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Case Report

Dermatology Section

Multiple, Eruptive and Generalised Lobular Capillary Haemangiomas- A Rare Morphological Variant

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ABSTRACT

Lobular Capillary Haemangioma or Pyogenic Granuloma (PG) is a common benign, vascular proliferative lesion, usually occurring at sites of preceding trauma. It is known to occur more commonly in children and young females. The most common presentation is a solitary lesion. Multiple, eruptive PG have been reported after antecedent burns, trauma and following retinoid therapy and as satellite lesions after treatment of a primary lesion. A 14-year-old female, without any co-morbidities presented with spontaneous onset of multiple, eruptive and generalised skin coloured papules and nodules over face, neck, upper chest and upper back. The lesions were eruptive in nature, involved the back, face and upper trunk over duration of four months. There was intermittent bleeding from the lesions after trivial trauma. Clinical diagnosis of lobular capillary haemangioma was confirmed by histopathology which showed lobules of variably dilated network of blood filled capillaries in the papillary dermis surrounded by typical epithelial collarette. The lesions were removed by Radiofrequency (RF) in subsequent sittings. The present case report features an interesting and unusual morphological presentation of generalised eruptive lobular capillary haemangioma, which occurred de novo in a young female without any associated skin or systemic disease.

Keywords: De novo, Morphological presentation, Proliferative lesion, Pyogenic granuloma, Radiofrequency

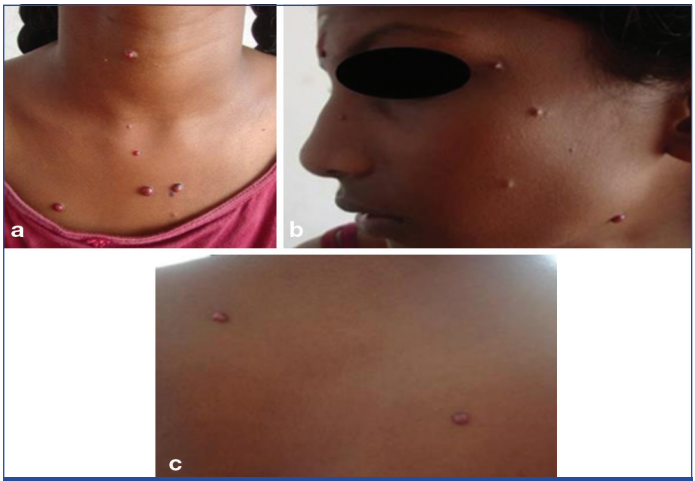
CASE REPORT

A 14-year-old, unmarried, school going female presented with multiple asymptomatic raised lesions over the chest, face and back for duration of four months. The lesions started on the back initially and later multiple lesions developed over face and anterior trunk over a period of four months. There was history of intermittent bleeding from the lesions after trivial trauma. There was no history of constitutional symptoms. There was no history of preceding trauma. The patient had not undergone any therapeutic procedure before the development of lesions. There was no history of systemic drug intake. The patient did not have any co-morbidities. There was no similar history in any of the family members.

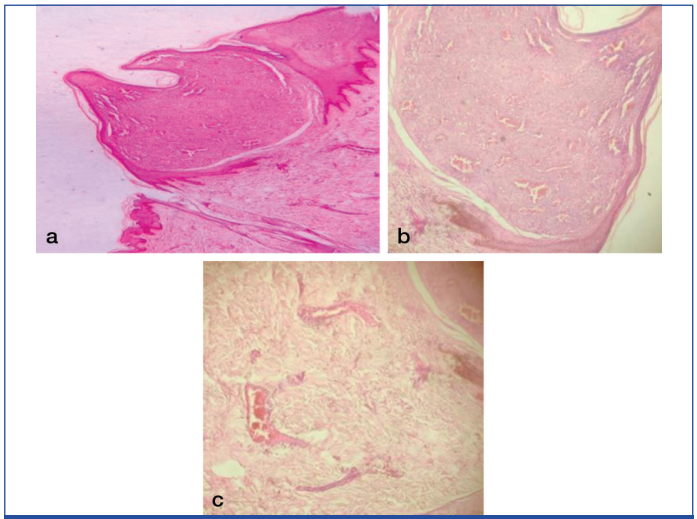
General examination and systemic examination were normal. On dermatological examination, there were 24 pink coloured to skin coloured, papules and nodules ranging from 0.5×0.2 cm to 2×1 cm, present diffusely over the face, neck, upper chest and upper back [Table/Fig-1].

