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Endometrial stromal sarcomas with extensive endometrioid glandular differentiation: report of a series with emphasis on the potential for misdiagnosis and discussion of the differential diagnosis

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Endometrial stromal sarcomas with extensive endometrioid glandular differentiation: report of a series with emphasis on the potential for misdiagnosis and discussion of the differential diagnosis

Aims: To describe a series of endometrial stromal sarcomas with large numbers of endometrioid-type glands.

Methods and results: The eight tumours occurred in patients aged 42–74 years. In three cases, the neoplasm arose in the uterine corpus and in the others there was either an extrauterine origin or the origin could not be determined since multiple sites were involved. In four cases, glands were present throughout the neoplasm and in the others there were areas of typical endometrial stromal sarcoma without glands. In one case, glands were present only in the recurrent neoplasm. The malignant stromal component comprised, for the most part, typical endometrial stromal sarcoma. In two patients, repeated biopsy specimens from the vagina or cervix were initially diagnosed as endometriosis, and in some cases there was a significant delay in diagnosis. Apart from endometriosis, other diagnostic considerations, depending on the tumour location and exact morphology, included adenomyosis, adenosarcoma and carcinosarcoma.

Conclusions: Endometrial stromal sarcoma with extensive endometrioid glandular differentiation is rare. The presence of glands often results in diagnostic difficulty with a significant risk of misdiagnosis or delay in diagnosis. It is likely that some cases reported in the literature as aggressive endometriosis represent this entity.

Keywords: endometrial stromal sarcoma, endometrioid glands, endometriosis

Abbreviations: ER, oestrogen receptor; ESS, endometrial stromal sarcoma; PR, progesterone receptor

Introduction

Endometrial stromal sarcomas (ESSs) are relatively uncommon neoplasms, usually arising within the uterine corpus but occasionally at extrauterine sites.[1,2] We use the term ESS to denote those morphologically bland neoplasms that were previously referred to as low-grade ESS, since the former is in widespread use and is the preferred terminology of the World Health Organization.[3] Most ESSs, especially those arising in the uterine corpus, have a characteristic and relatively constant morphological appearance with an infiltrative tongue-like growth pattern, prominent vascularity and a monotonous population of regular cells resembling proliferative phase endometrial stromal cells; as such. they are usually easily diagnosed. However, various features may complicate the appearance of endometrial

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stromal tumours, both ESS and the less common endometrial stromal nodule. These include sex-cord-like and smooth muscle differentiation and a fibrous or myxoid appearance.[4–7] Other rare variations described include a pseudopapillary architecture, an epithelioid, rhabdoid or clear cell phenotype, adipose tissue or skeletal muscle formation and nuclear atypia of symplastic type.[8–15] A small series of three uterine ESSs with extensive endometrioid glandular differentiation has been reported by Clement and Scully,[16] as well as a single similar case.[17] There has been a single report of a probable extrauterine ESS with extensive endometrioid glandular differentiation.[18] However, it is our opinion that the potential occurrence of endometrioid glands in endometrial stromal neoplasms is not well known to pathologists, and in our experience this may result in diagnostic difficulty. In this study, we report a series of a modest number of ESSs, both uterine and extrauterine, with large numbers of endometrioid-type glands. The majority of cases, most of which were seen in consultation, resulted in diagnostic problems, sometimes with considerable delay in diagnosis. We discuss the differential diagnosis in detail.

Materials and methods

The cases were derived from the pathology archives of the institutions to which the authors are affiliated. Most of the cases were from the consultation files of one of the authors (W.G.M.) and of Dr T. P. Rollason (Birmingham Women's Hospital, Birmingham, UK). Clinical details were obtained from the pathology reports, referral letters or liaison with the clinician or consulting pathologist. All available haematoxylin and eosin-stained slides were examined as well as immunohistochemistry, which had been performed as part of the work-up of the cases. Follow-up was obtained by liaison with the clinician or consulting pathologist.

Results

CLINICAL DETAILS

The patients in cases 1–8 were aged 74, 50, 64, 51, 62, 47, 42 and 59 years, respectively, at the time of first presentation. The primary site was considered to be in the uterine corpus in three cases (cases 1, 3 and 7) and the soft tissues of the pelvis and abdomen in two (cases 4 and 8). In the other cases, multiple sites were involved and the origin could not be confidently ascertained. The sites of tumour involvement are listed in Table 1. Table 2 lists the various surgical and biopsy procedures, together with the original pathological diagnoses made by the reporting or referring pathologists. All of the tissues biopsied or resected were considered on review to represent ESS with endometrioid glands, except where stated in Table 2.

