Extensive psammomatous calcification of the uterus and cervix associated with a uterine serous carcinoma

R I Cameron, W G McCluggage

This report describes a uterine serous carcinoma with bilateral ovarian metastasis, which was associated with widespread extensive psammomatous calcification of the uterine leiomyomata, the myometrium, and the cervical stroma. These psammoma bodies were not associated with tumour or epithelial elements. This psammomatous calcification is rare, with no previous reports of similar cases. The presence of psammoma bodies is probably related to the serous carcinoma, raising the possibility that psammoma body formation in serous carcinomas is the result of a factor secreted locally by the tumour, rather than the widely held theory that their formation is secondary to necrosis, with subsequent dystrophic calcification within a papillary neoplasm.

Psammoma bodies are well circumscribed, laminated, calcified structures that can be seen in a range of conditions. They are especially common in the female genital tract, and are often associated with serous type neoplasms, which can be benign, borderline, or malignant. In some serous tumours, they are abundant and the term psammocarcinoma has been used for a highly differentiated serous carcinoma of the ovary or peritoneum associated with massive psammoma body formation. Psammoma bodies may also be associated with benign, non-neoplastic proliferations of serous type epithelium, termed endosalpingiosis. Occasionally, especially in peritoneal lined structures, psammoma bodies are found without an associated epithelial component, and it has been speculated that this represents “burnt out” or atrophic endosalpingiosis.

“We describe an unusual phenomenon of extensive psammoma body formation within uterine leiomyomas, the myometrium, and the cervical stroma”

Rarely, psammoma bodies are seen in the endometrium or cervical mucosa, sometimes in association with a serous proliferation elsewhere within the female genital tract. In this report, we describe an unusual phenomenon of extensive psammoma body formation within uterine leiomyomas, the myometrium, and the cervical stroma. These psammoma bodies were not associated with tumour or epithelial elements, but a small uterine serous carcinoma was present, which had spread to both ovaries. We speculate on the possible pathogenesis of psammoma body formation in this case.

CASE REPORT
A 56 year old woman presented with abdominal pain, nausea, and vomiting. Clinical examination revealed an acute abdomen and an emergency exploratory laparotomy was performed. The uterus was found to be perforated and adherent to the descending colon. There was extensive ischaemic change of the colon and small intestine. A hysterectomy and bilateral salpingoophorectomy together with omentectomy, colectomy, and small intestinal resection was performed.

The uterus weighed 380 g and was perforated anteriorly. On sectioning, multiple fibroids were identified, the largest measuring 5 cm in diameter. No obvious endometrial or myometrial tumour was seen grossly. Both ovaries measured 4 cm in maximum diameter. Microscopically, the right ovary was entirely replaced by poorly differentiated serous carcinoma. Occasionally, especially in peritoneal lined structures, psammoma bodies are found without an associated epithelial component, and it has been speculated that this represents “burnt out” or atrophic endosalpingiosis.

Figure 1 Poorly differentiated serous carcinoma involving the right ovary.

Abbreviations: CK, cytokeratin; WT1, Wilms’s tumour gene product
An additional unusual feature was the presence of numerous psammoma bodies throughout the leiomyomata (fig 3), the myometrium, and the cervical stroma, just beneath the surface epithelium (fig 4), and also deep within the stroma (fig 5). These formed aggregates and were associated with hyalinised fibrous tissue (fig 3). They were not associated with tumour or benign epithelial elements.

**DISCUSSION**

Psammoma bodies are characteristically associated with non-neoplastic serous proliferations and with serous neoplasms, which may be benign, borderline, or malignant. They are thought to arise secondary to necrosis, with subsequent dystrophic calcification of the tips of papillary structures. In our case, the presence of numerous psammoma bodies throughout the uterine leiomyomata, the myometrium, and the cervical stroma was extremely unusual and we are not aware of previous reports of similar cases. A serous carcinoma involved the endometrium and had metastasised to both ovaries, without evidence of myometrial involvement, except for tumour on the serosal surface of the uterus. The psammoma bodies in the myometrium and the cervix were not associated with epithelial elements, but we think that they are probably related in some way to the uterine serous carcinoma. One possibility is that they are the remnants of previous tumour within the myometrium and cervix, but that the tumour here has spontaneously regressed. However, we feel that this is extremely unlikely, and think that the psammomatous calcification is probably secondary to a local factor secreted by the tumour cells. It is not uncommon to find psammoma bodies without epithelial elements in the omentum or peritoneum in association with a serous tumour of the ovary. In this situation, it can also be postulated that the psammoma body formation is secondary to secretion of a factor by the tumour cells.

The presence of serous carcinoma within the endometrium and both ovaries raises questions regarding the origin of the tumour; that is, whether this represents a primary uterine serous carcinoma with bilateral ovarian metastasis or a primary ovarian carcinoma with endometrial metastasis. A further possibility is independent synchronous primaries within the endometrium and ovaries. We think that it is most likely that this represents a primary uterine neoplasm, but we cannot exclude the other possibilities. A pointer towards a uterine primary is that there was no staining of tumour cell nuclei with WT1. WT1 positivity is usual in ovarian serous carcinomas but most uterine serous carcinomas are negative. Moreover, there was no evidence of omental involvement by tumour, which is usual when a serous carcinoma spreads beyond the ovary.
Take home messages

- We describe an unusual phenomenon of massive psammoma body formation, not associated with epithelial elements, throughout the uterine leiomyoma, the myometrium, and the cervical stroma.
- This was associated with a uterine serous carcinoma that had metastasised to both ovaries.
- This appears to be the first report of this phenomenon.
- We speculate that psammoma body formation was secondary to a local factor secreted by the tumour.

In summary, we describe an unusual phenomenon of massive psammoma body formation, not associated with epithelial elements, throughout the uterine leiomyoma, the myometrium, and the cervical stroma. This was associated with a uterine serous carcinoma that had metastasised to both ovaries. In a search of the literature, we could find no previous reports of this phenomenon. We speculate that psammoma body formation in our case is secondary to a local factor secreted by the tumour.

Authors’ affiliations

R I Cameron, W G McCluggage, Department of Pathology, Royal Group of Hospitals Trust, Belfast BT12 6LB, Northern Ireland

Correspondence to: Dr W G McCluggage, Department of Pathology, Royal Group of Hospitals Trust, Grosvenor Road, Belfast BT12 6LB, Northern Ireland; glenn.mccluggage@bhll.ni.nhs.uk

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