal and parietal 7 and 5 mm. (C) (D) One-year DSA reveals control of the two irradiate lesions and new small and distal fusiform lesions (E) (F).

At this time with almost 3 years of follow up the patient is asymptomatic (cardiologic and neurologic), scheduled for new angiography follow up in a year.

## DISCUSSION

Acute neurological complications of cardiac myxomas as stroke or aneurysm formation are well described but delayed complications particularly fusiform aneurysms are very limited.

The present case has both common and uncommon characteristics compared with previously described. Female patients are 71% of previously described, CM is in the left atrium in 91% and almost all aneurysms were fusiform (91%) these characteristics are compatible with our case<sup>8</sup>. On the other hand we present a case with some news aspects in clinical presentation, that may help the medical community to perform diagnosis.

The formation of aneurysms after the resection of cardiac myxoma is unclear. The tumor cells in the embolism lead to perivascular damage, the inflammatory response at the site of injury and the deposition of the tumor into the vasa vasorum were proposed hypotheses to explain the mechanism why there is progression of aneurysms despite the surgery.

The endocrine and secretory properties of CM cells support the tumor emboli deposition in the vessel wall. A 23- year-old patient who presented with multiple cerebellar hemorrhages presented high levels of IL-6 in the CSF. These IL-6 levels returned to normal after resection of the CM and they have thus suggested these interleukines as a marker for neurological manifestations.

Dyspnoea as initial isolated symptom of CM without constitutional symptoms was not previously described and this can be a differential diagnosis of progressive dyspnea that can confirmed or ruled out by a inexpensive, non invasive and fast exam – Echocardiogram<sup>9</sup>, that can show a pedunculated lesion occupying all left atrium as previous biopsy shows<sup>10</sup>. The neurological symptom of this case (seizure) is also a new feature, previously described neurological symptoms migraine like headache, motor strength deficits and conscious level alteration<sup>7</sup>.

Treatments of cerebral aneurysm secondary to CM are not well defined. The conservative management is the preferred choice. However, the finding that dividing tumor cells are responsible for aneurysm formation suggests the possibility of using chemotherapy to prevent aneurysmal growth, but the results of doxorubicin alone are equivocal<sup>10</sup>. Low-dose radiation therapy alone or in conjunction with chemotherapy revealed more encouraging results<sup>12,13</sup>. Cardiac surgery to remove the primary cardiac tumors usually eliminates early neurologic symptoms, but cannot completely abolish the risk of delayed cerebral aneurysm formation, presumably as a result of metastatic seeding prior to surgery<sup>11</sup>. There are no randomized controlled trials to address this issue.

We present a case that only two years later the patient presents multiple episodes of tonic – clonic seizure after the CM resection. Recurrent embolic strokes with later evaluation revealed multiple cerebral aneurysms secondary to a cardiac myxoma detected 10 and 25 years, respectively, as reported in the literature. Despite treatment of cardiac myxoma, intracranial complications can have delayed presentation. Our case is in keeping with the current literature of the preferential distribution to the MCA territory as compared to the vertebrobasilar circulation.

## CONCLUSION

CM are very rare lesions and association with cerebral aneurysms are even rarer. We present a case of a 45 years old female patient with multiple cerebral aneurysms 2 years after a successful resection of cardiac myxoma, submitted to radiosurgery for 2 bigger aneurysms with good results.

This report adds to literature new clinical findings and a hypothesis of radiosurgery as an effective option of treatment for cerebral aneurysms with origin in CM. However, randomized controlled trials are essential to address this issue and guide therapeutic approach. On the other hand, non-invasive evaluation of the cerebral arteries should be done on a regular basis to detect the presence of delayed formation of intracranial aneurysms after the resection of the CM. This is due to the new findings about cerebral vessel involvement with intracranial deposition in cardiac myxomas.

## Limitações do estudo

Este estudo tem várias limitações. A mais importante delas é o reduzido tamanho da amostra (29 doentes). É, portanto, imperativo que mais estudos sejam realizados para melhor caracterizar o perfil radiológico dos pacientes com doença de Pott.

#### **Protecção de pessoas e animais**

Os autores declaram que os procedimentos estavam de acordo com os regulamentos estabelecidos pelos responsáveis da Comissão de Investigação Clínica e Ética e de acordo com a Declaração de Helsínquia da Associação Médica Mundial actualizada em 2013.

