

Tufted Angioma Treated with Low-dose Aspirin in a 1-year-old Filipino Boy

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Abstract

Tufted angioma (TA) is a rare, benign, vascular neoplasm of the skin. The diagnosis of this condition is infrequent due to its rare occurrence. Only 158 cases have been described as of 2015. The treatment reported in the literature is very limited with no clear guidelines on its management. Currently, there are no reported cases in the Philippines of TA treated with aspirin. This is a case of a 1-year-old Filipino boy presenting with multiple dusky red papules and plaques on the left side of the cheek, pre- and postauricular areas, parieto-occipital areas, chest, and upper back. His lesions started at 2 months of age, noted to increase in size, number, and thickness over time. Dermoscopy revealed homogenous erythematous background with perifollicular lacunae separated by thin septa. Histopathology revealed dilated vessels in the papillary dermis with proliferation of endothelial cells in lobules, surrounded by dilated crescent-shaped vascular channels in the dermis. The patient was treated with low-dose aspirin (5 mg/kg/day) once a day for 1 month with improvement. After 4 months from treatment, no new lesions, no increase in size, nor symptoms were noted. Low-dose aspirin is an effective and safe option for monotherapy of TA in pediatric patients.

Keywords: Aspirin, case report, tufted angioma

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INTRODUCTION

Tufted angioma (TA) is a rare, vascular tumor presenting as dusky red or violaceous infiltrating macules, plaques, or nodules.^[1] They are located on areas of the head, neck, trunk, or extremities and can be associated with tenderness, hyperhidrosis, or hypertrichosis.^[2] Most cases occur within the 1st year of life, during infancy and early childhood.^[3] The etiology of TA is still unknown.^[1]

There are no reported treated cases of TA in the Philippines. Currently, no clear guidelines on its management exist, and treatment is limited due to its rare nature.^[3] TA can have variable clinical outcomes with the occurrence of complete

or partial spontaneous regression or persistence of lesions over the years.^[4]

CASE REPORT

A 1-year-old Filipino boy presented with multiple dusky red papules and plaques on the left side of the cheek, pre- and postauricular areas, parieto-occipital areas, chest, and upper back. Lesions developed when the patient was at 2 months of age, initially presenting with a solitary pinkish papule on the lower left cheek. The papule evolved into plaques, extending to the left preauricular area, left earlobe,

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and left parieto-occipital area with a gradual increase in size and thickness of lesions. At 10 months of age, new erythematous papules and plaques were noted on the upper chest, left shoulder, and middle upper back. The mother noted the patient to occasionally scratch affected areas and would become irritable when lesions are palpated. At 1 year of age, the persistence of lesions with increasing size and thickness prompted consultation.

Medical history was unremarkable. The patient's older sister (9 years of age) has a vascular malformation on her left forearm, noted since birth, with no resolution. No other family members have vascular malformations, and there are no other known heredo-familial diseases.

Physical examination revealed multiple, discrete to confluent, well-defined, indurated, dusky red papules and plaques on the left cheek, left pre- and postauricular areas, left parieto-occipital scalp, upper chest, left shoulder, and mid-upper back measuring 0.5 cm × 0.5 cm to 4 cm × 7 cm. Dermoscopic examination revealed homogenous erythematous background with perifollicular lacunae separated by thin septa. Histologic examination revealed dilated vessels and proliferation of endothelial cells in lobules, surrounded by dilated crescent-shaped vascular channels in the dermis [Figure 1]. Complete blood count and bleeding parameters were unremarkable. Magnetic resonance imaging and ultrasound imaging were advised; however, the mother did not consent.

The patient was treated with low-dose aspirin 100 mg/tablet, ½ tablet once a day (5 mg/kg/day) for 1 month with significant improvement. The patient was unable to follow up after 1 month of treatment. After 4 months, the patches

and plaques were more hyperpigmented with a decrease in size and thickness. After 4 months, the patches and plaques were more hyperpigmented with a decrease in size and thickness noted and with no new lesions, no relapse nor worsening of symptoms, and no adverse effects [Figure 2]. Dermoscopic findings showed hyperpigmented patches with perifollicular lacunae separated by thin septa [Figure 3].

