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Cervical Angiomyolipoma Remodeled by Calcification, a Rare Entity in a Case at the Nianankoro FOMBA Hospital in Ségou

Aminata Fofana[1*], Oumar Sountié Coulibaly[2], Abdoul Wahab Haidara[1], Alpha Singuepiré[3], Bagouma Traoré[4], Nouhoun Traoré[5], MS Ag Med Elmehdi[6], Ahmadou Dembélé[7], Kalifa Haidara[8], Aly Dembélé[1], Sidiki Dao[9], Thierno Nadio[10], Souleymane Berthé[10], Aïssata Maïga[10], Kadidiatou Mandio Koné[10]

[1]ENT Department Nianankoro Fomba Hospital (HNF), Ségou, Mali
[2]HNF Paediatric Surgery Department, Ségou, Mali
[3]HNF Neurosurgery Department, Ségou, Mali
[4]HNF Anesthesia and Resuscitation Department, Ségou, Mali
[5]ENT Department at Kayes Mali Hospital, Kayes, Mali
[6]ENT Service Reference Health Centre of Commune VI, Bamako, Mali
[7]ENT Department, Sikasso Hospital, Sikasso, Mali
[8]Urology Department Nianamkoro Fomba Hospital, Ségou, Mali
[9]ENT Service CSREF IV, Bamako, Mali
[10]Ophthalmology Department of the District Hospital of Bamako, Bamako, Mali
Email: *aminata4@yahoo.fr

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Abstract

Angiomyolipoma is a rare benign tumour composed of blood vessels, smooth muscle and adipose tissue, located mainly in the kidney. Localisation in the cervix is very rare and does not appear to be associated with Bourneville's tuberous sclerosis. **Aim:** To report a case of cervical angiomyolipoma. Patient and method: A 14-year-old girl consulted for a right cervico-lateral mass that had been evolving for 7 years. **Results:** A cervicotomy was performed, and a firm mass with a smooth, limited surface was found, yellow in colour, and vascularised. It was removed in one piece, weighing 1925 g. The anatomopathological examination of the operative specimen revealed "an angiomyolipoma remodelled by calcification". After 5 years, the patient was monitored clinically and ultrasonographically, and the follow-up was straightforward. **Conclusion:** Angiomyolipoma is a benign tumour that mainly affects the kidney. It accounts for 1 to 3% of solid tumours of the kidney and rarely affects other organs. Its cervical location is very rare.

Keywords

Angiomyolipoma, Cervical, Calcification, Bourneville Tuberous Sclerosis

1. Introduction

Angiomyolipoma (AML) is a rare, benign mesenchymal tumour consisting of three different components: a fatty contingent, often the most abundant, a smooth muscle cell contingent and a vascular contingent [1]-[5]. It accounts for 1 to 3% of solid tumours of the kidney, which is the organ most frequently affected. In this case, it is very often associated with tuberous sclerosis of Bourneville and is more frequent in women. It can be extrarenal and have other localisations such as cutaneous, nasal/oral, liver, spleen, and lymph node. In this case, it is more frequent in men [1]-[4].

In our case study, we have a cervical location suggesting a variant of cutaneous AML. Cutaneous AML was described for the first time in 1990 by Fitzpatrick. In the literature, 24 cases have been reported and only 1 case of cervical localization has been described, located in the sternocleidomastoid muscle [1] [2].

AMLs containing calcifications are rare and could result from secondary intratumoral haemorrhage or bone metaplasia, 4 cases have been described in the literature [6] [7], hence the interest of this work.

Our aim is to report a case of angiomyolipoma of cervical location with calcifications and to discuss its rarity in the neck, and the diagnostic and therapeutic difficulty.

2. Observation

B. T., female, aged 14, consulted a paediatric surgeon in 2019 for a large right latero-cervical mass that had been developing for 7 years. The mass was increasing progressively, with no notion of pain or bleeding. Her history was unremarkable.

On physical examination, the swelling was found in the right cervical region, extending from the right jugulocarotid region to the supraclavicular region, the right spinal column and the nape of the neck. It measured 25 × 20 cm and was firm and mobile in both planes. The skin opposite was healthy (**Figure 1**). Her general condition was perfectly preserved.

