Case Report

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Cutaneous leiomyoma of scalp: a rare case report with review of literature

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ABSTRACT

Skin is the largest organ of the body and many types of tumor arises from different part of skin layer. A leiomyoma is an uncommon, benign tumour of smooth muscle of skin derived from the arrector pili muscle of skin. These lesions can develop wherever smooth muscle is present, but malignant transformation probably does not occur. Cutaneous leiomyomas or piloleiomyomas appear as small (0.5-2 cm) firm skin coloured nodules. They arise from the arrector pili muscles which are responsible for making your hairs stand on end. Multiple lesions may develop, often in clusters and commonly in a segmental distribution. These nodules are benign, so are only of concern because of their appearance or their tendency to be tender if knocked. Female patients with cutaneous leiomyomas may also develop uterine leiomyomas (fibroids), and very rarely leiomyomas can be associated with renal cell carcinoma (hereditary leiomyomatosis and renal cell cancer). We report a case cutaneous leiomyoma of scalp in a 22 year old male who present with the complaints of scalp swelling.

Keywords: Cutaneous leiomyoma, Scalp, Skin

INTRODUCTION

Leiomyomas are benign soft tissue neoplasms that arise from smooth muscle. They can develop wherever smooth muscle is present. Skin is the second commonest location for leiomyoma after uterus which accounts for 95% of cases. Cutaneous leiomyomas account for 75% of extrauterine leiomyomas.¹

Superficial or cutaneous, leiomyomas are of two types. Those arising from the pilar arrector muscles of the skin may be solitary or multifocal and are often associated with considerable pain and tenderness. The other form, the genital leiomyoma, arises from the diffuse network of muscle in the deep dermis of the genital zones (e.g., scrotum, nipple, areola, vulva). In the scrotum they arise from the dartoic muscles (dartoic leiomyoma) and in the nipple from the muscularis mamillae and areolae. This form is nearly always solitary and rarely causes significant pain.²

CASE REPORT

22 year old male was present to surgical OPD with the complaints of scalp swelling since 6 months. He noticed a swelling on the scalp before the 6 months which was gradually increasing in size. Swelling was painless on starting but now it has become painful since 1 month. On examination a single well circumscribed lesion was present on right side of scalp which was measuring 5x3x2 cm in size and red brown in colour and soft in consistency. A provisional clinical diagnosis of sebaceous cyst was made and mass was removed under local anaesthesia on the OPD basis and patient was

discharged on the same day. Tissue was sent for histopathological examination to our department.

Gross examination

Single globular grey white soft tissue mass measuring 4.5x1.5x1 cm in size with skin attach on one side was received.

Microscopic examination

Multiple sections were taken and stained with hematoxyline and eosin stain.

Section showed thin layer of epidermis and dermis. Dermis showed hair follicles, sebaceous gland and sweat glands. Tumor was well circumscribed lobulated and encapsulated (Figure 1). Tumor cells were arranged in whorl like pattern (Figure 2). The tumor cells composed of interlacing bundles of smooth muscle fibres. Tumor cells were uniform fusiform or spindle-shaped cells (Figure 3). These cells had indistinct cell borders and abundant fibrillary eosinophilic cytoplasm. Nuclei were elongated with tapered ends, and have finely dispersed chromatin and small nucleoli with minimal pleomorphism (Figure 3). Mitotic figure was not seen. At the periphery of tumor myxomatous changes were present just below the capsule which appears as bluish protenaceous material in between the cells. In between the tumor cells, few scattered lymphocytes were present. Necrosis was absent.

Special staining was done with Masson's trichrome stain which shows which stain smooth muscle cells as red colour and nuclei as black. At places Masson's trichrome stain, tumor cells as red linear streaks traversing the cytoplasm in a longitudinal fashion (Figure 4).

On the basis of gross, histopathological and special staining finding a diagnosis of cutaneous leiomyoma (spindle cell tumor) was made.

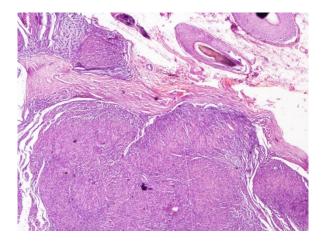


Figure 1: Section shows a well encapsulated tumor with hair follicles outside the tumor capsule with myxomatous changes at periphery (H&E, 4x).