#### **Confidencialidade dos dados**

Os autores declaram ter seguido os protocolos do seu centro de trabalho acerca da publicação de dados.

#### **Conflito de interesse**

Os autores afirmam não haver conflito de interesse relacionados com a presente pesquisa.

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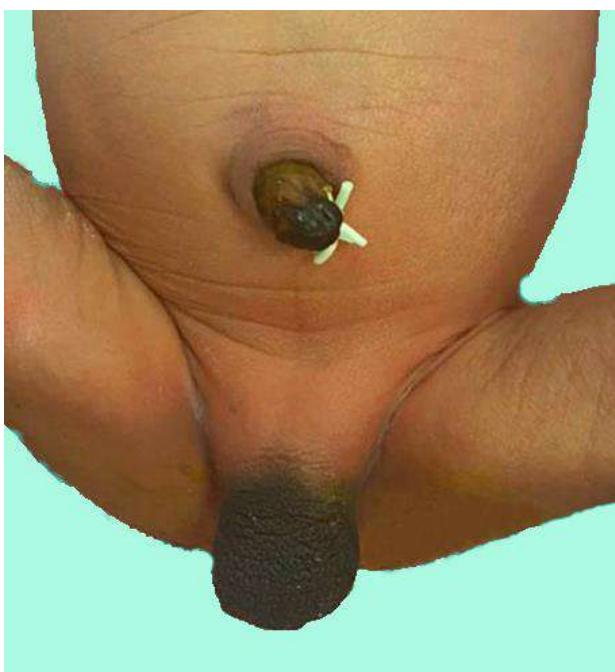
## Agenesia peniana com fístula vesicorectal: o primeiro caso reportado na literatura científica Angolana

### ***Penile agenesis with vesicorectal fistula: the first case reported in Angolan scientific literature***

Victorina Fato Botelho<sup>✉1</sup>, Augusta Sachiteque Marques<sup>2</sup>, Suelma Prata Tavares<sup>3</sup>, Maribel Diaz<sup>4</sup>

**Palavras – Chave:** Agenesia peniana; Fístula retrovesical; *Aphalia*

**Keywords:** Penile agenesis; Retrovesical fistula; *Aphalia*



**Figura 1 – Lactente com ausência total do pénis (Vista frontal).**

Trata-se de um lactente de 2 meses de idade, fruto de uma relação não consanguínea, referido do Hospital municipal da Caála, por apresentar ausência total do pénis. Este, apresentava o escroto bem desenvolvido com testículos descendentes bilaterais (Fig. 1), sem abertura urinária observável no períneo, com emissão de urina e mecoño.

A mãe é uma primigesta, de 20 anos de idade, gestação de termo, com seguimento pré-natal irregular, apenas três consultas, imunizada com duas doses antitetânicas, tomou suplementos com ferro e ácido fólico, sem referência pela mesma de intercorrências ao longo da gestação. Teve parto hospitalar, eutócico, cefálico, com 7 horas de duração, rotura de membranas duas horas antes do parto, líquido claro. Teve choro imediato e outros reflexos presentes e normais.

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Agenesia peniana ou Aphallia é uma anomalia congênita rara, que acomete indivíduos com o genotipo 46 XY, com uma prevalência mundial de 1 caso em 30 milhões de nascimentos<sup>1,2</sup>. A nível mundial encontram-se reportados na literatura científica, pouco menos de 100 casos, sendo apresentada pela primeira vez por Imminger em 1953. Ela resulta da incapacidade de formação e desenvolvimento do tubérculo genital<sup>3</sup>.

**Protecção de pessoas e animais:** os autores declararam que os procedimentos seguidos estavam de acordo com os regulamentos estabelecidos pelos responsáveis da Comissão de Investigação Clínica e Ética e de acordo com a Declaração de Helsínquia da Associação Médica Mundial.

**Confidencialidade dos dados:** os autores declararam ter seguido os protocolos do seu centro de trabalho acerca da publicação de dados.

**Consentimento informado:** obtido.

**Conflitos de interesse:** os autores declararam não ter qualquer conflito de interesse relativamente ao presente artigo.

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