Surface of the lesions was smooth. Since there was history of bleeding, palpation of the lesions was not done. Routine blood investigations, chest radiograph and serology for Human Immunodeficiency Virus (HIV), syphilis, hepatitis B and C were negative. Although clinical diagnosis was multiple, eruptive Pyogenic Granuloma (PG), differential diagnosis of bacillary angiomatosis and Kaposi's sarcoma was also considered. Excision biopsy of one of the lesions over upper back was done. Histopathology revealed normal intact epidermis and lobules of variably dilated network of blood filled capillaries in the papillary dermis, surrounded by myxoid stroma and inflammatory infiltrate consisting of lymphocytes and plasma cells. Typical epithelial collarette was present. There was no cellular atypia or extravasation of blood [Table/Fig-2]. Warthin-Starry and Giemsa stains were negative for bacillary angiomatosis.

The lesions were treated with Radiofrequency (RF) ablation under 2% plain lignocaine injections. The radiofrequency produces thermal effect on the target by resistive heating. The procedure was performed using a Monopolar and Monoterminal Radiofrequency unit (Derma India surge gold machine), with a partially rectified and markedly damp waveform (electrodessication). The procedure was



[Table/Fig-1]: Lobular capillary haemangioma: Multiple pink to skin-coloured papules and nodules over a): face; b): neck and; c): upper chest.



[Table/Fig-2]: Histopathology showing lobules of variably dilated network of blood-filled capillaries in the papillary dermis, surrounded by myxoid stroma and inflammatory infiltrate consisting of lymphocytes and plasma cells. Typical epithelial collarette can be seen. Haematoxylin and Eosin (H&E) stain; a) Scanner view, 4x magnification; b and c) 10x and 40x magnification, respectively}.

performed using angled needle electrode and power of 2 to 3. Electrodessication was performed from superior to base of individual lesions until the lesion was completely ablated. Topical 2% mupirocin was applied postprocedure. Two to three lesions were removed in one sitting. All the lesions were removed within total duration of 12 weeks. Intraoperative and postprocedure were uneventful. The treated lesions healed with postinflammatory hyperpigmentation. The patient was lost to follow-up due to the COVID-19 pandemic.

DISCUSSION

Lobular capillary haemangioma or PG is a common, benign and acquired vascular tumour that arises on skin and mucous membrane [1]. Eruptive PGs are classified as localised when they occur in a specific region and as disseminated or generalised when they occur diffusely. Generalised and eruptive lobular capillary haemangiomas are characterised by sudden eruption of widespread lesions in a short duration of time [2]. Such lesions have been reported in the setting of antecedent burns following lightning injury, systemic retinoid therapy, following therapeutic procedures, over underlying capillary malformation, following drug hypersensitivity and also rarely spontaneously [2-6]. Satellite lesions of PG erupting around a central lesion are also associated with trauma, excision or local treatment of the primary lesion [6]. Warner and Wilson-Jones syndrome refers to occurrence of multiple satellite nodules accompanying recurrent PG lesion after treatment [7].

Multiple eruptive PGs have been speculated as a vascular proliferative response to many angiogenic stimuli including trauma, infections, viral oncogenes, medications, vascular malformations and increased levels of female sex hormones [8]. The pathogenic mechanisms suggested were production of angiogenic factors by these stimuli, which in turn stimulates endothelial proliferation [9].

In the literature, multiple and eruptive PGs were treated with Pulsed-Dye Laser [10]. Diode excision, oral erythromycin and cryotherapy were also used for treatment [6,11-13]. In PubMed search using keywords: eruptive and PG (without any specific time period) de novo eruptive PG has been reported, as early as, in 1970 [10]. Only four such cases [Table/Fig-3] have been reported so far in India [6,14].

The present case is fifth in such series and the first one with generalised presentation, involving face, neck, upper chest and upper trunk compared to the relatively localised presentation in other four cases. De novo eruptive and generalised PG is a unique morphological variant which may look bizarre and alarming at presentation, but is completely amenable to treatment, cure and prevention of complications.

CONCLUSION(S)

The present case report is first de novo case of multiple, eruptive and generalised lobular capillary haemangiomas from India. The present report highlights the rare morphological presentation of eruptive PG occurring de novo. PG with generalised morphological pattern is a very rare manifestation in an adolescent female which should be diagnosed and treated early, as it can predispose to bleeding diathesis (due to injury), infection, scar formation and psychological distress in teenage.

