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Angiolymphoid hyperplasia with eosinophilia along with arteriovenous malformation: an unusual case

Sir,

Angiolymphoid hyperplasia with eosinophilia is a benign cutaneous disorder clinically characterised by red angiomatous papules or nodules, usually affecting the head and neck. Arteriovenous malformations are complex disorders manifesting as isolated entitities sporadically or as a component of genetic vascular syndromes. Progressively growing arteriovenous malformations may be associated with pain, bleeding and tissue destruction. In such cases, early diagnosis is essential to prevent invasion of adjacent tissues.[1]

A 42-year-old woman was admitted to our clinic with multiple red pruritic lesions on her scalp, present for last five years. The lesions appeared gradually and progressively increased in number, with a predilection for the left-posterior aspect. The lesions were severely pruritic and bled on scratching. She denied any history of local trauma, family history suggestive of hereditary vascular condition, personal history of headache, dizziness, hypertension, seizure or any relevant drug history. Focussed physical examination of the scalp revealed more than twenty-five discrete red papulo-nodules without any background inflammation. The lesions were localised on the left-posterior aspect of the scalp, extending from the crown to occiput. Additionally, we observed a swelling on the left side of occiput, extending from the external to internal occipital protuberance. This swelling was soft without any warmth or tenderness on palpation [Figure 1]. Cervical lymphadenopathy was absent, and remaining physical and systemic examination was unremarkable.

Routine hematology and biochemistry including complete and differential blood counts, and absolute eosinophil count was within normal limits. Lesional ultrasonography demonstrated a 4-mm wide vascular structure with arterial flow at a depth of ten millimetre from the scalp surface. No abnormality was detected on brain magnetic resonance imaging with or without contrast.

We excised a representative red papule for histopathology and observed dermal proliferation of blood vessels lined by epithelioid endothelial cells exhibiting abundant eosinophilic cytoplasm and large vesicular nuclei, along with perivascular eosinophils and sparse lymphocytes. Furthermore, we observed abnormal capillaries in the reticular dermis within the hair follicle structures [Figure 2]. A final diagnosis of angiolymphoid hyperplasia with eosinophilia with concomitant vascular malformation was made based on clinico-pathological correlation.

The patient underwent surgical resection of his vascular mass along with ligation of the four feeding arteries for excision biopsy. Subsequently, the operation site was covered by a flap, Histopathology demonstrated dilated and congested arteries with partly organised thrombosis and venous channels in the dermis and subcutis, along with a solitary reactive lymph node. [Figure 3]. Almost all skin lesions resolved few days after surgery. [Figure 4].

On post-operative examination we observed mild discharge at the surgical site after one week, and prescribed oral

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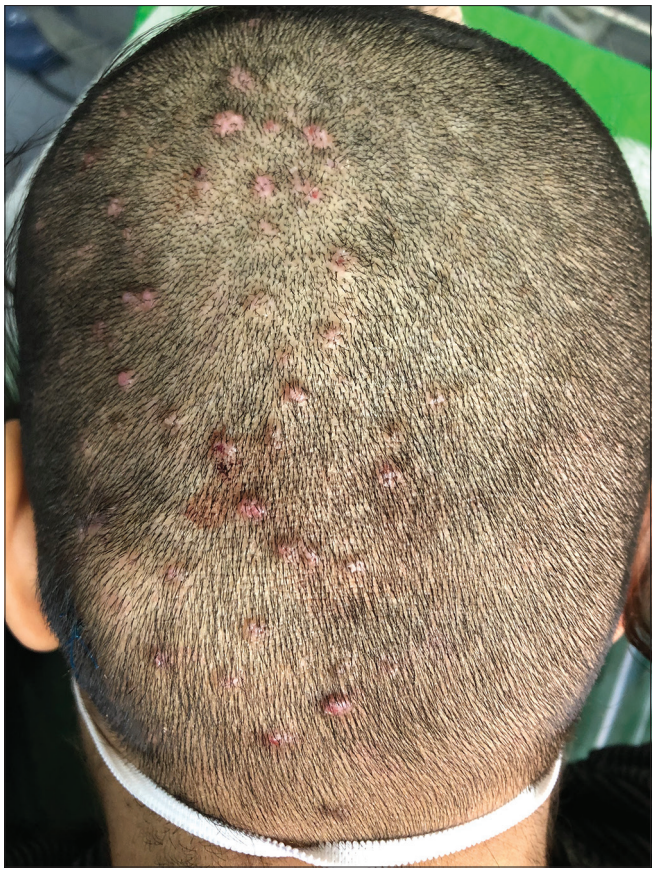

