ADENOACANTHOMA OF RECTAL ENDOMETRIOSIS

by

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Malignancy arising in endometriosis is unusually rare and such a change in bowel endometriosis is rarer still. The earliest reference to the occurrence of malignancy in bowel endometriosis is that of Hosoi and Meeker (1929) who described an adenocarcinoma in endometriosis of the transverse colon. Nylander as quoted by Jenkinson and Brown (1943) reported a spindle cell sarcoma of the rectum containing endometrial tissue. Weinrod, *et al* (1956) have recorded an adenoacanthoma of sigmoid endometriosis. There have been isolated case reports of adenocarcinomas arising in endometriosis of the rectovaginal septum which have infiltrated the rectum. (Docherty, *et al* 1954; Ferreira and Clayton 1958; Lash and Rubenstone 1959). The following case, therefore, appeared worthy of record.

Case Report

A married woman aged 51 years was admitted to Harrow hospital on the 13th of September 1966. She complained of constipation alternating with diarrhoea for about 15 years. For most of this time she passed mucus in her stools and for the last few years had frequent episodes of bleeding per rectum. She also complained of considerable pain in the rectum and perineum for a few months before admission. For about 12 years she suffered from recurrent bouts of pyrexia. She had a ruptured ectopic pregnancy 12 years previously and 2 years later underwent hysterectomy and right oophorectomy for endometriosis.

On admission her general condition was good and abdominal examination was normal. On rectal examination there was a hard, extramucosal, tender mass on the right side and anteriorly. Pressure on this mass produced pain she had recently experienced. Her routine blood tests, urinalysis, and barium enema were normal.

A laparotomy was performed on 10th October 1966. A hard mass was seen under the pelvic peritonium. Her remaining left ovary was atrophic. Histological examination of the biopsy from this mass revealed a poorly differentiated squamous carcinoma. Further exploration at the second laparotomy on 15th October 1966 showed this mass to be in the anterior rectal wall. It was adherent to the vaginal vault and the base of the bladder. There were no visible liver or lymph node metastases. Abdomino-perineal resection of the rectum and left iliac colostomy were performed. Postoperative recovery was uneventful.

Pathology: Examination of the specimen showed a tumour in the anterior wall of the rectum on the right side, 10 cm. above the anus. The mucosa overlying the tumour was largely intact with several ulcers, the largest about 5 mm. An irregular piece of vagina 3 x 2 cm in size was adherent to the anterior surface of the tumour.

Histology showed a poorly differentiated adenocarcinoma with areas of squamous metaplasia invading the rectal wall from without (Fig. 1 and 2). Between vaginal vault and the tumour lay an area of endometriosis, possibly the origin of this tumour. (Fig. 3 and 4). All glands were involved.

The patient died about a year later but details regarding the cause of her death were not available.

Discussion

It is not safe to assume that a malignant tumour has arisen in an endometrial

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tissue merely because endometriosis is also present or because the tumour morphologically resembles endometrial carcinoma. The clinical evidence for the view that this growth arose in endometriosis seems however satisfactory. Exploration of the abdomen at the time of resection showed no other intra-abdominal pathology. Gross and microscopic study of the specimen failed to show the origin of this tumour from the rectal mucosa. Endometriosis and cancer can be seen in one histological block in adjacent parts although direct transition between the two is not seen. Docherty *et al* (1954) felt that insistance on this criterion would only serve to discourage the reporting of bona fide cases.

Squamous metaplasia seen in this tumour deserves some comment. Since the first description of an adenoacanthoma in the coecum by Herxheimer in 1907 this histogenetically interesting neoplasm has occasionally been described at intraepithelial junctions, such as cervix (Hamperi and Hellung 1957), ano-rectal region (Key, 1954), oesophago-gastric junction (Dodge 1961) and at various other sites in the body (Nicholson (1923). Nevertheless, acanthosis in an adenocarcinoma is not common in tumours other than those of endometrial origin. The rarity of adenoacanthoma of the rectum is illustrated by the fact that all the 1,000 cases of rectal cancer reviewed by Dukes (1940) were adenocarcinomas. There seems to be a considerable stimulus to squamous metaplasia in endometrial carcinomas the reason for which is of interest but unknown. It was observed in 9 of 50 cases of endometrial carcinomas by Willis (1967) a proportion closely similar to Miller's figure of 15% (1950).

Nicholson (1923) found squamous metaplasia in 14 of 36 endometrial carcinomas and Charles (1965) in 55 of 148 cases. Other authors have been more critical. Healey and Cutler (1930) showed acanthosis in only 3 out of 100 endometrial carcinomas while Novak and Nalley (1957) state a still lower incidence. Squamous metaplasia is also a well recognised feature of ovarian tumours arising in endometriosis. Although epidermoid transformation is seen in ovarian cystadenoma, its malignant counterpart, and some other ovarian tumours, in Thompson's opinion (1957) most primary ovarian adenoacanthomas arise in endometriosis. Freedman, *et al* (1964) observed that 25 of 57 ovarian adenoacanthomas had their origin in endometriosis. Ferreira and Clayton (1958) found 23 cases in which conditions for malignant change in endometriosis were fulfilled. Of these, 13 were adenoacanthomas or of epidermoid type and 10 were adenocarcinomas. Assuming that other criteria are fulfilled it does not seem unreasonable to consider squamous metaplasia in an adenocarcinoma as additional evidence of the origin of a tumour in endometriosis.

Endometriosis is essentially a benign condition and various authors have emphasized the need to avoid unnecessary radical surgery. Surgical treatment of this condition is almost never dictated by any concern over possible malignant complication. Considering the frequency of endometrial carcinoma in the uterus it seems curious that malignancy in endometriosis should be so rare. The true incidence of such a change is extremely difficult to determine since the evolution of cancer from foci of endometriosis is often difficult to verify. Such cancers are frequently symptomless in early stages and when detected the tissue of their origin may be obscured. Also, there is considerable divergence of opinion among pathologists regarding the criteria for accepting the origin of malignancy in endometriosis.

In a study of 889 cases of ovarian endometriosis and 265 cases of ovarian malignancies Corner, *et al* (1950) found only 3 undoubted and 3 probable cases of malignant change in endometriosis. Thompson (1957) could demonstrate such a change in only 1 of 181 cases of endometriosis subjected to thorough histological study. Fathala (1967) reported 4 cases of endometrial carcinomas in a survey of 592 cases of ovarian endometriosis and 418 cases of primary malignant ovarian tumours. However, in the same series malignant tumours histologically similar to endometrial growths were encountered in 52 cases. Long and Taylor (1964) have questioned the assumption that malignancy in endometriosis is so rare. They studied 120 cases of malignant ovarian tumours and found 20 had structural resemblance to endometrial carcinoma suggesting an origin in endometriosis. Kottmeir (1952) and Docherty (1954) have also commented on the frequency with which endometrioid carcinomas are seen in the ovary. The low incidence of reported cases may be partly due to strict compliance with the requirements dictated by Sampson (1925). It is fair to state that malignant change in endometriosis is such an unlikely possibility considering the frequency of this condition that we are not justified in allowing this consideration to alter our surgical judgement regarding the treatment of this condition. The importance of recognising such a change as a rare occurrence in an individual case is however obvious.

Summary

A rare case of adenoacanthoma arising in rectal endometriosis is described. Adenoacanthoma, while rare in other sites, is a relatively frequent form of tumour arising in tissues of endometrial origin. Relevant literature is reviewed.

Malignant change in endometriosis is very rare. Incidence however seems to vary according to the criteria adopted and is probably a little more than the paucity of case reports suggests.

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See Figs. on Art Paper X