Author and year of publication	Presentation	Treatment and outcome
Sethuraman G et al., 2006 [6]	A 22-year-old woman developed 15 to 20 PG lesions on scalp over course of 4 years	Treated with surgical shave excision followed by electrocautery of the base. No recurrence at nine months follow-up.
	A 55-year-old man developed multiple PG lesions over cheek in 2 months.	Referred for treatment with pulsed-dye laser. Follow-up details not documented.
Supekar BB et al., 2021 [14]	A 66-year-old female had multiple PG lesions over forearm in duration of 6 months.	Treated with cryosurgery using liquid nitrogen. Significant resolution of nodules after three sessions of cryotherapy.
	A 27-year-old male with 3 months history of multiple PG lesions on forehead.	Referred for surgical excision. Follow-up details not documented.
Current case (2022)	A 14-year-old female with 24 PG lesions over face, neck, upper chest and upper back which developed over period of 4 months.	Treated with 12 sittings of RF over three months duration, each sitting one week apart due to multiple lesions.

[Table/Fig-3]: Reported cases of de novo eruptive Pyogenic Granuloma (PG) in Indian literature- presentation, treatment and outcome [6,14].

REFERENCES

[1] Wollina U, Langner D, França K, Gianfaldoni S, Lotti T, Tchernev G. Pyogenic granuloma- A common benign vascular tumour with variable clinical presentation: New findings and treatment options. Open Access Maced J Med Sci. 2017;5(4):423-26.
[2] Palmero ML, Pope E. Eruptive pyogenic granulomas developing after drug hypersensitivity reaction. J Am Acad Dermatol. 2009;60(5):855-57.
[3] Xu Y, Li H, Wang ZX, Yang S. Multiple eruptive pyogenic granulomas occurring in a region of scalded skin. Pediatr Dermatol. 2016;33(1):e27-28.
[4] Williamson DM, Greenwood R. Multiple pyogenic granulomata occurring during etretinate therapy. Br J Dermatol. 1983;109(5):615-17.
[5] Swerlick RA, Cooper PH. Pyogenic granuloma (lobular capillary hemangioma) within port-wine stains. J Am Acad Dermatol. 1983;8(5):627-30.
[6] Sethuraman G, Khaitan BK, Sirka CS, Prasad HRY, Agarwal S, Singh MK, et al. Pyogenic granuloma with multiple and satellite lesions. Indian J Dermatol. 2006;51(2):134.
[7] Ballin JS, Egan KM, Satter EK. What is your diagnosis? Recurrent lobular capillary hemangioma with satellitosis (Warner and Wilson-Jones syndrome). Cutis. 2014;93(3):125, 132-33.
[8] Whitaker S, Bouquot J, Alimario A, Whitaker T. Identification and semiquantification of estrogen and progesterone receptors in pyogenic granulomas of pregnancy. Oral Surg Oral Med Oral Pathol. 1995;78:755-60.
[9] Vara JT, Gurudu VS, Ananthaneni A, Bagalad BS, Kuberappa PH, Ponnapalli HP. Correlation of vascular and inflammatory index in oral pyogenic granuloma and periapical granuloma– an insight into pathogenesis. J Clin Diagn Res. 2017;11(5):ZC25-28.
[10] Shah M, Kingston TP, Cotterill JA. Eruptive pyogenic granulomas: A successfully treated patient and review of the literature. Br J Dermatol. 2006;133(5):795-96.
[11] Netchiporouk E, Moreau L, Ramirez LP, Castillo PAC, Bravo FP, Solar MCD, et al. Eruptive disseminated pyogenic granulomas following lightning injury. Dermatology. 2015;230(3):199-203.
[12] Ceyhan AM, Basak PY, Akkaya VB, Yildirim M, Kapucuoglu N. A case of multiple, eruptive pyogenic granuloma developed on a region of the burned skin: Can erythromycin be a treatment option? J Burn Care Res. 2007;28(5):754-57.
[13] Qadir S, Manzur A, Raza N. Multiple disseminated pyogenic granulomas. J Coll Physicians Surg Pak. 2013;23:588-89.
[14] Supekar BB, Wankhade VH, Chopkar AD, Singh RP, Bhat D. Multiple erythematous nodules: An intriguing entity. Indian Dermatol Online J. 2021;12(1):142.

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