Table 1. Sites of tumour involvement

	All sites of tumour involvement	Sites of tumour with endometrioid glands
Case 1	Uterine corpus, cervix, left ovary, paracervical tissues	Uterine corpus, cervix, left ovary, paracervical tissues
Case 2	Uterine corpus, cervix, vagina, bladder, serosa of rectum	Uterine corpus, cervix, vagina, bladder, serosa of rectum
Case 3	Uterine corpus	Uterine corpus
Case 4	Omentum, small bowel mesentery, pelvic lymph nodes	Pelvic lymph nodes
Case 5	Vagina, left ovary, omentum, peritoneum, serosa of rectum and sigmoid	Vagina
Case 6	Cervix, vagina, serosa of rectum	Cervix, vagina, serosa of rectum
Case 7	Uterine corpus, vaginal vault	Vaginal vault
Case 8	Intestinal serosa, peritoneum	Intestinal serosa, peritoneum

In cases 1, 4 and 5, the patients were commenced on unopposed oestrogen following the hysterectomy and unilateral or bilateral salpingo-oophorectomy. In case 2, there was a period of 7 years between the first vaginal biopsy and establishing a definitive diagnosis of malignancy. In case 4, residual disease remained following the tumour resection and the patient was given postoperative radiotherapy and gonadotropin-releasing hormone agonists. In case 5, the patient received six cycles of adriamycin and ifosfamide chemotherapy following resection of the tumour. In case 6, six cycles of adriamycin chemotherapy were administered before surgery.

FOLLOW-UP

In case 1, the patient is alive and well with no evidence of tumour recurrence 5 years following removal of the cervical neoplasm. Cases 2 and 3 are recent and there is no significant follow-up. The patient in case 4 is

Table 2. Surgical procedures and pathological diagnoses

	Original surgical procedures	Original pathological diagnoses	Final surgical procedures	Final pathological diagnoses
Case 1	Subtotal hysterectomy and BSO 3 years previously	Adenomyosis, endometriosis	Cervical biopsies and cervical stumpectomy	ESS with endometrioid glands
Case 2	Repeated vaginal and cervical biopsies. Rectal resection	Endometriosis	Hysterectomy, BSO and further rectal resection	Adenosarcoma
Case 3	Endometrial polypectomy	Carcinosarcoma	Hysterectomy and BSO	ESS with endometrioid glands
Case 4	Hysterectomy and BSO 5 years previously	Uterine leiomyomas (confirmed on review)	Removal of omental and mesenteric tumour	ESS with endometrioid glands
Case 5	Hysterectomy and right ovarian cystectomy 23 years previously. Vaginal biopsies	Ovarian serous cystadenoma (confirmed on review). Vaginal biopsies diagnosed as endometriosis	Partial vaginectomy, BSO and omentectomy	ESS with endometrioid glands
Case 6	Cone biopsy of cervix	Adenosarcoma	Hysterectomy, BSO, partial vaginectomy and rectal resection	ESS with endometrioid glands
Case 7	Hysterectomy and BSO 7 years previously	ESS of uterus (confirmed on review)	Removal of mass in vaginal vault	ESS with endometrioid glands
Case 8	Hysterectomy and BSO 5 years previously	Uterine leiomyomas and ovarian endometriosis (confirmed on review)	Small intestinal resection and peritoneal biopsies	ESS with endometrioid glands

BSO, bilateral salpingo-oophorectomy; ESS, endometrial stromal sarcoma.

currently alive and asymptomatic 40 months following resection of the omental mass, but with known residual tumour within the abdomen. She is maintained on gonadotropin-releasing hormone agonists. The patient in case 5 is alive and well with no evidence of tumour recurrence 11 years following the diagnosis of ESS. We have no follow-up in cases 6 and 8. In case 7, a recurrent tumour nodule was discovered in the recto-vaginal septum 7 years following removal of the vaginal mass. This was not removed, but has been followed up by imaging and has remained the same size for 5 years.

