ENDOMETRIAL STROMATOSIS

(A Case Report)

by

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Introduction

Endometrial stromatosis is a rare lesion characterized by the neoplastic proliferation of mesenchymal cells, resembling those of endometrial stroma. It arises from the stromal cells of the endometrium, foci of adenomyosis or nests of ectopic stromal cells within the myometrium. Its distribution within the uterine corpus, suggested a close relationship to adenomyosis and this probably accounts for the various diagnostic terminology used to describe this entity,—like stromal endometriosis, stromal adenomyosis and endometrioma interstitial. It was originally confused with haemangiopericytoma (Norris and Taylor, 1966).

This entity was first described by Doran and Lockyer in 1908, who described its malignant growth pattern and benign clinical behaviour. Since then there are several reports and reviews on this entity. There are 16 such case reports in the Indian Literature—the earliest that of Mangalik and Wahil (1954) and the last one by Bhavthanker *et al* (1977). In World Literature about 120 such cases have been studied and reported. It is a comparatively rare condition and the most controversial aspect of this condition concerns its malignant potentiality. Recurrences and distant metastases have been noted indicating that it is a form of malignant neoplasm and therefore several authors prefer to classify such tumors as endometrial stromal sarcomas (Koss *et al* 1965). Few others have indicated (Krieger and Gusberg, 1973) that its biologic behaviour is different and therefore warrants a seperate identity. Considering the rarity and the controversial nature of this condition we are reporting a case, with a review of the pertinent literature. This is the first such case, seen in our department here.

CASE REPORT: A 50 year old female came to the hospital with the complaints of irregular bleeding per vaginam since 5 years. She had 4 full term normal deliveries, the last one being 22 years ago.

Physical examination revealed only anaemia (haemoglobin 9 gms%). Systemic examination was unremarkable. Pelvic examination revealed on enlarged uterus which was thought to be due to "Fibroids". A total hysterectomy was subsequently performed. The uterus measured 9 x 5 x 3 cms. The cut surface of the corpus revealed multiple intramural tumour masses

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having a soft yellowish to tan brown appearance. The tumor nodules were of variable sizes, the largest being 1 cm in diameter. The myometrium was thickened (6 cms.). The cervix was unremarkable.

MICROSCOPIC EXAMINATION: Several sections were studied using the routine H & E staining procedure. The endometrium was involved by stromatosis. The neoplastic cells surrounded and compressed normal endometrial glands, in addition to separating them widely. The tumor tissue consisted of cells closely resembling the stromal cells. These tumor masses appeared as worm-like extensions, dissecting and separating bundles of smooth muscle cells (Fig. 1). Groups of tumor cells were seen bulging into lymphatic and vascular spaces. The neoplastic cells were remarkably uniform in size and shape, small, with oval elongated nuclei and scanty cytoplasm (Fig. 2). Pleomorphism was absent. There was no atypia or anaplasia of the nuclei. There were not more than 1-2 mitotic figures per 10 HPF. The supportive tissue was richly vascular consisting of uniformly distributed small delicate capillaries or venules. Well formed endometrial glands were not seen in any of the sections. Sections from the cervix were unremarkable.

Discussion

Doran and Lockyer (1908) who first described this entity called it perithelioma of the uterus. Since then several authors have described it variously. It has been called stromatosis by Robertson *et al* (1942) and Hunter (1953), Hill (1947) called it fibromyosis uteri. Pedowitz (1954) considered it to be a Haemangiopericytoma, while Rosenberg *et al* (1964) called it Stromeloma. Koss (1965) considered it to be an endometrial sarcoma.

Norris and Taylor (1966) studied the nature and behaviour of these tumors and divided them into a benign pushing type and a malignant type with infiltrating margins. Hart and Yoonesi (1977) have laid down specific histopathologic criteria to distinguish this condition from stromal sarcomas. Stromal sarcomas have high mitotic rates—10-20 mitoses per 10 HPF and the cells have anaplastic nuclear changes. They have a destructive growth pattern when they infiltrate the myometrium and do not have a prominent intravascular component. In contrast stromatosis have sparse mitotic activity 5-6 per 10 HPF or less with hardly any nuclear anaplasia. Goldfarb *et al* (1970) calculated that the nuclear DNA content of stromatosis was of a diploid or tetraploid distribution while malignancy was associated with aneuploid DNA content. Ultrastructural studies by Akhtar *et al* (1975) and Komorowski *et al* (1970) have shown similarities between the cells of stromatosis and the stroma of an early or mid proliferative endometrium.

The clinical course of patients with stromatosis is more favourable-while recurrences are seen, they are not inevitable and often it is a number of years before they become clinically apparent (Baggish *et al* 1972). Norris and Taylor (1966) recorded a 5 year survival rate of 100% for 19 patients with infiltrative stromatosis. Hart and Yoonesi (1977) noted a recurrence in 7 of their 9 patients, the interval varying from 3 to 14 years.

Optimum therapy for patients with stromatosis should consist of a minimum of total abdominal hysterectomy. The propensity for intravascular extension into the parametrium, broad ligament and adenexal organs strongly suggest that bilateral salpingo-oophorectomy should also be performed. The possible stimulatory effects of oestrogen from retained ovaries on the neoplastic stromal cells can also be considered theoretically—but there is no proof for the same. Baggish and Woodruff (1972) have suggested an ovarian oestrogen dependence since their

reported case responded to bilateral ovariectomy. Norris and Taylor (1966) on the other hand, find this therapy to be of no benifit at all. Progestational hormonal therapy has been of value in a few reported cases (Baggish and Woodruff 1972; Krumholz *et al* 1973; Pellillo 1968). Radiation therapy has also been tried in a few cases (Koss 1965, Norris and Taylor, 1966). But on the whole the role of hormonal therapy, chemotherapy and radiotherapy for the treatment of stromatosis remains very much speculative.

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