Multiple aneurysms and Rosai-Dorfman disease: association or coincidence?

Yadala Ganesh,1 Vivek Yadala,2 Bhavana Vemula,2 Nalini Y Kammela3

1Department of General Medicine/Internal Medicine, Apollo Hospitals, Hyderabad, India
2Department of Internal Medicine, Apollo Hospital, Hyderabad, India
3Department of Radiation Oncology, Apollo Hospitals, Hyderabad, India

Correspondence to
Dr Yadala Ganesh,
yadalaganesh@yahoo.com

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DESCRIPTION

A 29-year-old man presented with a 1-month history of pain and numbness in his right arm and hypertension for 2 months, which was refractory to a combination of antihypertensive drugs. Physical examination revealed multiple pulsatile swellings in upper limbs and bilateral proptosis. No lymphadenopathy was noticed. The patient’s blood pressure was 210/105 mm Hg. CT angiogram showed aneurysmal dilations in the left external carotid artery, left internal carotid artery, both axillary arteries and both renal arteries (figure 1). The right renal artery showed thrombosis (figure 2) and haemorrhage with non-functioning right kidney. Except for serum creatinine of 1.6 mg%, laboratory reports, including antinuclear antibody profile, c-ANCA (antineutrophil cytoplasmic antibody), p-ANCA, whole body positron emission tomography (PET) CT scan and thyroid profile, were normal. The patient had undergone surgical excision of a sinonasal mass 2 years earlier, the histopathological examination of which revealed Rosai-Dorfman disease (RDD).

RDD is a rare histiocytic disorder with excessive production of white cells called non-Langerhans sinus histiocytes.1 These cells accumulate in various areas of the body, leading to malfunction. They mainly deposit in lymph nodes, particularly in the cervical region. The condition was first described as a distinct pathological entity by Rosai and Dorfman in 1969.2 Involvement of extra nodal sites including orbits with proptosis, salivary glands, testis and upper respiratory tract, has been reported.3 There have been reports of unconfirmed vasculitis in RDD and we are reporting a case of multiple aneurysms in a patient with RDD. A definite association between RDD and multiple aneurysms still needs to be established.

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REFERENCES


Figure 1 CT angiogram showing aneurysmal dilations in left internal carotid artery, left external carotid artery, both axillary arteries and both renal arteries.

Figure 2 Right renal artery showing thrombosis and haemorrhage, and aneurysm in the left renal artery.