Case Report

Zosteriform cutaneous leiomyoma relieved with topical nitroglycerine

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Abstract

Cutaneous leiomyomas (CL) are one of the painful benign tumors of the skin arising out of smooth muscle. CLs are reddish brown to skin colored firm nodules and may be single or multiple depending on site of occurrence. Zosteriform distribution is rare to occur. The cause of pain has been argued upon; however the pain may be severe and exacerbated on cold weather and pressure. The treatment, when surgical is met often with recurrence. Various pain- alleviating measures have been used; we report herein a case of zosteriform CL not only for its rarity, but also to share our experience with a compounded nitroglycerine cream to much success.

Keywords: cutaneous leiomyoma, painful tumor, nitroglycerine.

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Received Date: 24/03/2015 Revised Date: 02/04/2015 Accepted Date: 05/04/2015

Access this article online

Quick Response Code:

Website: www.statperson.com

DOI: ---

INTRODUCTION

Cutaneous leiomyomas (CL) are one of the painful tumors of the skin. These are benign tumors of smooth muscle and may arise from the arrectores pilorum muscle, muscular lining of the cutaneous arterioles or the dartos muscle of the scrotum. Accordingly, they are known as piloleiomyoma, angioleiomyoma and genital leiomyoma, respectively.[1] Clinically, CLs present as reddish-brown to skin-colored, firm nodules, mostly over the face, neck, trunk, or extremities.[2-3] While lesions may be solitary, lesions especially on the trunk can be multiple (80%[4] of patients). Multiple CLs may be disseminated or zosteriform. Zosteriform (segmental or dermatomal) distribution of lesions is a rare occurrence.[5-7] CLs often look similar to other benign cutaneous neoplasms and then distinction can only be made by histology. On histopathology intertwining bundles of smooth muscle, collagen fibers are seen with variable lymphocytic and mast cell infiltrate occupying most of deeper dermis.[1,4] Patients usually are hypersensitive to light touch and cold temperature and report moderate to incapacitating pain[8-11] with trauma and pressure. The character of the pain may be burning, pinching, or stabbing. Segmental and multiple disseminated lesions are likely to cause greater pain.[1,2,6] The cause of the pain is not decided upon.[2,10] Some authors have noted increased numbers of nerve fibers in these tumors and state that compression of these to be the cause of pain.[9,12] Other authors however have not been in agreement and have described muscle spasm as the cause. Successful treatment using different oral and topical agents has been reported.[8,12] Oral agents like alpha blockers, calcium channel blockers, nitroglycerin, antidepressants, ethaverine, and analgesics have been used successfully.[4,5,7,9,14] Topical treatments have included nitroglycerine paste, lignocaine, phentolamine, hyoscine hydrobromide[3] and liquid nitrogen cryotherapy in one case.[10]

CASE REPORT

A 39 year old man presented to the clinic last winter (2013) complaining of recurring intense pain over some

How to site this article: Loknath Ghoshal, Saumen Nandi, Soumya Kundu, Souvik Ray, Falguni Mandi. Zosteriform cutaneous leiomyoma relieved with topical nitroglycerine. *International Journal of Recent Trends in Science and Technology* April 2015; 14(3): 531-533 http://www.statperson.com (accessed 08 April 2015).

tumors which he had on his right upper trunk for the preceding 12 years. The lumps initially had started as small mildly painful elevations which attained the present size with time. The pain, which had significantly increased in intensity and frequency over the last 5 years, generally increased with exposure to cold and mental stress. Examination revealed multiple painful, firm reddish nodules of different sizes over the right upper trunk in the sub-clavicular region along the distribution of the first thoracic (D_1) dermatome. There was clear sparing of the left side of the trunk (**Figure 1, 2**). Rest of the body and systemic examination was normal. As far as his knowledge would go, there was no history of similar problem in his family. One of the nodules was subjected to excision biopsy. Histopathology revealed a tumourous mass composed of smooth muscle fibers with irregular arrangement and interlacing with collagen fibers invading full thickness of the dermis (**Figure 3**). The fibers had elongated, cigar shaped nuclei with rounded ends, suggestive of smooth muscle (**Figure 4).** The patient was counseled about the possible modalities of treatment; he refused surgical modalities and opted for measures aimed solely at pain relief. We started treatment with diclofenac sodium 50 mg tablets to be taken twice to thrice daily coupled with a compounded cream containing 2% nitroglycerine. The patient was instructed to apply the cream twice a day, once in the morning and once again 6-8 hours later. The patient was told to replace the cap tightly, wash his hands after such application and warned about symptoms of postural hypotension. At the follow-up visit 10 days later, the patient did not have pain, though he stated having two bouts of dizziness on the first day of application of the cream. We advised continuation of the nitroglycerine cream only on as required basis. The patient has been learnt to use the nitroglycerine cream and is symptom-free since then.

Figure 1(a): Unilateral segmental distribution of the tumors; 1 (b) Close up of fig 1(a)

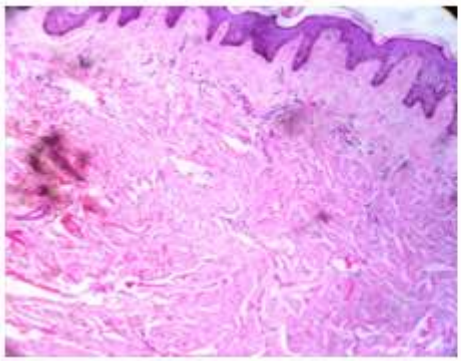

Figure 2: Histopathology of a tumor (magnification X100, H and E) showing a tumourous mass composed of smooth muscle fibers almost filling up the deeper dermis

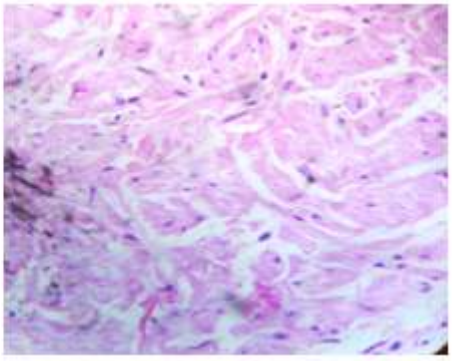

Figure 3: Histopathology of tumor (magnification X 400, HandE) elongated, cigar shaped nuclei with rounded ends, suggestive of smooth muscle

DISCUSSION

The main differential diagnosis was congenital smooth muscle hamartoma, the possibility of which was excluded by the history and the relative lack of collagen between the smooth muscle fibers which is more characteristic of the latter. The standard of treatment for cutaneous leiomyomas is surgical excision, but the high recurrence rate of 50%[13] makes it less practical. The present patient having refused surgical excision was offered only pain management. We could find only one case report[5] in the

English literature employing similar treatment (nitroglycerine paste) for this condition. The mechanism of action could be the smooth muscle relaxing property of the latter.

CONCLUSION

The purpose of report of this case was not only its rarity, but also to highlight upon the usage of nitroglycerine cream as a potential low cost (almost no cost) management of this condition.

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Source of Support: None Declared
Conflict of Interest: None Declared