

Pyogenic Granuloma Arising Within a Port-wine Stain

Daniel J. Sheehan, MD; Jack L. Leshner, Jr, MD

GOAL

To gain a thorough understanding of pyogenic granulomas

OBJECTIVES

Upon completion of this activity, dermatologists and general practitioners should be able to:

1. Explain the presentation of pyogenic granulomas.
2. Describe the risk factors for development of pyogenic granulomas within port-wine stains.
3. Discuss the pathogenesis of pyogenic granulomas and port-wine stains.

CME Test on page 194.

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Pyogenic granulomas are common overgrowths of vascular tissue that usually arise on the face, lips, or hands after episodes of minor trauma or during pregnancy. Pyogenic granulomas rarely have been reported to arise in congenital capillary malformations such as port-wine stains, a presentation that most often occurs after laser treatment of port-wine stains or in the setting of

pregnancy. This co-occurrence of the 2 lesions represents an underreported event, and the presentation can be alarming or mimic malignancy when there is no prior history of trauma or other known circumstances in which pyogenic granulomas occur. We report a case of a pyogenic granuloma emerging within a port-wine stain in a 35-year-old man with no predisposing factors. A review of the literature regarding pyogenic granulomas that arise within port-wine stains is presented and yields informative conclusions regarding the clinical scenarios where this sometimes alarming event is likely to happen.

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From the Section of Dermatology, Department of Medicine, Medical College of Georgia, Augusta. Dr. Sheehan is a dermatology resident, and Dr. Leshner is Professor and Chief. Reprints: Jack L. Leshner, Jr, MD, Section of Dermatology, Medical College of Georgia, 1004 Chafee Ave, Augusta, GA 30904 (e-mail jlesher@mail.mcg.edu).

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Pyogenic granuloma, often referred to by many other names, including *lobular capillary hemangioma*, is a benign vascular hyperplasia that tends to occur on the gingiva, lips, nasal mucosa, face, and hands.^{1,2} The lesion often is preceded by minor trauma, has a history of bleeding easily, and appears as a pedunculated or sessile papule with a rough red surface. There is a well-known tendency toward local recurrence after treatment.³ Results of a histopathologic examination shows lobular aggregates of capillaries and venules with plump endothelial cells.⁴ The pathogenesis of this lesion is poorly understood.

Port-wine stains are a type of nevus flammeus that present as a deep red or purple macule and are typically unilateral.⁴ Unlike salmon patches, the other type of nevus flammeus, port-wine stains tend to persist into adulthood.⁵ These congenital malformations are composed of capillary and venule ectasia in the dermis, and the ectasias progress over the course of a lifetime with increasing erythrocyte stasis.⁶⁻⁸ As Finley⁶ pointed out, in time, the port-wine stain can develop a thickened “cobblestone pattern,” as well as nodules that are

thought to be arteriovenous malformations or “localized tumor formations.” Klapman and Yao⁸ recently noted that the thickening and nodules are most likely to occur in the areas of the face innervated by the second and third branches of the trigeminal nerve than in other parts of the body. Port-wine stains have been associated with other abnormalities such as nevus anemicus, glaucoma, choroidal angiomas, and Sturge-Weber syndrome.⁴ The pathogenesis of the port-wine stain has been a controversial issue, but one possibility is that it represents a deficiency of sympathetic innervation of blood vessels.⁴

The occurrence of vascular neoplasms arising in capillary malformations is a rare, but documented, event.⁹ Pyogenic granulomas infrequently have been reported to occur in association with other vascular abnormalities such as port-wine stains or hemangiomas, particularly after laser treatment.^{10,11} A number of other vascular neoplasms, such as capillary hemangiomas, tufted angiomas, and cavernous hemangiomas, have been reported in association with port-wine stains, as well.^{6,12,13} Very few pyogenic granulomas have



Figure 1. Friable tumor with surrounding contact dermatitis emerging from underlying port-wine stain on left upper back.

