

Multiple Pyogenic Granulomas within Port-Wine Stain

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Pyogenic granuloma is a rapidly growing vascular tumor that often arises at sites of minor trauma. It can also appear within an existing capillary malformation (port-wine stain), but most commonly there is no history of preexisting dermatologic conditions. We report a rare case of multiple pyogenic granulomas within a port-wine stain without any trauma history in early childhood. This case suggests that in a few cases there may be an association between vascular tumors and vascular malformations. (*Ann Dermatol* 16(4) 201~203, 2004)

Key Words: Port-wine stain, Pyogenic granuloma

INTRODUCTION

There were several clinical or histological settings in which vascular tumors and vascular malformations were associated¹. Pyogenic granuloma (PG) is a common, benign vascular tumor of the skin, and port-wine stain (PWS) is a congenital vascular malformation. These two lesions rarely occur in association. As reported earlier^{2,3}, formation of a PG within the PWS supports the assumption that arteriovenous anastomoses are associated with its pathogenesis.

We report a case of multiple PGs developed within a PWS without any trauma or foreign body reaction, and the literatures are reviewed.

CASE REPORT

A 3-year-old Korean boy presented with multiple papules in a segmental distribution within a faint port-wine stain on the left side of his central back for 1 month. The port-wine stain had been present since birth. At first, there were a few bright red colored macules within the stain, and then they grew slowly and increased in numbers. There were no

associated abnormalities and his past history, including any trauma, was otherwise unremarkable. Examination revealed multiple slight elevated, bright red papules, 1 - 5 mm in diameter (Fig. 1). All lesions were inside the margins of the port-wine stain. The biggest one was lobulated and pedunculated, and excised for histopathologic examination.

Histopathologically, the tumor was composed of a variably dilated network of blood-filled capillary vessels and groups of poorly canalized vascular tufts and lobulated by fibrous stroma, which indicated



Fig. 1. Multiple pyogenic granulomas within a port-wine stain on the back.

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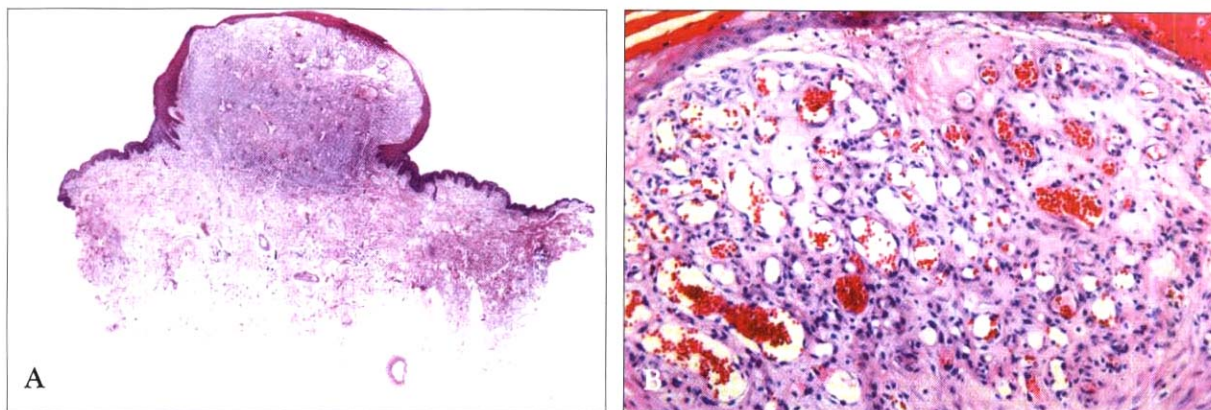


Fig. 2. A. Low-power view of pyogenic granuloma showed lobules of capillaries and dilated capillaries in subjacent dermis (H & E, $\times 12.5$). B. Lobules of capillaries were well outlined by fibrous stroma (H & E, $\times 100$).

typical pyogenic granuloma (Fig. 2A and B). There were a small number of greatly ectatic, thin-walled capillaries at the periphery of the lesion.

DISCUSSION

In 2000, Garzon *et al*¹ identified clinical or histologic settings in which vascular tumors and vascular malformations were associated: (1) coexistence of hemangioma and vascular malformation at the same anatomic site or in close proximity; (2) pyogenic granuloma arising within a vascular malformation; (3) hemangioma associated with developmental vascular anomalies; (4) spindle-cell hemangioendothelioma and venous malformation; (5) kaposiform hemangioendothelioma and lymphatic malformation. They suggested that in a small minority of patients there was an association between vascular tumors and vascular malformations.

The occurrence of a PG within a PWS is a rare event in early childhood. There were several reports of cases which had developed during pregnancy, after laser treatment, or spontaneously. Some theories have been put forward to explain the pathogenesis of PG.

It is well known that vascular lesions, including PG, occur with a higher frequency during pregnancy and hormonal factors are thought to play a major role in the development of these lesions. Barter *et al.*⁴ reviewed hemangiomas in pregnancy and reported several cases. One of their cases was a hemangioma resembling a PG developed in the PWS

of a pregnant woman. Katta *et al.*⁵ also reported a PG arising spontaneously within a PWS during pregnancy and suggested that hormonal factors played a role in the association.

Beers *et al.*⁶ believed that the formation of a PG following laser treatment of a PWS was not rare, but it might occur more frequently than the literature would suggest. It would appear that the PWS might already be predisposed to the spontaneous occurrence of this lesion, perhaps because of the increased numbers of microscopic arteriovenous anastomoses. Thus, the trauma of laser therapy might easily accelerate such a tendency. In Korea, Tak *et al.*⁷ reported the multiple PGs developed in a PWS following Nd-YAG laser therapy.

It has been postulated that arteriovenous anastomoses are associated with the pathogenesis of PG. Rusin and Harrell² suggested that PWS was composed of numerous arteriovenous fistulae and PG might also represent a form of acquired arteriovenous fistula. They suggested that highly vascularized areas such as the fingers, hands, lips, and tongue might be favored sites for PG due to increased numbers of microscopic arteriovenous anastomoses in these locations. Swerlick and Cooper³ supported that the spontaneous development of PG in a PWS might be associated with microscopic arteriovenous anastomoses.

Some authors suggested that an underlying inflammatory or neoplastic disease could trigger spontaneous development of multiple PGs with release of angiogenic factors⁸. Shimizu *et al.*⁹ demonstrated the involvement of nitric oxide synthase effector mecha-

nism in promoting angiogenesis and the rapid growth of PG.

Pyogenic granuloma within a port-wine stain have been successfully treated with pulsed dye laser⁷, cryotherapy³, excision^{3,5}. Spontaneously developed pyogenic granuloma in pregnancy may disappear completely after pregnancy⁴. But, if there is a cosmetic problem or frequent bleeding, it should be treated⁴. In our case, we excised two pyogenic granulomas which bled easily after minor trauma, but didn't treat the other uncomplicated ones.

Our observation of the spontaneous development of multiple PGs within a PWS is interesting, and possible pathogenesis may be associated with microscopic arteriovenous anastomoses.

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