SCIENCE WORLD JOURNAL OF CANCER SCIENCE AND THERAPY

Unusual Presentation of Pyogenic Granuloma in a Finger of a Pregnant Woman: A Rare Case Report

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Article Information

Article Type: Case Report
Journal Type: Open Access
Volume: 1 Issue: 2
Manuscript ID: SWJCST-1-107
Publisher: Science World Publishing

Recieved Date: 25 January 2021
Accepted Date: 20 February 2021
Published Date: 24 February 2021

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Citation:

Hope JMV (2021). Unusual Presentation of Pyogenic Granuloma in a Finger of a Pregnant Woman: A Rare Case Report. Sci World J Cancer Sci Ther, 1(2);1-5

ABSTRACT

Pyogenic granuloma is a benign vascular lesion usually occurring in the skin or mucous membrane. In the modern English literature, reported cases of pyogenic granuloma of fingers arise from the volar, dorsal or lateral surfaces with a possible link to traumatic events. Therefore, we report the first case of pyogenic granuloma occurring during the course of pregnancy with rare location at the medial surface of the mid-phalanx of the left index finger.

KEYWORDS: Pyogenic granuloma; Medial surface; Pregnancy; Finger

INTRODUCTION

Pyogenic granuloma (PG), also known as lobular capillary hemangioma, is a benign vascular tumor that occurs on the skin and mucous membranes, occasional it can be found subcutaneously or intravascularly [1-3]. It was first described by Poncet in 1897 as botryomycosis hominis [4]. PG can arise spontaneously, in sites of injury, or within capillary malformations [1-3]. Etiologies of pathogenetic importance are trauma, BRAF mutations and probably herpes virus type 1, Orf virus and/or human papilloma virus type 2 [4-7]. It occurs in all age group and gender [3-5]. Pyogenic granuloma lesions commonly occur in gingiva, lips, tongue and upper limbs and has no potential for malignancy. The reported digital location is either lateral, volar or dorsal surfaces [1, 2, 3-5]. Numerous options of treatment are available [1, 8, 9]. Here, we report the first case of pyogenic granuloma occurring during the course of pregnancy with rare location at the medial surface of the mid-phalanx of the left index finger.

CASE REPORT

A 37-year-old woman presented, one month after delivery, with a painless swelling of the proximal medial surface expanding to the dorsum of the middle phalanx of the left index finger, progressively increasing in size for the past three months. Initially it was a bean-sized and it tripled its size one month later after delivery. Examination of the left index finger revealed a red, firm, well-circumscribed, fungating 45mmx30mmx15mm mass over the proximal medial surface of the middle phalanx expanding to the dorsum (Figure 1). There was neither tenderness nor local heat around the tumor. Systemic review was unremarkable and the newborn (boy) was well healthy. Blood examination revealed WBC 5200/µl, Hb 14.5 g/dl, C-Reactive Protein 0.13 mg/dl, rheumatoid Factors not detected and no remarkable inflammatory reaction. Conventional left hand radiographs detected a radiolucent soft tissue mass on the medial side of mid-phalanx of the index without bony involvement (Figure 2).

Figure 1: Exophytic and hemorrhagic mass over the medial surface of the middle phalanx of the left index finger with dorsal expansions measuring 4.5cmx3cmx1.5cm

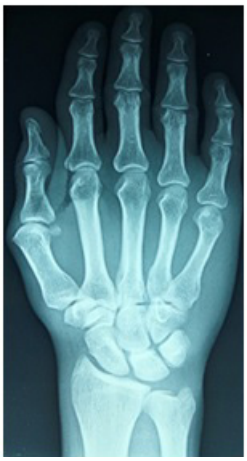

Figure 2: Anteroposterior radiography of the left hand showing a radiolucent soft tissue mass on the medial side of mid-phalanx of the index. No bony lesions were detected.

The consent was made with the patient to undergo wide margin surgical resection of the tumor under local anesthesia with subsequent pathological examination. It was excised with 3 mm margin of normal skin and subcutaneous tissue. There was no adhesion to bone or tendon and the surgical wound was closed primarily. Macroscopically, it was a large, firm, lobulated, reddish, exophytic, mushroom-like tumor with tendency to bleed profusely and measuring 45mmx30mmx15mm. Microscopically, pathological examination with hematoxylin and eosin staining showed monotonous lobular endothelial hyperplasia of capillary vessels proliferation and edema of stroma, and invasion of inflammatory cells (Figure 3 & 4). In immunolocalization of vascular endothelial growth factor (VEGF), VEGF was detected at capillary vessels inside the tumor. Any findings of malignant change were not seen; thus it was diagnosed as pyogenic granuloma. The wound healed uneventfully. The patient was followed up annually and there is no recurrence of the tumor after three years of the excision (Figure 5).