PATHOLOGICAL FINDINGS

There were no particular gross lesions noted in the pathology reports, except where stated in this paragraph. In case 1, the cervical stump was totally replaced by tumour, which also involved the paracervical tissues. A 90-mm, partly solid and partly cystic mass involved the ovary in the prior hysterectomy specimen. In case 2, no gross lesion was noted on the pathology report, the tumour being identified on histological examination. In case 3, an 18-mm polypoid lesion was located at the fundus of the uterus in the hysterectomy specimen. In case 4, a 120-mm omental and mesenteric mass was submitted as well as a 40-mm lymph node mass. In case 5, an 80-mm mass was present in the left ovary as well as several tumour nodules in the omentum. In case 6, a 25-mm tumour was identified in the hysterectomy specimen involving the cervix and posterior vaginal fornix and extending onto the serosal surface of the rectum. In case 7, the original uterine tumour measured 50 mm in

maximum dimension and had a gross appearance of multiple polyps, but also with obvious tumour invasion of the myometrium. The recurrent tumour removed from the vaginal vault measured 115 mm in maximum dimension and was yellow. In case 8, multiple tumour deposits were present on the serosa of the small intestine.

In four cases, the entire neoplasm contained endometrioid-type glands and in the others there were areas of typical ESS without glands (Table 1); in the latter cases, glands involved from 10% to 50% of the neoplasms. In case 7, glands were present only in the recurrent neoplasm. In three of the cases (cases 1, 2 and 7) with uterine corpus involvement, there was widespread infiltration of the myometrium with a tongue-like pattern (Figure 1). In the other case (case 3), there was only limited infiltration of the inner half of the myometrium.

The areas of pure ESS and the stromal component in the areas with glands were composed of a monotonous population of small bland cells with ovoid to short spindle-shaped nuclei and generally scanty cytoplasm. There was at least focally a network of small arteriole-like vascular channels, and densely hyalinized acellular collagen bands were sometimes present. Mitotic figures were always identified with between 5 and 15 per 10 high-power fields with the area of a single high-power field being 0.55 mm^2. In case 6, the stromal cells focally had abundant eosinophilic cytoplasm, suggestive of pseudodecidualization. In cases 3 and 5, the stromal cells focally had abundant clear cytoplasm. Foam cells were present in the stroma in one case (case 6). In case 1, scattered individual or small groups of stromal cells were enlarged and atypical with multinucleate forms, the features being reminiscent of symplastic change in a uterine leiomyoma.

The endometrioid glands were usually relatively evenly distributed, either throughout the whole of the neoplasm or in focal areas, and were typically lined by a single layer of bland cuboidal epithelial cells (Figure 2). For the most part, the glands were of small calibre, but sometimes they were cystically dilated (Figure 3). In all cases, even in the areas with glands, the stromal component predominated and the glands were usually widely separated by an obviously expansile low-grade malignant stromal component. In the case with tumour confined to the uterine corpus (case 3), the endometrioid glands focally had a back-to-back architecture with stromal exclusion and associated

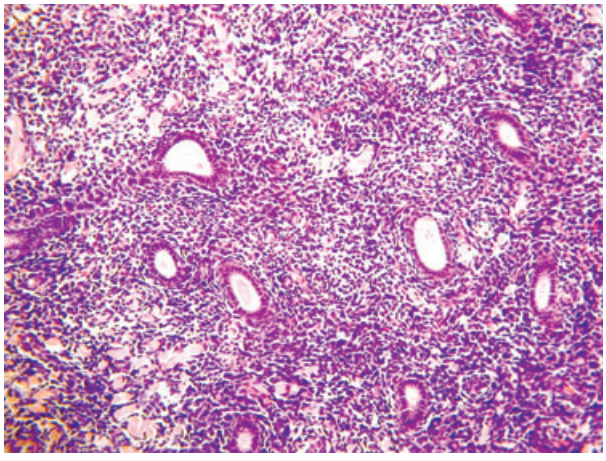

Figure 2. Endometrial stromal sarcoma containing benign endometrioid-type glands that are evenly distributed.