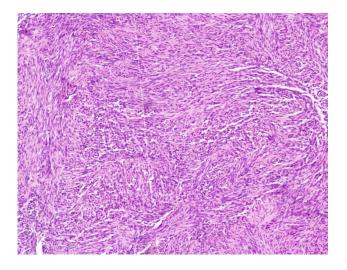


Figure 2: Section shows interlacing smooth muscle cells arranged in whorled pattern (H&E, 10x).

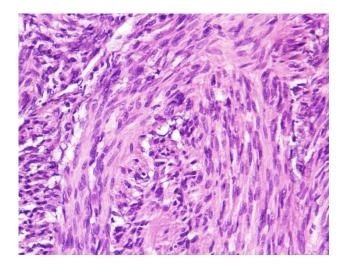


Figure 3: Section shows spindle cells with tapering ends, vesicular chromatin and abundant eosinophilic cytoplasm (H&E, 40x).

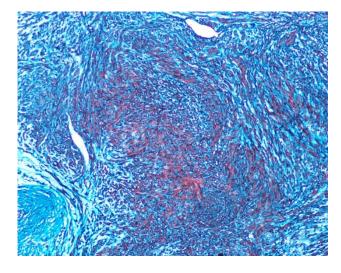


Figure 4: Masson's tichrome stain shows red colour smooth muscles fibres (MT, 10x).

DISCUSSION

Skin is the largest organ of the body and directly in contact with environment. In the skin lining arrector pilli muscles attached to the hair follicles and present in the dermis. In the skin smooth muscle tumor as leiomyoma and leiomyosarcomas are uncommon tumors. The anatomical distribution of cutaneous leiomyomas is extensive. Leiomyomas present clinically either solitary or multiple lesions. Lesions appear as skin colored or red surface, and are most commonly located in the extremities.³ In our case patient present with solitary leiomyoma present on scalp which is an uncommon location for solitary leiomyoma.

Lesions of leiomyoma clinically may present as plaques, zosteriform groups, or, in symmetrical distribution, nevus. Usually two or more areas are affected and multiple lesions were present. Lesions of cutaneous leiomyomas are usually tender but not always. Sometime may give painful paroxysms. Pain can be spontaneous or triggered by exposure to cold, pressure, trauma, or emotion.

Adults are commonly affected by leiomyoma. In children isolated lesion of cutaneous leiomyoma are also reported, including a solitary cutaneous leiomyoma on the heel of a neonate at birth.⁴

The five types of leiomyomas of the skin are described as following (1) multiple piloleiomyomas and (2) solitary piloleiomyomas, both arising from arrectores pilorum muscles (3) solitary genital leiomyomas, arising from the dartoic, vulvar, or mammillary muscles; (4) solitary angioleiomyomas, arising from the muscles of veins; and (5) leiomyomas with additional mesenchymal elements.

Multiple piloleiomyoma, most common type of cutaneous leiomyoma, are small, firm, red or brown intradermal nodules arranged in a group or linear pattern, primarily affecting the trunk and extremities but may occur on the face and neck and rarely in the mouth.⁵

We extensively search for large series studies on leiomyoma cutis on internet but we found only some studies and not found any study in which more than 40 case were reported. Here we describe some studies on cutaneous leiomyoma.

Malhotra P et al.⁶ conducted a study on 37 cases of leiomyoma cutis and they found that piloleiomyoma was most common variant, followed by breast leiomyomas, angioleiomyoma and least common was scrotal leiomyomas. Patients with an average age of 38.2 years at presentation with male predominance (M:F-2.2:1). Cutaneous leiomyoma were more commonly present as single lesion than multiple lesions. The trunk and upper limbs were involved most commonly, followed by lower limb, face, breast, and scrotum.

Ghanadan A et al.⁷ conducted a study and they found that cutaneous leiomyoma were more common in were male than female with the male to female ratio 4:1 and mean age of presentation or diagnosis was 44.6 ± 13.6 in male and 50.8 ± 14.6 in females. The lesion was mostly present on extremities (52%). Lesion of cutaneous leiomyoma was present with average size of 5.5 mm. They examine all the tumors pathologically and found interlacing short fascicles of bland spindle cells with elongated nuclei and blunt ends. Two subtypes of cutaneous leiomyoma were diagnosed, Pilar leiomyoma and angioleiomyoma. Pilar leiomyoma were present mostly in upper dermis extending to deep dermis and all angioleiomyoma were placed in deep dermis. Angioleiomyomas revealed myxoid and hyaline changes in 75% of the tumor. 75% of Pilar leiomyoma present with acanthosis and elongated rete ridges with hyperpigmentation of skin were seen but not in angioleiomyoma. Smooth muscle bundles which are interdigitated with elongated rete ridges were also a histological feature purely seen in Pilar leiomyoma but not in angioleiomyoma.