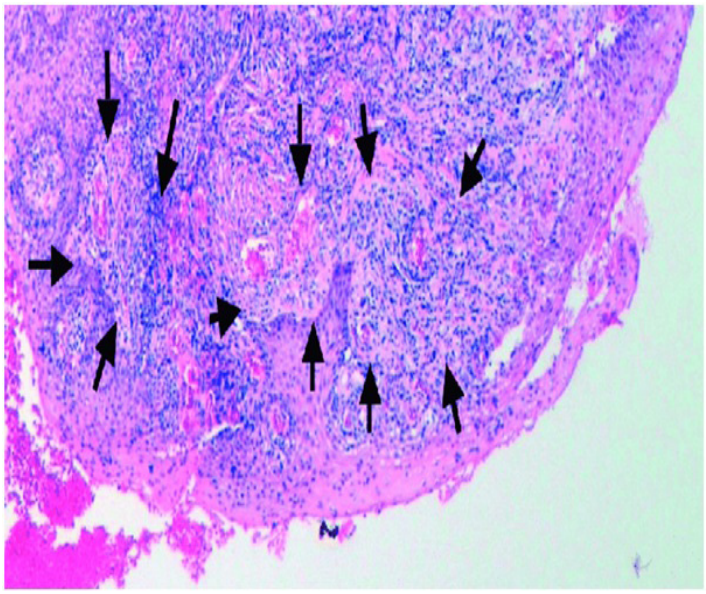

Figure 3: The histological type of the pyogenic granuloma is non-lobular capillary hemangioma showing vascularized tissue with capillaries proliferation, inflammatory infiltrate and no malignancy. Arrow heads label blood vessels surrounded by connective tissue.

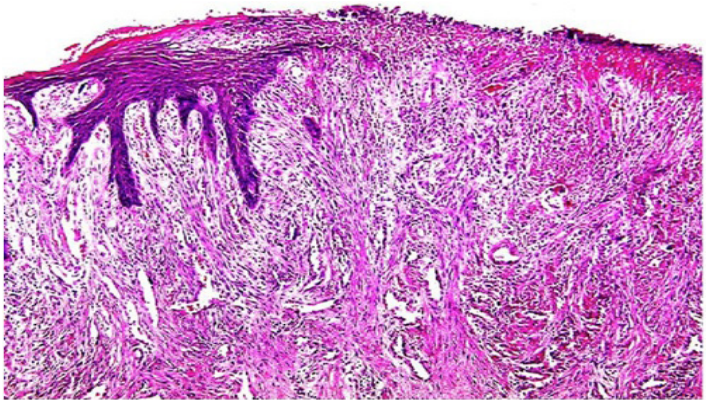

Figure 4: Higher magnification shows monotonous endothelial hyperplasia and capillary proliferation (**H&E,** × 100)

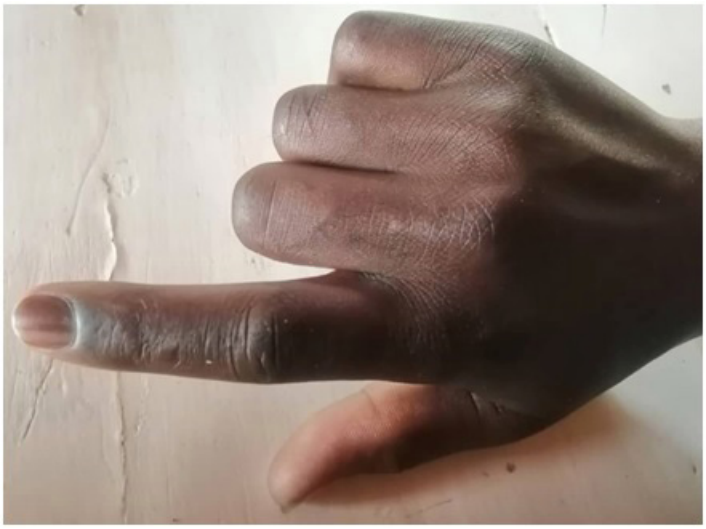

Figure 5: Well healed surgical wound cosmetically acceptable 3 years post excision

DISCUSSION

The etiology of pyogenic granuloma (PG) is unknown. However, PG is frequent in patient who is in certain condition, such as in pregnancy, suffering from repeated minor trauma (proliferative response of capillaries as a result of repetitive injury or chronic irritation, peripheral nerve injury and cast immobilization), immunocompromised host, taking retinoids or antiretroviral drugs, anti-TNF alpha drug (etanercept) and inflammatory and infectious agents are also hypothesized as possible factors [10,11]. Besides, hormonal factors may play a role [5]. In particular, mechanical trauma (e.g., manicure, pedicure), friction (e.g., a prolonged walk), or an inflammatory cause (penetration of a foreign body) can predispose the development of a PG. In childhood, PGs are often self-caused because of nail biting or improper nail trimming, with penetration of spicules in the soft tissues of the nail folds and subsequent foreign body reaction, especially if the lesion is associated with digital swelling [1, 2, 5-11]. PG has been associated with certain medications such as oral contraceptives, retinoids, gefitinib and afatinib [2,12,13]. Curr et al., reported a case of multiple pyogenic granuloma in a rectal cancer patient who takes 5-fuluorouracil and speculated 5-fuluorouracil that were the cause of pyogenic granuloma [14]. Piguet et al., reported a case of pyogenic granuloma-like lesions during capecitabine therapy [15]. Most tumors occur as solitary lesions, but multiple grouped or disseminated tumors have been described. Multiple disseminated tumors are an adverse cutaneous effect of melanoma treatment with selective BRAF inhibitors like vemurafenib or encorafenib [16]. Other PG occur with targeted oncological therapies using epidermal growth-factor receptor inhibitors or mitogen-activated protein kinase (MEK) inhibitors and rituximab [17-18]. In our case, it was a solitary tumor and pregnancy is believed to be the predisposing factor to the occurrence of this pyogenic granuloma.