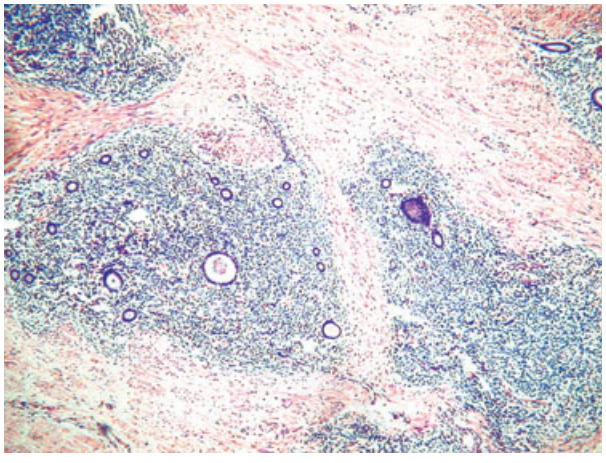

Figure 1. Endometrial stromal sarcoma with endometrioid glands diffusely infiltrating the myometrium, resulting in an appearance closely resembling adenomyosis.

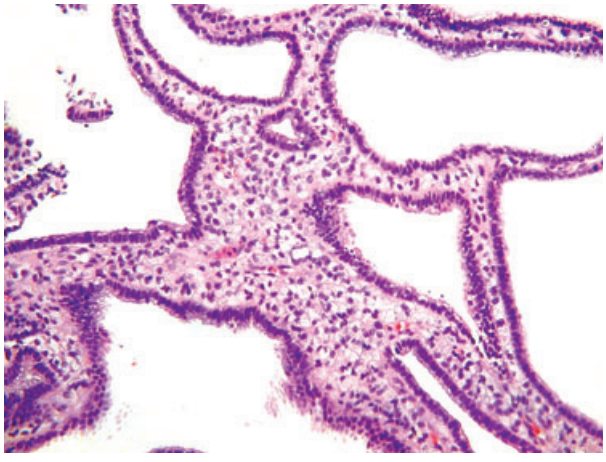

Figure 3. In some cases, the endometrial glands are cystically dilated.

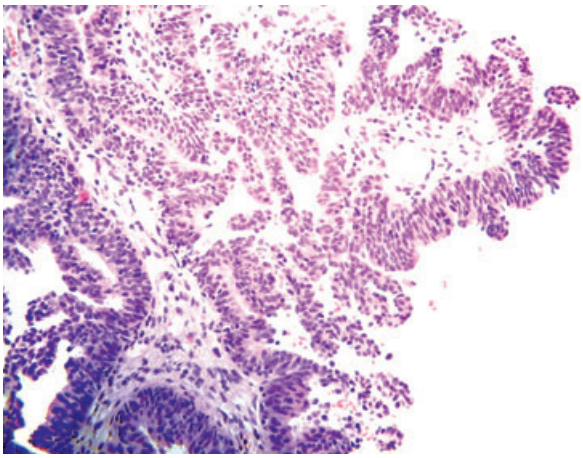

Figure 4. Area of glandular crowding and nuclear atypia amounting to grade one endometrioid adenocarcinoma.

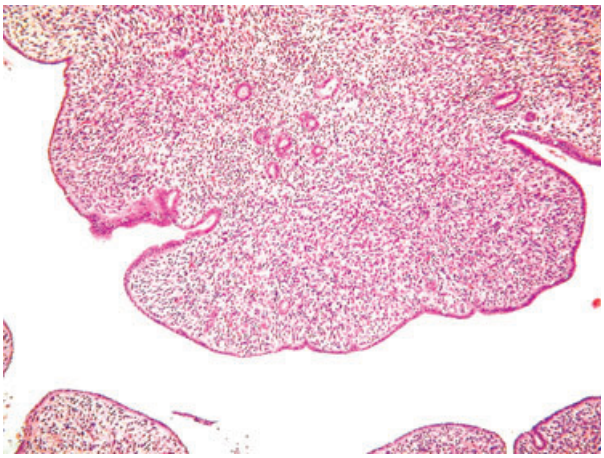

Figure 6. Endometrial stromal sarcoma with glandular differentiation where the tumour has a polypoid architecture with periglandular cuffing, the morphology being reminiscent of adenosarcoma.

nuclear atypia, amounting to a grade one endometrioid adenocarcinoma (Figure 4). In two other cases, there were focal areas of glandular crowding, resulting in an appearance reminiscent of complex hyperplasia within the endometrium (Figure 5).

