Eruptive Disseminated Pyogenic Granulomas following Lightning Injury

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Abstract

Background: Pyogenic granuloma (PG) is a common benign acquired vascular tumor. It classically presents as a solitary friable nodule on the face or distal extremities. Disseminated eruption is rare and can occur spontaneously or secondary to various triggers, including burn injury. To date, the literature reports only 13 cases of eruptive PGs following burn injury, most from exposure to boiling milk or water. We report the first case of disseminated eruptive PGs following a lightning injury.

Case: A 17-year-old previously healthy boy developed second- and third-degree burns following lightning injury. Two weeks later, he developed widespread dark-purple polypoid exophytic tumors ranging from 1 to 10 cm in diameter extending beyond the limits of the initial burn injury. The lesions were friable and often formed erosions and crusts. The patient was otherwise well and laboratory and microbiological investigations were normal. Excisional biopsy of a lesion was diagnostic of PG and the patient was treated with surgical excision of the lesions, without recurrence. Conclusion: The exact pathogenesis of multiple PGs remains unknown. Several pathogenic mechanisms have been suggested, including production of angiogenic factors that stimulate endothelial proliferation and formation of minute arteriovenous fistulas by trauma.

Introduction

Pyogenic granuloma (PG), also known as lobular capillary hemangioma, was first described by Poncet and Dor in 1897 [1]. Although its name has stood the test of time, it is really a misnomer because this benign acquired vascular tumor is neither infectious nor granulomatous [2]. It classically presents as a solitary friable nodule on the face or distal extremities in children or young adults, but may occur anywhere on the skin or mucous membranes. Two cases of congenital PGs have been reported to date [3]. The typical lesion develops over weeks and eventually becomes a fibrotic ‘angioma’. Occasional nodules may spontaneously involute [4]. Predisposing factors include pregnancy, vascular malformations and skin irritation resulting from trauma, infection, inflammatory skin conditions and oral retinoid therapy [3]. In addition, other medications and neoplastic processes have been linked to the development of PGs (table 1, left column). Despite their benign nature, treatment is often warranted because of associated pain and bleeding. Common therapeutic options for solitary lesions include cryotherapy, silver nitrate application, laser ablation, shave excision with electrodesiccation and complete surgical excision with direct closure [2, 5].

Aside from classic solitary mucocutaneous PG, several other clinical variants have been recognized (table 1, right column) [2, 3, 6–11]. ‘Epulis gravidarum’ (granuloma gravidarum) is a term reserved for PG arising in the oral mucosa during pregnancy, usually during the second or third trimester of gestation [10]. Following treatment, PGs may recur as isolated lesions, but when recurrence is accompanied by multiple satellite nodules, this phenomenon is known as Warner and Wilson-Jones syndrome [2]. Other rare variants in-
The eruption of multiple PGs is rare and can be classified as 'localized' or 'disseminated', depending on the extent of skin involvement. Disseminated eruption is characterized by the sudden appearance of numerous widespread PGs, either spontaneously or secondary to various triggers, including burn injury (table 1, left column) [7]. To date, only 13 cases of eruptive PGs (localized and disseminated) following burn injury have been described in the literature, most after contact with boiling milk or water [12–18]. We report the first case of disseminated eruptive PGs following lightning injury.

**Case Report**

A 17-year-old previously healthy farmer from Huancavelica, Peru developed second- and third-degree burns over his face, chest and limbs following lightning injury. He also sustained a femoral fracture requiring surgery. Two weeks following the initial accident, he was transferred to the Cayetano Heredia Hospital in Lima, Peru. At that time, his burn wounds were healing, but he developed widespread dark-purple, polypoid and exophytic tumors ranging from 1 to 10 cm in diameter over his face, chest and limbs, extending beyond the limits of the initial burn injury (fig. 1a). These lesions were friable and often formed ero-
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Discussion

In the present case, the history of rapid growth, friability and vascular appearance were typical findings of PG. The differential diagnosis of such lesions includes Kaposi’s sarcoma, verruca peruana, bacillary angiomatosis, disseminated atypical mycobacterial infection and angiolymphoid hyperplasia with eosinophilia. These entities were ruled out by histopathology and/or microbiological cultures. The exact pathogenesis of multiple PGs is unknown, however they are considered to be a vascular proliferative response to various angiogenic stimuli, including trauma, increased levels of female sex hormones, infections, viral oncogenes, medications, inflammatory disorders, vascular malformations and malignancies [4]. Several pathogenic mechanisms have been suggested, including production of angiogenic factors that stimulate endothelial proliferation, formation of minute arteriovenous fistulas by trauma and female hormone-enhanced angiogenesis [7, 19]. A local trauma, such as a burn, is typically followed by sequential phases of wound healing. The early inflammatory phase leads to innate immune system activation as well as release of cytokine and growth factors that include vascular endothelial growth factor (VEGF) [20]. It is followed by a proliferative phase, with formation of granulation tissue consisting of macrophages, fibroblasts and endothelial cells, which will fill the wound area. In the final remodeling phase, the granulation tissue is replaced by a scar. Surprisingly, PG is not listed among the complications of wound healing, despite the established relation with skin injury (up to 50% of patients with PG describe previous injury) and histopathology resembling granulation tissue [21]. Recently, Godfraind et al. [22] performed genome-wide transcriptional profiling of blood vessels derived from PG. Their findings suggest that PG may result from tissue injury, followed by a deregulated wound healing process, during which vascular growth is driven by FLT4 (VEGF signaling) and nitric oxide pathways. Most of the genes found to be upregulated in PG (BDKRB2, NTPDase1, ENG, FLT4, ST3GAL6, NDST1, HSPG2, HOXD3, EMCN, PODXL and ENTPD1) are known to be involved in vascular injury, wound healing or cancer angiogenesis [22].