Figure 1: Multiple discrete reddish papulonodules of angiolymphoid hyperplasia with eosinophilia coexistence with a bulge of artriovenous malformation on the external protuberance of the occiput that extended to the internal protuberance of the scalp

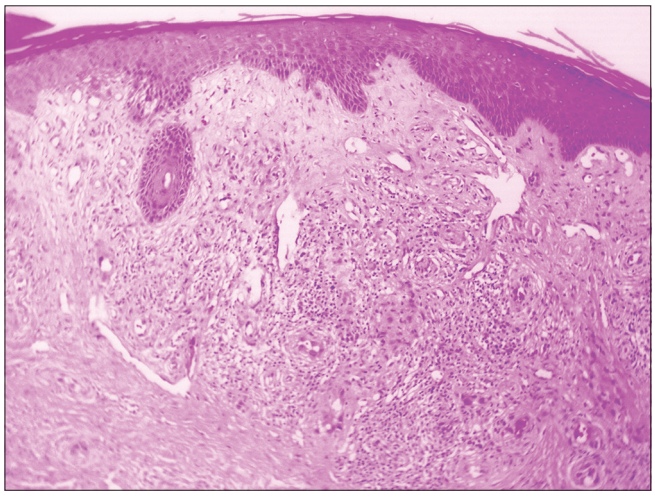

Figure 2: Multiple dilated vascular channels with inflammatory cells in the dermis (haematoxylin-eosin ×200)

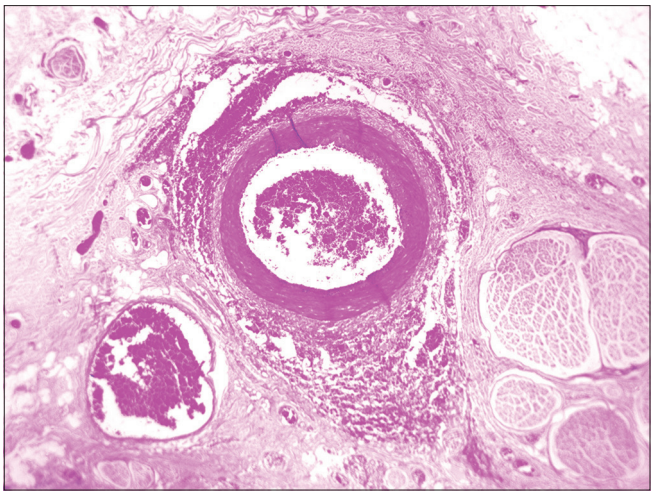

Figure 3: Dilated and congested arterial and venous channels with partly organised thrombosis associated with a reactive lymph node (haematoxylin-eosin ×200)