Areas of smooth muscle differentiation were present in one case (case 4), and sex-cord-like elements in two (cases 3 and 7). In case 7, the sex-cord-like elements were present only in the original uterine ESS that did not contain glands. Small foci of metaplastic bone were present in case 5. In four cases, there were areas with a polypoid architecture (in two cases involving the cervix and two the vagina), sometimes with periglandular increased cellularity, the morphology resembling that of an adenosarcoma (Figure 6). However, this was always a focal phenomenon with areas of typical ESS with or without glands elsewhere. In some cases, there were individual areas of tumour that were indistinguishable from endometriosis (extrauterine tumour) or adenomyosis (uterine tumour), although these areas were a minor component and, for the most part, the stromal component was obviously malignant and widely separated the glands. In case 6, there was definite endometriosis involving the left ovary, and in case 8 there was definite endometriosis in both ovaries in the earlier resection specimen.

Vascular invasion was seen in three cases (Figure 7); in one case, this was by ESS with glands and in the others by ESS without glands. Perineural infiltration was seen in one case (Figure 8).

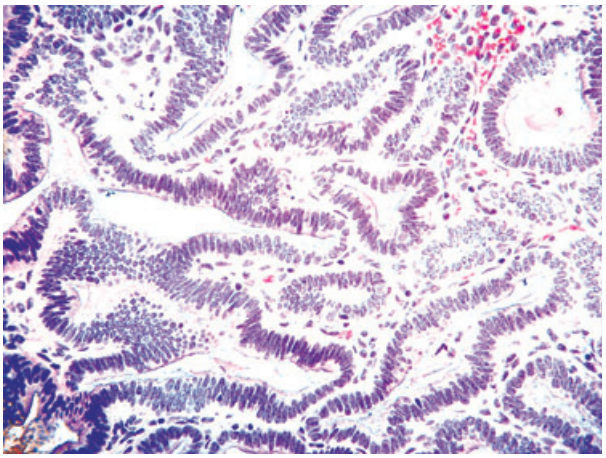

Figure 5. Case where glands are crowded, resulting in an appearance reminiscent of complex hyperplasia.

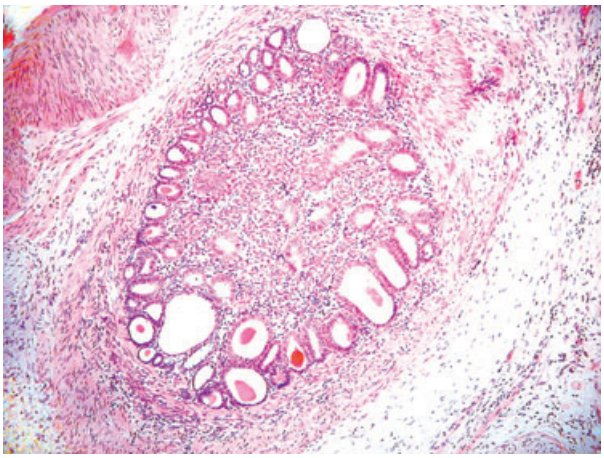

Figure 7. Endometrial stromal sarcoma with glandular differentiation within a thick-walled muscular blood vessel.

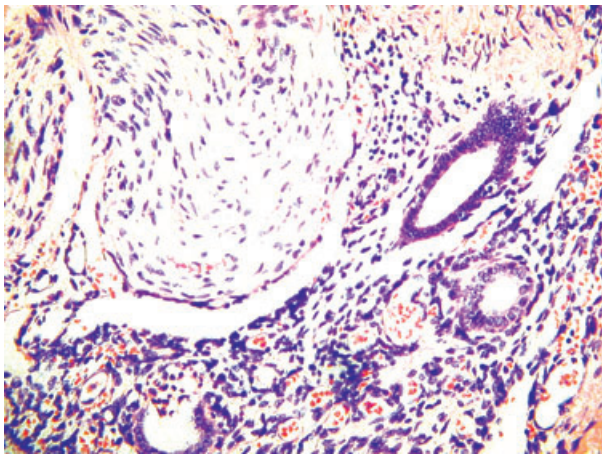

Figure 8. Endometrial stromal sarcoma with glandular differentiation exhibiting perineural infiltration.

Immunohistochemically the neoplastic stromal cells were CD10+ in two of three cases tested. Four of four and three of three cases, respectively, were positive with oestrogen receptor (ER) and progesterone receptor (PR). Two cases were desmin and one smooth muscle actin negative. The sex-cord-like elements in case 3 were focally cytokeratin and inhibin positive and diffusely positive with CD56.

Other pathology included uterine leiomyomas in four cases and atypical hyperplasia of the endometrium in one. In the latter case (case 1), the atypical hyperplasia was clearly separate from the ESS within the myometrium.