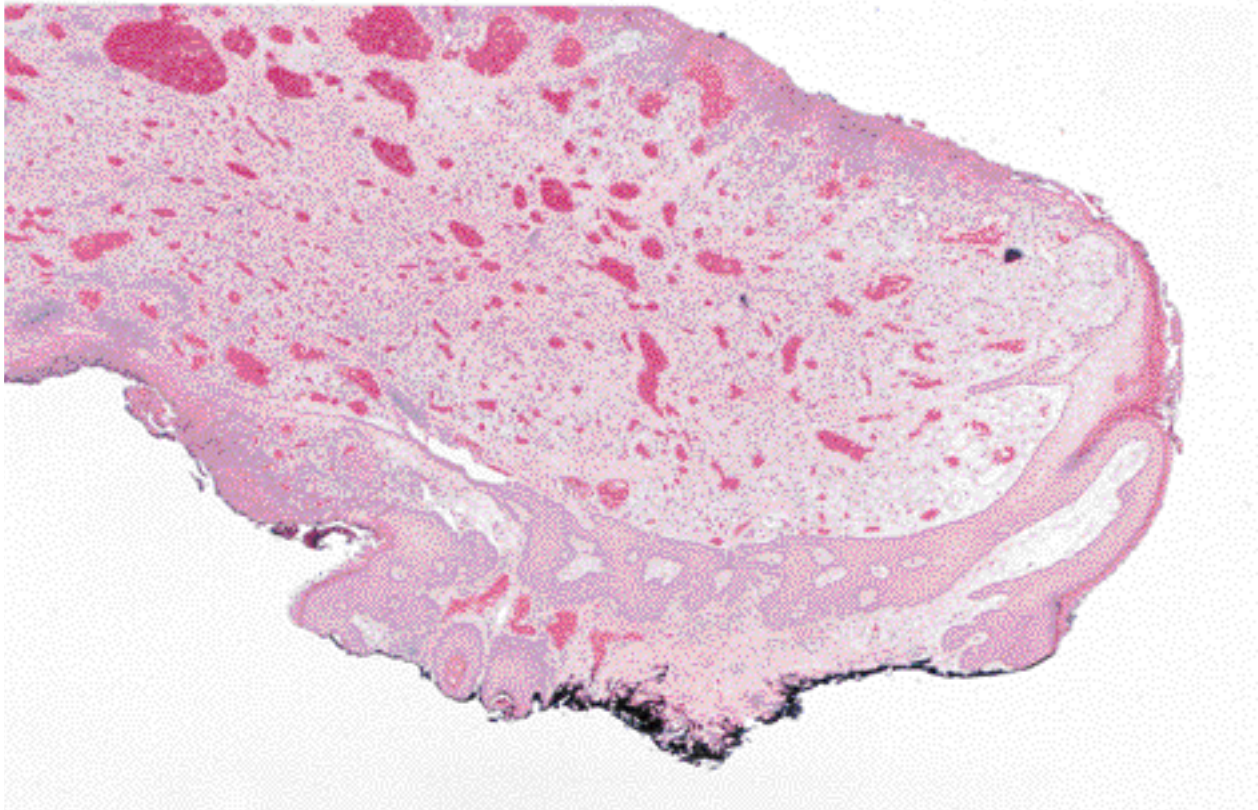


Figure 2. Exophytic papule composed of lobules of vascular tissue in a loose stroma. Underlying the papule in the dermis, there is visible vascular ectasia and erythrocyte stasis (H&E, original magnification $\times 4$).

arisen in port-wine stains without a history of trauma, laser treatment, or pregnancy. We now report a pyogenic granuloma occurring within a port-wine stain without a history of trauma or laser treatment in a man, and we contend that this occurrence offers unique insight into the pathogenesis of these poorly understood but commonly encountered entities.

Case Report

A 35-year-old Asian man with a history of a port-wine stain on his left upper back presented for evaluation of a “mushroom” growing within his birthmark (Figure 1). He noted that some lesion had been present in this location for several months but that it had grown rapidly in the previous 3 weeks and begun to bleed easily. There was no history of trauma or treatment of the area. On physical examination, the patient had a 1.4-cm pedunculated, moist tumor with a rough surface within a port-wine stain with surrounding contact dermatitis from the bandaging he had used to protect the friable papule. Saucerization was performed, and the extremely vascularized base was cauterized. Results

of histopathologic examination revealed lobules of vascular hyperplasia consistent with pyogenic granuloma (Figure 2). Vascular ectasia was seen below the pyogenic granuloma that represented the underlying port-wine stain.

Comment

The Table documents the reported cases of pyogenic granulomas arising within port-wine stains.^{3,10,11,13-23} Many have occurred after laser treatment of port-wine stains.^{11,15-17,24,25} Several cases have occurred after other manipulation of port-wine stains, such as Grenz-ray therapy¹⁵ or local trauma.²⁰ Three cases occurred during pregnancy, which is a hormonal state that is known to predispose the patient to the formation of pyogenic granulomas, including one patient who previously was treated with the 577-nm pulsed dye laser and subsequently became pregnant.^{14,15,21} Holloway et al¹⁸ noted the development of a proliferation that had a superficial component of pyogenic granuloma and a deeper component of cavernous hemangioma. Other authors have seen the occasional association of port-wine stains and cavernous