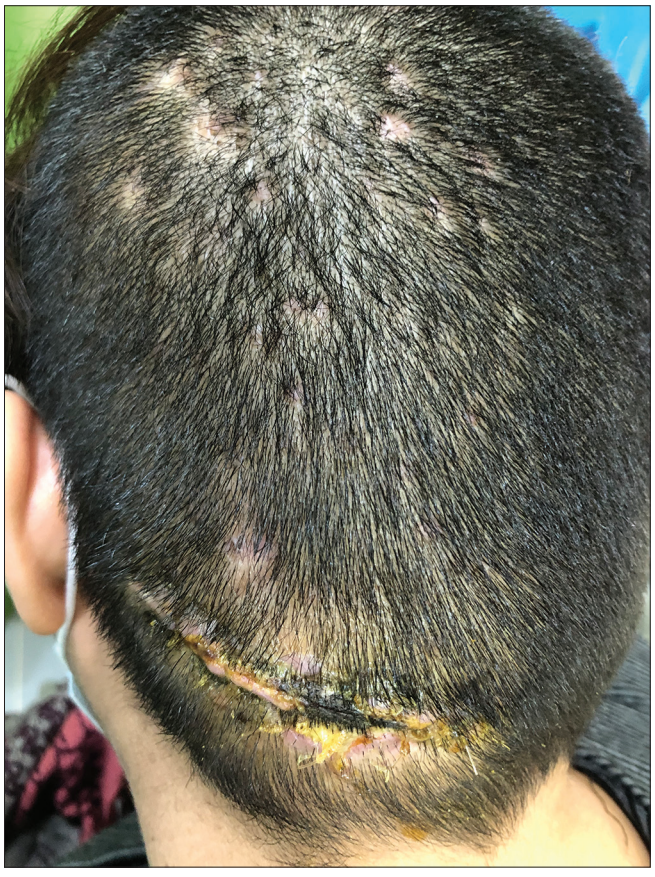

Figure 4: Subsidence of all skin lesions of angiolymphoid hyperplasia with eosinophilia after radical excision of an arteriovenous anomaly

and topical antibiotics for two weeks. The wound healed completely, and no recurrence of scalp lesions has been observed at three-months follow-up.

Angiolymphoid hyperplasia with eosinophilia predominantly involves the head and face, classically presenting as a solitary lesion, although multiple lesions have been occasionally reported.[2] Peripheral blood eosinophilia is not a constant association, and is observed in only 21% cases. Diagnosis of angiolymphoid hyperplasia with eosinophilia is based on histopathological confirmation. [3]

Vascular malformations represent disorganisation of the typical vascular pattern, mostly involving capillaries which typically act as conduits between the high-pressure arterial system and thin-walled venous system. Arteriovenous malformations indicate direct connection between the arteries or arterioles and the venous vessels without any intervening capillary bed.[4]

Arteriovenous malformations comprise 4.7–15% of all vascular malformations[1] and may be congenital or acquired (post-traumatic). However, the exact etiopathogenesis remains unknown. Almost 60% cases appear at birth, while the rest develop during adolescence or adulthood.[1] Such malformations have been reported in almost every organ or tissue, but those located outside the brain are called peripheral or extracranial arteriovenous malformations.[1,4]

The etiopathogenesis of angiolymphoid hyperplasia with eosinophilia remains obscure till date.[5] In our patient, the growth of multiple satellite skin lesions, histologically confirmed as angiolymphoid hyperplasia with eosinophilia on the left-posterior aspect of scalp, along with concomitant arteriovenous malformations on the lower scalp, suggests a causal relationship between them. The feeding arteries of this arteriovenous malformation served as the backdrop for developing secondary vascular abnormalities in the reticular dermis and subsequent cutaneous lesions. This causal relationship is evidenced by the simultaneous disappearance of almost all skin lesions following the complete excision of arteriovenous malformation, including its feeding and draining vessels. Thus, in our case, underlying arteriovenous malformation promoted the development of angiolymphoid hyperplasia with eosinophilia. Few authors have reported histologic evidence of arteriovenous shunts within angiolymphoid hyperplasia with eosinophilia, but arteriography has been performed rarely.[2] In our patient, vascular abnormalities within the skin lesion may represent arteriovenous shunts in angiolymphoid hyperplasia with eosinophilia, secondary to the occipital arteriovenous malformation. Thus, our patient developed reactive skin lesions due to underlying arteriovenous malformation.

Although surgical excision remains the first-line treatment for angiolymphoid hyperplasia with eosinophilia,[5] multiple therapeutic modalities have been tried including corticosteroids, pentoxifylline, interferon alfa-2a, imiquimod, tacrolimus, thalidomide, oral isotretinoin, acitretin, cytotoxic agents and other options such as radiotherapy, photodynamic therapy, cryotherapy, electrodesiccation and laser therapy.[5]

In conclusion, we suggest meticulous physical examination along with imaging techniques such as B-mode ultrasonography, colour Doppler and angiography if multifocal angiolymphoid hyperplasia with eosinophilia is suspected, especially involving the scalp. Appropriate therapy should be instituted after diagnostic confirming. Additionally, one must rule out underlying contributory factors such as vascular anomalies, as treatment of the primary pathology is essential in such cases, to induce regression of the skin lesions.

Declaration of patient consent

Patient's consent not required as there are no patients in this study.

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Conflicts of interest

There are no conflicts of interest.

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