Case Report

Multiple cutaneous and uterine leiomyomas: A case report

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ABSTRACT

Piloleiomyomas are uncommon benign papulo-nodular tumors arising from the erector pili muscle, often found with a segmental cutaneous distribution. While the presenting complaint for these patients may be intermittent pain in the swellings, they are associated with hereditary syndromes which, more importantly, require further surgical attention, namely Reed Syndrome (multiple cutaneous and uterine leiomyomatosis) and hereditary leiomyomatosis and renal cell carcinoma syndrome. We present a case of a 30-year-old woman who was referred to us for excision of multiple painful cutaneous lesions which were present over her left breast, who had a past history of primary infertility with multiple uterine fibroids requiring laparoscopic myomectomy 3 years back, with no significant family history. Few small lesions had been treated with cryotherapy before she was referred to us. The patient underwent excision under local anesthesia of the largest of the lesions causing the most pain and the histopathological examination determined it to be a piloleiomyoma. Abdominal ultrasonography revealed bilaterally normal kidneys with a small asymptomatic uterine fibroid. Post excision, the patient was pain-free and no surgical site recurrences were observed on follow-up after 1 year.

Key words: Hereditary leiomyomatosis, Leiomyoma, Piloleiomyoma, Reed syndrome, Renal cell carcinoma

Piloleiomyomas are rare, benign, cutaneous tumors arising from the erector pili muscle and the pilosebaceous unit, often in a segmental cutaneous distribution [1]. The presenting complaint is pain in the lesions, but they are associated with autosomal dominant syndromes causing uterine leiomyomas or renal cell carcinomas, which need a willful search and definitive management, which might eclipse the presenting complaint and existing treatment strategy of the patient [2]. Data on prevalence is limited due to its rare presentation [3] and hence, because of their association with more morbid visceral conditions, such lesions need to be investigated thoroughly.

We report a case of a 30-year-old woman with segmental piloleiomyoma with a syndromic association.

CASE REPORT

A 30-year-old woman came to our outpatient department with complaints of multiple painful skin lesions over her left breast for the past 3 years. The patient had a history of primary infertility with dysmenorrhea and was diagnosed with uterine fibroids 3 years back, for which she underwent a laparoscopic myomectomy.

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The lesions were insidious in onset, progressively increased in size and number, and were restricted to the upper quadrants of the left breast. The lesions were multiple, papulo-nodular, smooth, well-defined, soft to firm in consistency, light brown to skin-colored, not fixed to deeper structures, and painful to touch, without any surrounding edema or erythema, the largest of which was 2 × 1 cm and associated with the most discomfort to the patient (Fig. 1). There were no ulcers or any discharge from the lesions, nor was there any such history. Bilateral breast tissue and axillary examination were normal. There were no other areas of distribution of such lesions on the patient. The rest of the skin was normal on examination. The patient was vitally stable, of average height, and built with a body mass index of 24, with no history of diabetes, hypertension, or any other medical comorbidity.

After an initial dermatology consultation and treatment of smaller lesions with cryotherapy, the patient was referred for definitive surgical therapy for the largest lesion. Abdominal ultrasonography was done to screen for uterine or renal pathology, and bilateral kidneys were found to be normal, without any cysts or masses. She was taken up for excision of the largest cutaneous lesion under local anesthesia (Fig. 2), and primary closure was achieved with subcuticular monofilament absorbable sutures. The perioperative and postoperative course was uneventful.

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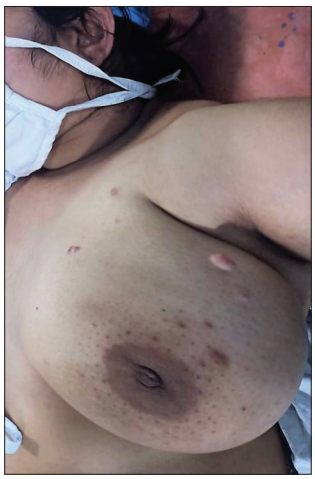