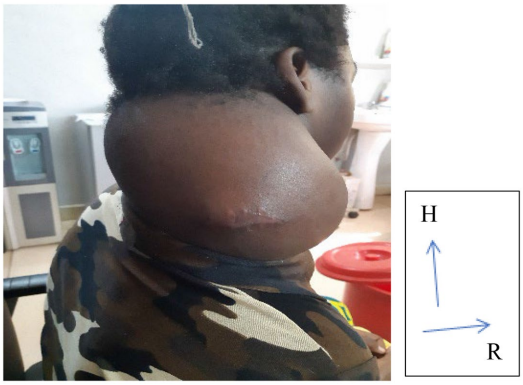

Figure 1. Posterior view of the patient.

Cervical Doppler ultrasound revealed a large heterogeneous hyperechoic tissue

mass with mixed vascular flow (arterial and venous).

A cervical CT scan revealed an isodense, heterogeneous tumour mass in the cervical soft tissue with multiple components (tissue, fat and calcium) without locoregional extension or cerebral metastasis. After injection, there was a discrete enhancement of the tissue component. The CT scan suggested a teratoma (Figure 2 and Figure 3).

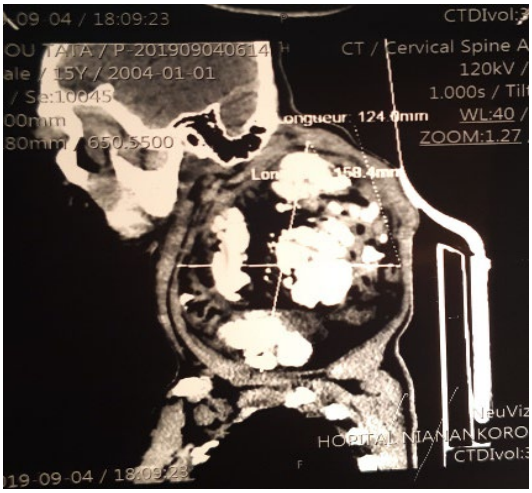

Figure 2. Multiple calcifications within the mass.

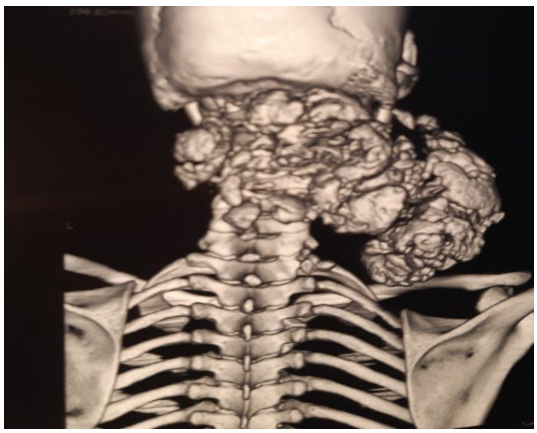

Figure 3. 3D reconstruction.

A biopsy was performed in favour of a haemangioma.

A cervicotomy with a modified Paul André L incision down to the nape of the neck was performed. When the mass was discovered, it was a firm mass with a smooth, limited surface, yellow in colour and vascularised. It was removed in one piece, weighing 1925 g (Figure 4).

The anatomopathological examination of the operative specimen was as follows: "an angiomyolipoma remodelled by calcification".

The immediate post-operative course was straightforward. After 5 years, the post-operative course was still straightforward.

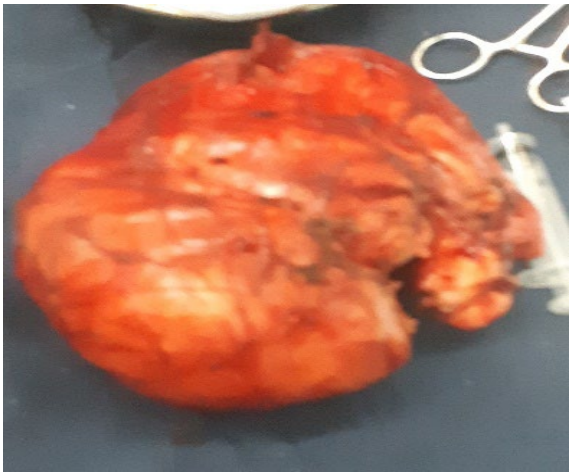

Figure 4. Operating piece.