Discussion

We have described a series of eight ESSs containing large numbers of endometrioid glands. It has been claimed that small numbers of endometrioid glands are found in a significant percentage of uterine endometrial stromal neoplasms, either due to entrapment of surface endometrial or adenomyotic glands or secondary to direct production by the neoplasm (epitheliogenesis).[19,20] In one study, glands were found in 40% of ESSs,[19] although in most other series they have been identified with a much lower frequency. However, in our experience the presence of endometrioid glands in endometrial stromal neoplasms is relatively unusual and, judging by the number of cases reported in the literature, the presence of significant numbers of endometrioid glands is rare. There has been a single small series of three primary uterine ESSs with extensive endometrioid glandular differentiation[16] together with a single case report.[17] In one of the cases, the endometrioid glands were present only in the recurrent neoplasm, which involved the vagina and pelvis and which was removed 10 years following hysterectomy for a primary uterine ESS.[16] A probable primary extrauterine ESS with numerous endometrioid glands has also been described.[18] In this case a hysterectomy had been performed some years previously; histological examination was not carried out and so a uterine primary could not be totally excluded.

In some of our cases, glands were present throughout the neoplasm and in others they were a focal, but significant, finding. The stromal element was always predominantly ESS with the characteristic morphology of small bland ovoid to spindle-shaped cells and the typical vascular pattern of small arteriole-like vessels. The presence of large numbers of endometrioid glands resulted in significant diagnostic difficulty in many of the cases. In some, the time interval from first biopsy to diagnosis of malignancy was considerable, in one instance 7 years. One uterine neoplasm was originally misdiagnosed as adenomyosis and several extrauterine cases as endometriosis; in several of the latter cases, especially case 2, the histological features in the original biopsy specimens were virtually indistinguishable from endometriosis and it was only on reviewing these specimens together with the later specimens that an unequivocal diagnosis could have been made. Other diagnostic considerations included adenosarcoma and carcinosarcoma. The differential diagnosis is discussed in detail below. However, we make the point that ESS with endometrioid glandular differentiation should be suspected when repeated biopsies reveal an appearance consistent with endometriosis in the presence of a suspected tumour mass.

Apart from the presence of endometrioid glands, other relatively unusual features included smooth muscle differentiation in one case and sex-cord-like elements in two. Both features are well described in otherwise typical endometrial stromal neoplasms, and it is not unexpected that they should occasionally occur in an ESS with endometrioid glandular differentiation. There was pseudodecidualization of the neoplastic stromal component in one case. This may occur in association with progestogen therapy, but may also occur idiopathically;[21] as far as we are aware, this patient was not taking hormonal preparations. In two cases, the neoplastic stromal cells focally had a clear cell appearance, a feature that has rarely been described in endometrial stromal neoplasms.[22] Foam cells were present in one case. These have been described in endometrial stromal neoplasms and may represent either histiocytes or neoplastic cells or a

combination.[23] Heterotopic bone was present in one case. To the best of our knowledge, this has not been described previously in endometrial stromal neoplasms, although heterotopic ossification occasionally occurs in the stroma of many neoplasms. Scattered atypical nuclei with an appearance reminiscent of symplastic change in a uterine leiomyoma were present in one case; this phenomenon has been described previously in a single case of ESS.[14]

Most extrauterine ESSs are thought to arise from endometriosis,[1,2] and it is possible that some of our cases did so, although unequivocal endometriosis was identified in only two cases. In other cases there were foci which, taken out of context, were virtually indistinguishable from endometriosis but which, given the totality of findings, we consider to represent tumour. There are several reports in the literature of 'aggressive endometriosis';[24,25] we suspect, as postulated by Clement and Scully, that many, if not all, of these represent examples of extrauterine ESS with endometrioid glandular differentiation. One case report we believe illustrates this point, in that a patient with a history of uterine ESS underwent removal of a mass involving an ovary.[25] Histology showed what was interpreted as aggressive endometriosis within large vascular channels; we suspect this represents recurrent ESS with glandular elements confined to the recurrent neoplasm, similar to one of the cases reported by Clement and Scully[16] and to case 7 in this series.