Figure 1: Clinical photograph showing segmental distribution of the tumors

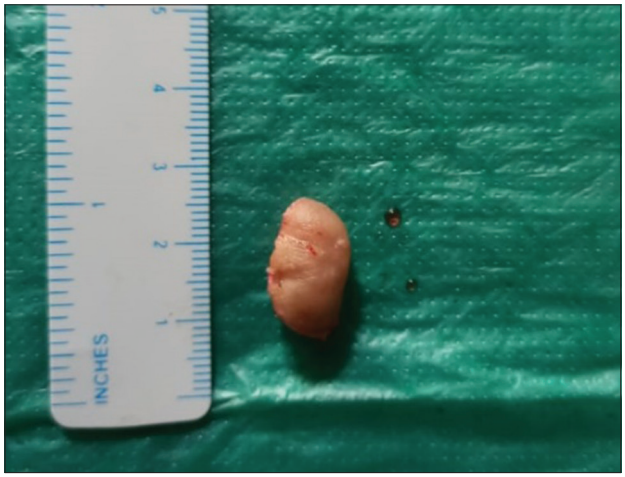

Figure 2: Excised specimen

The histopathology report described a well-circumscribed tumor mass composed of a fascicular arrangement of spindle cells with features suggestive of piloleiomyoma (Fig. 3). Combined with a positive history of uterine leiomyomas, a clinical diagnosis of Reed Syndrome was reached. Occasional pain in the remaining lesions was managed by over-the-counter analgesics and cryotherapy. On a follow-up of 1 year, the surgical site showed no recurrence and was not painful to touch.

DISCUSSION

Leiomyomas are rare, benign tumors arising from smooth muscle cells, with the most common site being uterine leiomyomas. Piloleiomyomas, a subset of cutaneous leiomyomas, consist of 75% of all extra-uterine leiomyomas and are often multiple, with segmental distribution. They arise from the erector pili muscle and pilosebaceous unit and are papulo-nodular in appearance, and are associated with pain to touch, cold temperatures, or emotional stimuli. Other types of cutaneous leiomyomas include angioleiomyomas and genital leiomyomas [1].

Piloleiomyomas do not show any gender or racial predisposition. They can be sporadic or due to an autosomal dominant genetic

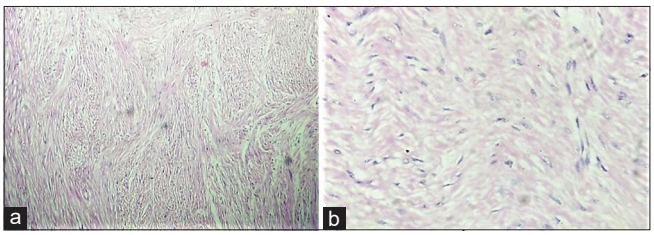

Figure 3: Microscopic view of piloleiomyoma (a) ×40 magnification and (b) ×100 magnification

syndrome. Multiple cutaneous and uterine leiomyomatosis, also known as Reed syndrome, occurs because of a germline mutation in the gene encoding fumarate hydratase (FH) on chromosome 1q42.3-43. When syndromic in presentation, they tend to occur earlier in life, with an average age of onset of lesions at 25 years old (ranges from 10 to 50 years old) [2-6].

Although unique in their clinical presentation, other similar lesions which may be considered during evaluation include dermatofibromas, Schwannomas, neurofibromas, angiolipomas, nevus, lipomas, eccrine spiradenoma, metastases, and angioleiomyomas [7,8].

While the gold standard treatment of piloleiomyomas is surgical excision, they have a high chance of recurrence, often up to 15 years. Other case reports have mentioned treatment modalities including cryotherapy, laser ablation, nifedipine, phenoxybenzamine, and gabapentin [9,10]. Usually, the uterine leiomyomas associated with FH mutation are larger (up to 10 cm), more numerous, and develop at a younger age. However, the most concerning feature of an FH mutation is the association with an aggressive variant of renal cell carcinoma (type 2 papillary renal cell carcinoma), which develops in approximately 15% of patients that are often metastatic on presentation in patients with hereditary leiomyomatosis and renal cell carcinoma [5-7].

Therefore, the presence of benign cutaneous leiomyomas must incite a search for more concerning abdominal pathology not only in the patient but also in first-degree relatives due to the possibility of autosomal dominant inheritance of renal cell carcinoma. In patients with diagnosed FH mutations, annual magnetic resonance imaging screening for renal cell carcinoma must be undertaken, and their children should also undergo annual screening, starting at the age of 8. Annual pelvic examination for uterine leiomyomas is also initiated in female patients [11].

CONCLUSION

While piloleiomyomas themselves have relatively easy surgical management, one must be prudent not to miss associated red herrings, namely uterine leiomyomas causing infertility and dysmenorrhea, and most importantly renal cell carcinomas, which might lead to significant morbidity and mortality. Appropriate genetic and clinical screenings must be initiated, and patients counseled regarding this seemingly benign disease with dire associations.

AUTHOR'S CONTRIBUTIONS

ST collected the clinical data, was part of the operating team, and contributed to the case discussion and literature review. BG guided the data collection, was the lead surgeon, and guided manuscript generation. SD guided the data collection and manuscript generation. AD contributed to the case discussion, manuscript generation, and literature review. VN contributed to the collection of the clinical data and literature review.

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