3. Discussion

Angiomyolipoma is a rare tumour located in the kidney. Extrarenal forms of angiomyolipoma, particularly in the neck, have been rarely reported in the literature [1] [2]. The extrarenal form is not associated with Bourneville's tuberous sclerosis (BTS), unlike the renal form where BTS is found in more than 20% of cases [5]-[7].

Extrarenal forms of AML are found in men, unlike our case study, which involved a 14-year-old girl. However, women are more likely to develop AML than men due to hormones [7].

In the majority of cases, AML is asymptomatic, discovered incidentally during imaging examinations carried out for other pathologies [3]. However, the tumour may increase in size and compress neighbouring organs, and may express itself as a voluminous cervical mase, as in our patient. There are very few clinical signs specific to calcified AML apart from the large palpable mass, as calcification itself does not cause any symptoms [1]-[4]. In our patient, there were no warning signs apart from the aesthetic problem (large cervical mass), there was no clinical or biological anaemia, and no neck pain either.

The diagnosis of angiomyolipoma in its classic, uncomplicated form is generally made radiologically, when a fatty contingent is identified.

On ultrasound, the fatty component of angiomyolipomas is hyperechoic with an acoustic shadow cone. In the case of a low-fat content, this ultrasound appearance will not be described. However, this hyperechoic appearance is not specific and, in no case, allows a diagnosis of certainty [8]-[10].

Computed tomography (CT) is the gold standard, with a sensitivity of 90%. In the majority of cases, it is used to make a positive diagnosis of AML by demonstrating the characteristic fatty component, as well as calcification, which is often peripheral or curvilinear.

MRI, in addition to CT, provides a better indication of the tissue nature of the mass, in particular the fatty and vascular components [7] [8].

In our review of the literature, we found 4 cases of angiomyolipoma with calcifications [7]. The mechanisms thought to explain the presence of calcification are

- Intratumoral haemorrhagic recurrence;
- Bone metaplasia associated with fibrous scarring.

Our case study is, therefore, interesting not only because of its cervical location but also because of the exceptional calcification of the angiomyolipoma.

In our study, neither imaging nor biopsy of the tumour was able to provide a presumptive diagnosis of AML even though all the radiological arguments were in favour. This diagnostic difficulty is probably due to the cervical location, as the neck is not a preferred site for angiomyolipoma. It is almost exclusively renal [7]-[10].

The biopsy suggested a haemangioma, probably because the biopsy was taken in the vascular tissue of the angiomyolipoma.

There is no consensus governing the management of this type of tumour, although surveillance may be accepted for small masses of less than 4 cm and symptomatic forms or those larger than 4 cm may be treated by embolisation or partial or total surgery [3].

Management must be adapted on a case-by-case basis. Calcification does not affect management options but may indicate chronicity or previous intra-tumour haemorrhage.

The delay in managing our patients is due to the socio-economic situation and cultural considerations, where access to primary healthcare is still a luxury for most of our populations.

Cervicotomy was performed on the basis of a teratoma. In our patient, the diagnosis was given by histological study of the surgical specimen, which concluded that it was an angiomyolipoma remodelled by calcification.

The follow-up to the surgery was 5 years, and imaging was used for monitoring: for the 1st year, an ultrasound scan was performed twice a year. The other 2 years were followed by one ultrasound scan per year. Progress was favourable. Angiomyolipoma is considered a benign tumour with no risk of recurrence when excision is complete and there are no metastases or degeneration [4].

4. Conclusion

The angiomyolipoma is a benign tumor that is more commonly located in the kidney, lymph nodes, and rarely in the liver. However, it can have other very rare locations such as the neck.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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