ELEPHANTIASIS OF VULVA

(Two Case Reports)

by

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Elephantiasis of the Vulva is an elephantoid condition found sporadically in gynaecological practice. Its occurrence is less common than its counterpart in the male (Elephantiasis of the Scrotum). This may be encountered in this country more commonly due to lymphatic obstruction by filaria sanguinis. It may rarely be caused by granuloma inguinale, lymphogranuloma venereum, syphilis, tuberculosis, cancer, sarcoma and occasionally it may be idiopathic. Two cases of Elephantiasis of Vulva (one with associated R.V.F.) are reported because of their rarity.

Case I

Mrs. S. S., aged 35 years P_3 + O, Hindu was admitted in Eden Hospital on 10-11-77 with the complaints of gradual enlargement of vulva for last 5 years, which to start with were small bilateral swellings but later on had rapidly grown to the present size. Her bladder and bowel functions were normal.

Menstruation, regular. No history of tuberculosis or sexually transmitted diseases.

Examination: She was of average built, B.P. 125 mm. of Hg. Systemic and abdominal examinations revealed no abnormality. Local examination showed huge swelling of Vulva (fig 1), involving both labia majora, minora and mons veneris. The right labia was enlarged to 6″ x 4″ x 3″ and the left to 5″ x 3″ x 3″ . These swellings were solid, irregular and sessile, not fixed to the bone. The overlying skin was thickened, rugose, and polypoid. The inguinal lymph nodes were not palpable.

Vaginal examination showed normal healthy vagina. Cervix was normal. Uterus was anteverted and normal in size. Appendages were not palpable.

Investigations: Hb, 10.5 gm%; T.C.-7850 per c.m.m., poly, 66% lympho, 25%, nomo, 1% and Eosino, 8%, E.S.R., 15 m.m. (1st hour by Wintrobe method). Blood for microfilaria and V.D.R.L. were negative. X'Ray chest and Mantoux test-negative, Frie's test-negative.

Operation: Simple vulvectomy was done on 16-11-77 and the wound healed by primary union.

Her condition was satisfactory at follow up after 6 weeks.

Histological Report

Squamous lining of the vulva is for the most part atrophic and thin with presence of many dilated lymphatics and dense chronic inflammatory cellular exudate underneath epidermis. The connective tissue element looks hypertrophied. The picture is consistent with filarial elephantiasis of vulva.

Case II

Mrs. A.S.R, aged 35 years, P_4+ $_0$ was admitted on 26-4-77 with bilateral swellings of vulva for last 4 years, on which ulcer deve-

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loped during last 1 year. She was also feeling burning sensation during micturation for 2 years. Associated with this there was leakage of stool from the vagina since her last child birth, 6 years ago.

Menstrual History—Menarche 13 years, Cycle 3-5 days. She was having no menstruation $\frac{28 + 2}{}$ since her last child-birth 6 years ago.

Obstetrical History—She had 4 normal pregnancies and labour, the last one was prolonged and according to her statement the presentation was breech but she could not recollect whether the delivery was assisted by instrument or not. She noticed purulent offensive discharge during defaecation few days after delivery followed by vaginal bleeding. Though vaginal bleeding ceased to occur, the leakage of stool through vagina persisted even after medical treatment. Her family and past histories were not significant, but her husband as she stated was suffering from penile ulcer. He died 6 years ago.

On examination she was of average built, B.P. $\frac{110}{80}$ m.m. of Hg. Systemic and abdominal examination revealed no abnormality. Local examination of vulva showed bilateral swellings, the right labia was 8″ x 7½″ x 6½″ and the left 9½″ x 7½″ x 7″. The masses were irregular in shape and sessile. The overlying skin was thickened and there was an indolent ulcer on inner aspect of right labia majora. Polypoid growth was present over perineal region (cutaneous).

Vaginal examination was done with difficulty which revealed healthy vagina and cervix. Uterus was normal in size, mobile and retroverted. Fornices were clear. One fistulous opening 1″ in diameter with thick irregular margins was communicating from the lower part of the vaginal wall to the rectum and anus. Rectal examination confirmed the communications. Rectal wall was thickened, oedematous and irregular around the fistulous opening. There was an annular narrowing of the rectum at that level. Rectum was fixed. Inguinal lymph nodes were not enlarged.

Investigations: Blood for V.D.R.L. and microfilaria—negative, Chest X-Ray showed no abnormality. Total W.B.C. counts—8900, with 12% eosinophilia, E.S.R. 90 mm, 1st hour (wintrobe). Urine and stool showed no abnormality. Mantoux test-negative Frie's skin test-inconclusive. No Donovan bodies in large mononuclear cells. Barium enema X-Ray showed no obstruction. Proctoscopic examinations—Perineal induration extending to vulva. Ulceration of anal canal, bleeds on touch. Large R.V.F., upper limit of which could no be visualised properly. Clinically it might be confused with papillary carcinoma of rectum due to surrounding polypoid indurated mass around anal canal. Biopsy report of both sided vulval swellings showed marked epithelial hyperplasia with thickening of fibrocollagenous tissues, enclosing marked round cell infiltrations. Biopsy report of anal wall showed squamous epithelium with hypertrophy and chronic inflammatory cells in the subepithelial tissue. No evidence of tuberculosis or malignancy seen.

Diagnosis: Elephantiasis of valva with R.V.F.

Treatment: The patient was treated with Tetracycline (500 mg x 4 times daily) continuously for 6 months, when the vulval swelling almost subsided. The R.V.F. though still persisted was spontaneously healing. The patient is under treatment and operative repair of fistula if necessary at all will be undertaken after months when the perirectal induration is likely to subside.

Discussions

Two cases of Elephantiasis of Vulva have been presented. The condition may clinically be present in different forms, small warty growth, huge masses or simply a browny induration with thickening of the skin. Superficial ulceration which was present in the 2nd case is usually a feature of elephantiasis of vulva and it has got a tendency to spread to the groin, upper part of the thighs and buttocks.

In the first case the patient came from an area where filaria is endemic. Though the patient did not show any evidence of active filariasis, such as rise of temperature or microfilaria in the blood, yet the general appearance of the skin overlying the vulval swellings, the presence of many dilated lymphatics on biopsy examination of the vulval tissue suggested that the elephantiasis of vulva in this

case was possibly be of filarial origin, though the total histological picture was also compatible with chronic vulvitis, which may be idiopathic.

In the second case association of R.V.F. with elephantiasis of vulva posed a diagnostic problem. On etiological basis R.V.F. may be associated with elephantatic condition of vulva in lymphogranuloma venereum, hypertrophic type of tuberculosis of vulva, vulval carcinoma involving rectum; however in all these conditions, elephantiasis of vulva develops earlier followed by R.V.F. In the 2nd case the reverse was true, and the patient gave a history of difficult labour before vulval swellings appeared.

The etiology thus seemed to be quite different from the first case. The history of penile sore in the husband, annular stricture in the rectum of the patient, ulceration in the anal canal, fistula, proctitis, histopathological report of marked round cell infiltration, almost complete suppression after tetracycline therapy suggested that the elephantiasis of vulva in this case may be due to lymphogranuloma venereum. Inconclusive Frei's test does not support this diagnosis, yet all the findings taken together, go in favour of the above mentioned etiology. Absence of Donovan bodies negatives the diagnosis of granuloma inguinale. The possibility of tuberculosis is ruled out in the absence of giant cell systems in histological report and the condition occurs only in advanced tuberculosis elswhere. Malignancy of the rectum is unlikely to give rise to such vulval swellings as more spread was expected. In the present case R.V.F. is a chance association.

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See Fig. on Art Paper VII