

Case Report

Interesting case of vulval lymphangiectasia following tubercular lymphadenitis

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We present an interesting case of acquired cutaneous lymphangiectasia of vulva following tubercular lymphadenitis. This was a young lady who initially had cervical lymphadenitis proven positive for tuberculosis on needle aspiration cytology and received Anti-tubercular Treatment. Soon after completion of therapy, she developed large oozing lesions on the vulva. There was initial diagnostic dilemma which was cleared on biopsy which confirmed acquired lymphangiectasia. Her symptoms were finally relieved with simple vulvectomy and she was doing well in two years of follow up. She had also conceived her second pregnancy by the end of second year of follow up.

Keywords: Acquired vulval lymphangiectasia, vulval lymphangiectasia, Tuberculosis

Key message: Unusual presentations of diseases may be encountered in clinical practice. Pulmonary TB is known to have high prevalence in India. Since, TB could affect any part of the body, the manifestations could also be challenging as in the present case.

INTRODUCTION

Acquired cutaneous lymphangiomas are characterized by presence of a circumscribed eruption of thin-walled, translucent vesicles. Diagnosis and treatment of these vesiculo-bullous lesions is important because they may be associated with pain, chronic drainage, cellulites, infection and psychosexual problems. When these arise on the vulva, diagnostic and treatment difficulties pose a challenge. We report a case of acquired lymphangioma of the vulva following tuberculosis.

The aim of this report is to bring forth unusual clinical manifestations following tuberculosis, the prevalence of which is high in our country.

Background

Vulval acquired cutaneous lymphangiomas pose diagnostic and treatment difficulties.

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Case report

30 year old, Ms. J, P₁L₁ presented in the Department of Medicine initially with cervical lymphadenitis; these proved positive for tuberculosis on aspiration cytology. She was given Anti Tubercular Treatment (ATT) for 8 months. After 1 month of completion of anti-tubercular treatment, she visited STD clinic with oozing and discharge from vulva and enlargement of labia (Figure 1). She was investigated for STDs and also the lesion was biopsied (Figure 2). The histopathological report confirmed lymphangiectasia. She was started on tablet Prednisolone 40 mg daily along with analgesics. There was initial symptomatic improvement but vulval edema persisted. Steroid therapy was tapered off slowly over 3 months but this was associated with reappearance of the disease. She was referred to us in the

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Figure 1. Preoperative appearance; acquired vulval lymphangiectasis. Multiple scars along inguinal ligament are seen following healing of the abscesses of tubercular lymph nodes

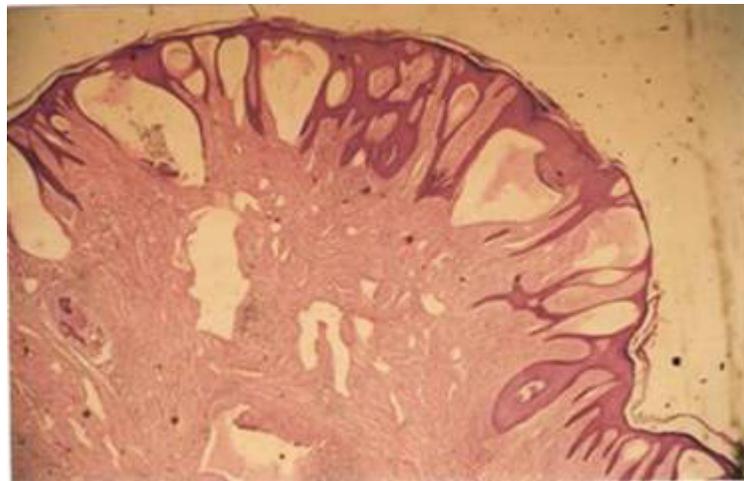


Figure 2. Dilated lymphatic channels in deep dermis with lymphocytic infiltration

Gynecology Department for opinion and definitive management.

On examination, she was a young woman of average built. There were multiple scar marks present along inguinal ligament suggesting ruptured abscesses. Bilateral labia were swollen and tender with multiple pustules and vesicles (Figure 1). Rest of the examination including gynecological examination was unremarkable. Superadded infection with citrobacter sensitive to ciprofloxacin was

proven on pus culture. She was given a course of the antibiotic for a week when there was slight improvement of symptoms.

Pelvic ultrasonography, chest X-ray and sputum for AFB did not reveal any abnormality.

After considering all options available for treatment, simple vulvectomy was performed. The procedure was fairly simple and there was no intraoperative or immediate postoperative problem. Histopathological report confirmed



Figure 3. Postoperative appearance after simple vulvectomy

acquired lymphangiectasia of vulva. The vulval scar healed well.(Figure 3)

The patient remained in our follow up for two years with no apparent recurrence of symptoms. During this period she conceived her second child too.

DISCUSSION

Lymphangiectasia is a term applied to lesions consisting of one or multiple groups of translucent vesicles, which represent an acquired or congenital dilatation of lymphatic channels. It is a nonfatal condition with a tendency to high recurrence after initial treatment. The known extrinsic factors include scarring from surgical procedures for keloid or following recurrent cutaneous infections, scleroderma, tuberculosis, repeated trauma and radiotherapy (Leshin et al., 1986; Ambrojo and cogolludo 1990). Cutaneous lymphangiectasia could be categorized as congenital (lymphangioma circumscriptum) and acquired forms. Lymphangioma circumscriptum occurs as a result of a congenital anomaly of the deep dermal and subcutaneous lymphatics. It is more localized and it may show up later in adult life. The pathogenesis of both these conditions involves saccular dilatation of local superficial lymphatics secondary to increased intra lymphatic pressure as a result of build up of lymph in the superficial vessels due to damage to previously normal deep lymphatics. The lesions

are usually thin walled vesicles often filled with clear fluid. However these vesicles may develop a much firmer hyperkeratotic appearance and often misdiagnosed and treated as viral warts (Sharma and Tomar 2002; Harwood and Mortimer 1993).

Recurrence following treatment is common in both conditions since removal/ablation of full depth of the abnormal lymphatics is difficult. Acquired lymphangiectasia are more problematic since the obstruction is more diffuse and complete surgical clearance may be nearly impossible.

Vulva is an uncommon site for the development of lymphangiectasia though rare lesions have been described both in scrotum and vulva (Russell and Prisie 1967). In a recent review of literature only 31 cases had been described (11 congenital and 20 acquired) (Vlastos et al., 2003). Majority of these lesions were following cancer of the cervix while three cases were following tuberculosis.

Recognition and appropriate treatment of vulval lymphangioma is important, primarily because the lesions may act as portal of entry for infection and severe cellulitis requiring parenteral antibiotics as in the present case. In addition persistent leakage of lymphatic fluid, the cosmetic appearance and the social repercussions of misdiagnosis as a sexually transmitted disease, may all pose significant problems.

Many treatment modalities have been advocated like electrodesiccation, laser therapy, sclerotherapy, cryotherapy and surgical excision (wide local excision,

simple vulvectomy or radical vulvectomy) though as yet there is no consensus as regards the best mode of treatment. Disease recurrence with any of these methods is high ranging from 25%-100% (Murugan et al., 1992; Short and Peacock 1995; Gordon and Kaufman 1999). This has also lead some clinicians to advocate observation of asymptomatic patients. We preferred surgery due to the ease of its availability and the patient was symptomatic.

Follow up care is essential for early treatment of recurrences.

Rarely malignant changes may occur in the lesions. Lymphangiosarcoma (Stewart-treves syndrome) an aggressive tumor may occur in chronic edematous limbs and early detection is critical.

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