Reported Cases of Pyogenic Granulomas Arising in Port-Wine Stains*†

Reference	Age, y	Sex	Location(s)	Notes
Barter et al ¹⁴	33	F	Cheek	Patient was pregnant, reported as a hemangioma
Warner and Jones ³	8	M	Sternum	Recurred with multiple satellites
Swerlick and Cooper ¹⁰	22	F	Thigh	Developed several pyogenic granulomas in 3 y
	8	M	Shoulder/back/neck	
Lanigan and Cotterill ¹⁵	21	F	Neck	Occurred after treatment with 577-nm pulsed dye laser, patient was pregnant when it arose
	17	M	Forehead	Occurred after treatment with 577-nm pulsed dye laser
	61	F	Buttock to calf	Developed 6 recurrences
	16	M	Scalp and neck	Was previously treated with grenz-ray therapy
Beers et al ¹⁶	35	F	Face	Occurred after treatment with argon/carbon dioxide lasers
Garden et al ¹⁷	?	?	?	Occurred after treatment with pulsed dye laser
Holloway et al ¹⁸	57	M	Scalp and neck	Giant proliferative hemangioma had superficial component of pyogenic granuloma and deeper component of cavernous hemangioma
Abd-El-Raheem et al ¹¹	29	F	Back	Occurred after treatment with pulsed dye laser
Hagiwara et al ¹⁹	5	F	Forehead	Patient had phacomatosis pigmentovascularis type IIb and Sturge-Weber syndrome
Kim et al ¹³	27	F	Shoulder	
	25	F	Chest	
Lee et al ²⁰	16	M	Eyebrow	History of local trauma (squeezing)
Katta et al ²¹	16	F	Nose	Patient was pregnant
Valeyrie et al ²²	31	F	Heel	3.5-cm tumor simulated amelanotic melanoma
	67	M	Wrist	4-cm tumor simulated malignant vascular proliferation
Castanedo-Cazares et al ²³	25	F	Submaxillar area	
Present case	35	M	Back	

*F indicates female; M, male.

†Listed in chronological order.

hemangiomas.⁶ Not surprisingly, pyogenic granulomas have occurred within port-wine stains in the setting of other syndromes, such as phacomatosis pigmentovascularis and Sturge-Weber syndrome.¹⁹

Only rarely does the pyogenic granuloma seem to emerge from the port-wine stain without a history of manipulation or pregnancy, as it did in the present case. When there is no history of predisposing

factors, the presentation can be very alarming, simulating other processes such as amelanotic melanoma.^{22,23} Interestingly, as opposed to the thickening and nodules that have shown to be more likely to develop in port-wine stains in the distribution of the trigeminal nerve, most of the pyogenic granulomas that have arisen in port-wine stains did so outside of this distribution, occurring more often on the neck, trunk, or extremities.

Clearly, the co-occurrence of these 2 vascular abnormalities must offer some information on the pathogenesis of these poorly understood conditions. Several years ago, Swerlick and Cooper¹⁰ speculated that the co-occurrence supported the possibility of pyogenic granulomas developing at the sites of microscopic arteriovenous anastomoses. More recently, as discussed by Garzon et al,⁹ a number of studies indicate that abnormalities in embryonic vasculogenesis are involved in forming vascular malformations such as port-wine stains, and these same abnormalities may predispose patients toward forming vascular hyperplasia or tumors. The well-known hormonally driven vascular changes that happen during pregnancy also may be able to generate pyogenic granulomas in the setting of port-wine stains.²¹

On a basic science level, chemical mediators such as inducible nitric oxide synthase have been implicated in the formation of pyogenic granulomas, and mast cells have been found to be dramatically increased in the settings of both pyogenic granulomas and port-wine stains, though this may not be evidence of a cause-and-effect relationship.^{26,27} With regard to laser-induced pyogenic granulomas in port-wine stains, speculation has involved several factors including local tissue trauma in an area of increased microscopic arteriovenous anastomoses, retention of abnormal vasculature, and nonspecific laser interaction with tissue.^{15,16}

Pyogenic granulomas tend to develop on the fingers, lips, face, and hands (places known to be rich in arteriovenous anastomoses), suggesting that this vascular phenomenon tends to occur with minor trauma in the setting of such anastomoses.^{13,20} This conclusion is consistent with the available data. The present case of a pyogenic granuloma arising without recognized trauma in a port-wine stain also supports this hypothesis.

Conclusion

Pyogenic granuloma is a well-known and probably underreported complication of port-wine stains. Unlike thickening and nodule formation that tend to occur in port-wine stains in the trigeminal nerve distribution, pyogenic granulomas arising in port-

wine stains tend to occur more commonly on the neck, trunk, or extremities. The pyogenic granuloma should not be confused with a true malignancy because it represents hyperplasia of vascular tissue in response to trauma, but the performance of an excisional biopsy is warranted.

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