DISCUSSION

The patient is clinically and histologically diagnosed with TA. This is the first reported case of TA in the Philippines treated successfully with low-dose aspirin monotherapy. Only one other case was previously published in the *Journal of the Philippine Dermatological Society* in 1998, a case of a 12-year-old boy histologically confirmed with TA. No therapeutic interventions were done due to the paucity of symptoms and benign progression of lesions.^[5]

The definitive diagnosis of TA is made histologically with its characteristic cannonball appearance of multiple scattered lobules or tufts lobules of endothelial cells and capillaries in the dermis.^[6] Tufted hemangiomas may express endothelial and lymphatic vascular markers such as CD31, CD34, VEGF-A, and D2-40. Dermoscopy findings would reveal a red background intermixed with few telangiectasias, thin white septa, and prominent perifollicular brown areas. Dermoscopy can also be utilized as a tool for monitoring the response of these tumors to treatment.^[7] Genetic testing can also be done. A study by Broek *et al.*^[8] showed two cases of TA with mutations in NRAS and GNA14.

There are three clinical patterns of TAs: (1) TAs without complications, (2) TAs with chronic coagulopathy without thrombocytopenia, and (3) TAs with Kasabach–Merritt phenomenon. Kasabach–Merritt phenomenon is characterized by thrombocytopenia, low fibrinogen, and elevated D-dimer levels. A complete blood count and coagulopathy testing is needed for its assessment.^[2]

Treatment options for TAs have variable efficacy.^[3] Pharmacologic treatments include ticlopidine, vincristine plus aspirin, heparin, corticosteroids, and sirolimus.^[2] Cryotherapy and electrocautery showed unsuccessful results. Pulsed dye and argon-tunable dye lasers have variable results. Surgical excision can be done for symptomatic patients with small lesions; however, recurrences occur. Despite treatments reported, there is still no uniformly effective and reproducible approach identified.^[9]

The patient was treated with aspirin for 1-month duration, which he tolerated without adverse effects. No new lesions were also noted after 3 months of observation. Only two

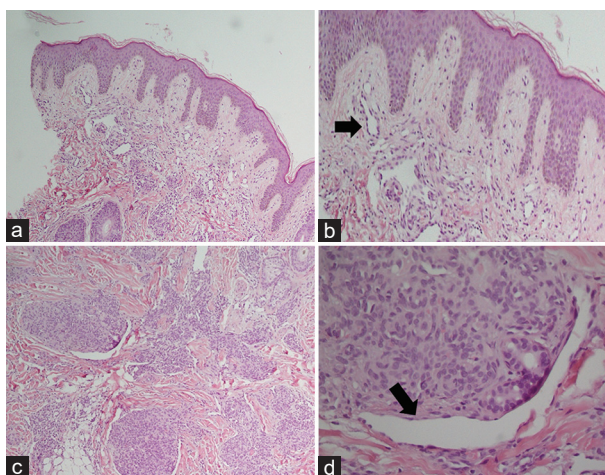


Figure 1: Histopathologic findings: (a) Normal epidermis, (b) Dilated vessels in the papillary dermis (black arrow), (c) Endothelial cells in lobules surrounded by dilated crescent-shaped vascular channels, (d) Dilated crescent-shaped vascular channels (black arrow)



Figure 2: Comparison of lesions at baseline and 4 months after on (a,b) left cheek, left pre- and postauricular areas, left parieto-occipital scalp (c) upper chest, and (d) mid-upper back

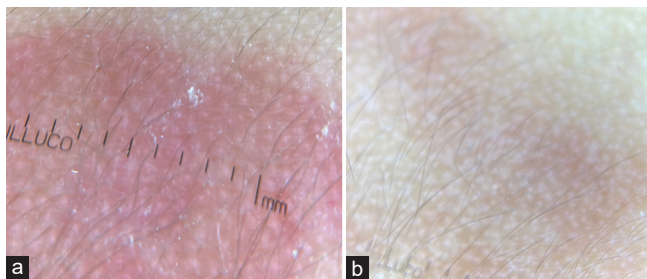


Figure 3: Comparison of dermoscopic findings of erythematous plaque on back (a) at baseline (b) and after 4 months

patients were likewise treated with aspirin for a duration of 3.5 and 7 years, respectively, as reported in a study by Javvaji and Frieden in 2013.^[10] The evolution of the disease varies considerably with the occurrence of complete or partial spontaneous regression or persistence of lesions over the years.^[7] Therefore, close follow-up and monitoring is essential.

Aspirin is an orally administered agent that interferes with platelet function by inhibiting platelet release reaction. Lower doses of aspirin are important for minimizing side effects, specifically decreasing the risk of Reye's syndrome. No reported cases of TA treated with aspirin developed Reye's syndrome. Other possible side effects include encephalopathy and liver failure; hence aspirin should be used with caution in children.^[10] The patient was treated with aspirin at a dose of 5 mg/kg/day for 1 month with a significant improvement of TA, with no systemic adverse effects. Therefore, low-dose aspirin is an acceptable treatment modality for cases of TAs in the pediatric age group.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and

other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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