Heatley MK, et al.⁸ 1989 conducted a study on 28 cases of solitary cutaneous leiomyomata were encountered in a 10 year period and they found female preponderance with Male to Femle ratio 9:19 with the mean age of the presentation is 53 years.

The lesions present most commonly in there study on the lower limbs followed by head and neck, and the genital skin and mostly were pain less. The size of lesion ranged from 0-5 cm in diameter.

Mostly cutaneous leiomyoma was misdiagnosed by clinician as fibromas, sebaceous cysts and fibroepithelial polyps.

Histologically tumor composed of interweaving bundles of smooth muscle cells which stained red with Masson's trichrome.

Histopathology: Piloleiomyomas and genital leiomyomas are similar in histolopathologic appearance.⁹

Most Pilar leiomyomas are 1-2 cm in size and tumor present in the dermal connective tissue and are separated from the overlying atrophic epidermis by a clear grenz zone.

Bhaskar S et al.¹⁰ reported a case of solitary cutaneous leiomyoma over the anterior abdominal wall in a 55 year old male patient presented with long history (4 year) solitary swelling which was painless and adjacent tender area probably showing the dormant nature of the disease.

Prasad P et al.¹¹ report 3 cases of leiomyoma cutis in female with painful and tender papules and plaques on the breast is presented with the involvement of the skin over the breast and all cases was histopathologically diagnosed. Prasad P et al. found that the leiomyoma cutis

rarely found at unusual location like breast. All the patients were treated with oral nifedipine 10 mg bid and in two patients noticed considerable decrease of pain and tenderness.

Dilek N et al.¹² report a case of Pilar leiomyoma on the face and neck in the form of multiple, large papulonodules plaque so the treatment by excision and grafting would be difficult because of the large surface area involved. The patient was treated with nifedipine.

They found that Skin is the second most common location for leiomyoma and often localized on the trunk or extremities. Less than 1% of leiomyomas present as papulonodules in the head and neck.

Sankar G et al.¹³ report a case of leiomyoma. On histopathological examination they found leiomyoma, with mucoid degeneration which was an unusual association.

Smith CG et al.¹⁴ report case of piloleiomyomas in a 45 year old female with multiple painful piloleiomyomas in a zosteriform pattern on her left flank and they report this case as a first case of zosteriform leiomyoma cutis.

In H&E staining muscle bundles of various leiomyoma stain pink as collagen fibre. The nuclei of the fibroblasts in the collagen fibre are shorter than the nuclei of the smooth muscle fibres and show tapering at their ends, while smooth muscle nuclei have blunt ends.

Smooth muscle cells show sometime vacuolization, especially in cross-sections, due to perinuclear clear zone.

A more definitive method for identifying collagen bundles is possible with special stain, such as the aniline, von Gieson and trichrome stain.

With the aniline blue stain, muscle stains red and collagen blue; with the trichrome stain, muscle stains dark red and collagen green or blue. Arrectores pilorum gives rise to smooth muscle tumors like leiomyoma. Electron microscopic examination has shown that piloleiomyomas are composed of normal-appearing smooth muscle cells. Each of these cells has a central nucleus surrounded by an area containing endoplasmic reticulum and mitochondria and, peripheral to this area, numerous myofilaments arranged in bundles. Both cytoplasmic and marginal dense bodies are present.

The cause of the frequent pain or tenderness was revealed by electron microscopy, as ultrastructural damage to nerve fibres through distortion and disruption of the myelin sheath, and only scanty unmyelinated nerve fibers are postulated factors.

Cutaneous leiomyomas do not undergo malignant transformation.

Total surgical excision is sufficient for cure. The lesions are often so numerous that total surgical excision is not possible. Laser therapy has been used with some success.

CONCLUSION

Clinical diagnosis of cutaneous leiomyoma was difficult in single lesion. Cutaneous leiomyomas are rare benign tumor. These form an important clinical differential diagnosis of painful papulonodules and must be biopsied to confirm the diagnosis. Complete excision of tumor is sufficient for treatment because recurrence is rare in this tumor.

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