One point that is worthy of note is the possible association between tumour development or growth and the administration of unopposed oestrogens. Unopposed oestrogens should not be given to women with an intact uterus because of the well-known risk of development of endometrial proliferative lesions, including adenocarcinoma. In three of our cases there was a history of unopposed oestrogen intake following hysterectomy. It is possible that this contributed to the development of ESS in cases 4 and 5, possibly from foci of unsuspected endometriosis, and stimulation of the neoplasm in case 1. Unopposed oestrogens have been speculated to be involved in the development of ESS and in malignant transformation of endometriosis.[26,27]

The differential diagnosis depends on the exact morphology and whether the tumour has a uterine or extrauterine location. In uterine cases, adenomyosis is obviously the main diagnostic consideration. Although individual areas may be indistinguishable from adenomyosis, the expansile tongue-like growth pattern rather than the atrophic appearance characteristic of most cases of adenomyosis is a pointer towards ESS. Areas of typical ESS without glands may be a further clue, although in some cases of adenomyosis there are foci consisting entirely or predominantly of stroma, so-called stromal adenomyosis or adenomyosis with sparse glands.[28] The presence of vascular invasion, especially if more than an occasional focus, is suggestive of ESS. Occasional intravascular foci are found in some cases of adenomyosis, but this is rarely a prominent feature.[29] With an extrauterine location, misdiagnosis of endometriosis or ESS arising in endometriosis is likely. Although it could be considered that some of the extrauterine cases represent ESS with areas of endometriosis, we feel these represent ESS with endometrioid glands. We base this on the fact that the stromal component was always obviously expansile and low-grade malignant, sometimes with vascular invasion, even in those areas containing glands. In order to confirm this, it would have been interesting to undertake molecular studies on the various components, since up to 80% of ESS harbour a *t*(11;17) chromosomal translocation involving the *JAZF1* and *JJAZ1* genes.[30]

Another diagnostic consideration in some cases was adenosarcoma or adenosarcoma arising in endometriosis.[31,32] The resemblance to adenosarcoma was greatest in those cases involving the cervix or vagina with a polypoid architecture with or without periglandular increased cellularity. However, in all such cases, there were areas with the typical appearance of ESS and absence of the intraglandular stromal projections characteristic of adenosarcoma, and we consider these to represent ESS with glandular differentiation rather than adenosarcoma. Adenosarcoma may exhibit sarcomatous overgrowth and contain areas devoid of glands. However, the areas of sarcomatous overgrowth are typically high grade, whereas in our cases the pure stromal component was of typical endometrial stromal type. It is possible that some cases reported in the literature as adenosarcoma may represent ESS with endometrioid glandular differentiation, and we admit that in some cases there may be considerable morphological overlap between these two neoplasms.

In the majority of cases, the endometrioid glands, although numerous, were for the most part quite widely separated by the predominant stromal element. Indeed, as discussed, we consider this dichotomy between the number of glands and the expansile stroma to be a clue to the diagnosis of ESS rather than endometriosis or adenomyosis. However, in several cases the endometrioid glands were crowded, at least focally, the morphological features resembling complex hyperplasia within the eutopic endometrium. In one case, there was a back-to-back glandular architecture with nuclear atypia, and focally this amounted to a grade I endometrioid adenocarcinoma. In one of the previously reported ESSs with endometrioid glandular

differentiation, there were foci of grade I endometrioid adenocarcinoma.[16] This admixture of adenocarcinoma and ESS raises the possibility of a carcinosarcoma. However, in carcinosarcoma both the epithelial and mesenchymal elements are typically high grade and the sarcomatous elements derive from the carcinoma, in effect a dedifferentiated carcinoma or carcinoma with sarcomatous metaplasia. In our case, benign endometrioid glands were present away from the foci of carcinoma, excluding a carcinosarcoma.

A variety of immunohistochemical markers were applied as part of the work-up of the individual cases. Most tumours tested exhibited the typical immunophenotype of endometrial stromal neoplasms with diffuse positivity for CD10, ER and PR.[33] However, immunohistochemistry contributes little to the diagnosis since the mesenchymal elements are obviously of endometrial stromal type and the stromal component of those lesions in the differential diagnosis would also typically be positive with these markers.

There is no evidence that the behaviour of ESS with endometrioid glands is any different from those neoplasms without glands. We have follow-up in only a few cases, and no patient has died of disease. However, two patients have persistent tumour following resection. These patients were treated with postoperative radiotherapy and gonadotropin-releasing hormone agonists in one case and progestogens in the other; these may be of value in management of residual tumour or in an adjuvant setting. One patient was treated with chemotherapy and is alive and well 11 years following diagnosis.

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