The endothelial cells in PG express CD34, ICAM-1, VCAM-1 associated with an increased microvascular density. Recently, BRAF c.1799T>A mutation had been identified in endothelial cells as a major driver mutation in the pathogenesis of PG. This explains the occurrence of multiple PG in patients treated with BRAF inhibitors [19-20]. The participation of viral particles in PG pathogenesis has been discussed. Alpha-herpes viridiae type 1 is considered as a possible indirect factor stimulating angiogenesis in PG. In some patients, dermatotropic parapoxvirus (Orf) could be identified by polymerase-chain reaction (PCR). Human papilloma virus DNA could be identified in 44% of these lesions with HPV type 2 as the most common [6-7]. Portwine stains are at risk for secondary PG. Their treatment with vascular lasers may induce PG as well [21]. Satellitosis has been observed in paediatric PG [9-22]. Histologically, PG is composed of capillaries and venules with plump endothelial cells separated into lobules by fibromyxoid stroma. The development can be classified into cellular phase, capillary phase or vascular phase, and involutionary phase [19, 20]. Our case was in phase III (involutionary phase). Histological examination showed lobular hyperplasia of capillary vessels and the expression of vascular endothelial growth factor at capillary vessels inside tumor and no malignancy.

PG occurs in all age groups. There is no clear predominance of a gender. PG appear as small or large, smooth or lobulated, reddish, exophytic vascular nodules that can grow rapidly. Larger lesions become lobulated and sometimes develop into mushroom-like, pediculated tumors. PGs have a tendency to bleed profusely. Bleeding is the leading symptom [3]. The most common locations are the digits of both hands and feet, followed by lips, gingiva, tongue, arm, palm and thigh [1, 10-11]. Considering the fingers, most lesions occur on the nail folds, subungual, periungual, dorsal, volar and lateral sides of the finger [3, 4-11]. During pregnancy, large intraoral PG may develop [23]. Uncommon sites are vulva and penis, esophagus, gut, and tracheobronchial tree. Gastrointestinal PG can cause severe anemia. Extremely rare are intravascular tumors which bear the risk of thrombosis [24-27]. Our case is a 37-year-old woman with a large, reddish, exophytic and hemorrhagic tumor of the medial surface of the proximal mid-phalanx of the left finger. It has occurred during pregnancy.

There are various options of management of pyogenic granuloma reported: electrocautery, laser therapy, radiation, cauterization and surgical excision with variable outcomes described in terms of recurrences [1, 2, 5, 8, 11-28]. Poorer outcome was noted using shave excision and/or curettage and cautery as compared with surgical excision where the recurrence rate is 10% and 3.6% respectively [2-5]. A better outcome was noted when surgical excision is combined with electrocautery, with some cited 0% of recurrence [1, 2-5]. Smaller lesions may be treated using sclerotherapy with ethanolamine oleate or laser therapy as the cure rate for the former

is almost 100% [2-5]. Regardless of the definitive treatment modalities, the risk of recurrence is still unavoidable. Completeness of excision will reduce the risk of recurrence [2, 3, 5, 29-30]. In our case, wide margin surgical resection was performed and the wound healed with acceptable cosmesis. She was followed up annually and no recurrence at three year follow up time.

CONCLUSION

After extensive literature review, there is no question that pyogenic granuloma is a rare medical occurrence. This case represents a rare lesion of the hand happened during pregnancy. With this notion, it has been proven by this case in particular due to the many risk factors that pregnancy, repetitive injury or chronic irritation, hormonal disturbances, with an unexplained mass, concern should be elicited for the diagnosis of pyogenic granuloma. The case presented here displays the unusual location, at the medial surface of the middle phalanx of the left index finger, of a small mass progressively increasing in size recognized during pregnancy and pathologically diagnosed PG over a 3-month course which healed well with surgical excision. Surgical excision remains the best treatment modality for pyogenic granuloma as it has the lowest recurrence rate, cosmetically acceptable and generally the best option for large lesions. Regular follow up is therefore needed to monitor recurrence.

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