Despite frequent occurrence of burn injuries, only 13 cases of eruptive PGs following a burn have been previously described (table 2). In all cases, PGs developed within 2 weeks of burn injury, most frequently involving contact with hot water or milk. Ten patients were children. There was no apparent gender predilection. Interestingly, seven of the reported cases were from Turkey. In six patients, the lesions resolved spontaneously or with antibiotic treatment, while the other patients were treated surgically.

Our patient represents the first case of eruptive PGs following lightning injury. Vascular injury can result from electrical coagulation of small blood vessels or direct cellular damage [23]. Imbalance of angiogenesis promoters and inhibitors following a burn injury may represent the underlying pathophysiologic process [24, 25]. Ruan et al. [26] documented an increase in VEGF production following electrical burn in an animal model, which correlated with clinical phases of wound healing. In addition, formation of minute arteriovenous fistulas has been hypothesized to occur following trauma and laser treatment and may contribute to PG formation [27–29].

Conclusion

We report a case of disseminated eruptive PGs occurring following lightning injury. Some patients with eruptive PG ex-
experience spontaneous involution of their tumors. For this reason, observation for spontaneous regression may be a reasonable therapeutic approach. In case of tumor persistence, surgical excision offers the lowest recurrence rate. We hope this case report and review will help clinicians to effectively recognize and manage this condition.

Acknowledgment

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Disclosure Statement

The authors report no conflicts of interest.

Table 2. Previously reported cases of PG following burn injury

<table>
<thead>
<tr>
<th>Reference</th>
<th>Age, years</th>
<th>Country of origin</th>
<th>Gender</th>
<th>Trigger</th>
<th>Onset of PG</th>
<th>Location</th>
<th>Number of lesions</th>
<th>Degree of burn</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>de Kaminsky et al., 1978 [16]</td>
<td>1.5</td>
<td>Argentina</td>
<td>F</td>
<td>boiling milk</td>
<td>1 week</td>
<td>right arm, trunk, face</td>
<td>18</td>
<td>second</td>
<td>electrocoagulation</td>
</tr>
<tr>
<td>Momeni et al., 1995 [18]</td>
<td>1.5</td>
<td>Iran</td>
<td>M</td>
<td>boiling milk</td>
<td>2 weeks</td>
<td>face, neck, trunk, thigh</td>
<td>28</td>
<td>second/third</td>
<td>spontaneous resolution</td>
</tr>
<tr>
<td></td>
<td>5</td>
<td>Iran</td>
<td>F</td>
<td>boiling milk</td>
<td>2 weeks</td>
<td>trunk, thigh</td>
<td>65</td>
<td>second</td>
<td>spontaneous resolution</td>
</tr>
<tr>
<td></td>
<td>35</td>
<td>Iran</td>
<td>F</td>
<td>boiling milk</td>
<td>unknown</td>
<td>face</td>
<td>90</td>
<td>second</td>
<td>spontaneous resolution</td>
</tr>
<tr>
<td>Ceyhan et al., 1997 [15]</td>
<td>1.5</td>
<td>Turkey</td>
<td>F</td>
<td>boiling milk</td>
<td>1 week</td>
<td>arm, trunk, face</td>
<td>unknown</td>
<td>second</td>
<td>excision</td>
</tr>
<tr>
<td>Aliğaçoğlu et al., 2006 [13]</td>
<td>5</td>
<td>Turkey</td>
<td>F</td>
<td>unknown</td>
<td>2 weeks</td>
<td>upper arm</td>
<td>unknown</td>
<td>unknown</td>
<td>excision</td>
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<tr>
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<td>2</td>
<td>Turkey</td>
<td>M</td>
<td>boiling milk</td>
<td>10 days</td>
<td>left forearm</td>
<td>5</td>
<td>unknown</td>
<td>excision</td>
</tr>
<tr>
<td>Ceyhan et al., 2007 [1]</td>
<td>1.5</td>
<td>Turkey</td>
<td>M</td>
<td>boiling water</td>
<td>2 weeks</td>
<td>left arm</td>
<td>unknown</td>
<td>second</td>
<td>erythromycin</td>
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<td>Özbayoğlu et al., 2011 [12]</td>
<td>8</td>
<td>Turkey</td>
<td>M</td>
<td>flame</td>
<td>2–3 weeks</td>
<td>abdomen</td>
<td>unknown</td>
<td>second</td>
<td>excision</td>
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<tr>
<td>Durgun et al., 2013 [17]</td>
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<td>F</td>
<td>boiling water</td>
<td>2 weeks</td>
<td>face, neck</td>
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<td>second</td>
<td>excision</td>
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<tr>
<td></td>
<td>7</td>
<td>Turkey</td>
<td>M</td>
<td>oven</td>
<td>2 weeks</td>
<td>left forearm</td>
<td>unknown</td>
<td>second</td>
<td>excision</td>
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<tr>
<td>Liao et al., 2006 [19]</td>
<td>41</td>
<td>China</td>
<td>M</td>
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<td>10 days</td>
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<td>cefazolin, amikacin</td>
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<td>19</td>
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<td>M</td>
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References


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