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Ovarian lipoleiomyoma: a rare benign tumour

There are only two published case reports of lipoleiomyoma in the English literature. We report a case of lipoleiomyoma of the ovary in a woman who was in her fifties. She presented with a gradually progressive abdominal mass of 8 months duration. Abdominal imaging showed a large mass that was fed by right ovarian pedicle. Right ovary was not visualised. The excised mass was nodular and encapsulated; the cut section was yellowish with scant grey-white whorled areas. Microsections showed a mature adipose tissue admixed with leiomyomatous areas without cellular atypia and adjacent compressed ovarian tissue. Immunohistochemical staining showed diffuse positive reaction for desmin confirming that spindle cells were smooth muscle cells. A diagnosis of lipoleiomyoma of the ovary was made based on the radiological, histopathological and immunohistochemical findings.

Primary leiomyoma is a rare tumour of the ovary.¹ Lipoleiomyoma of the ovary is even more uncommon. There are only two published case reports of lipoleiomyoma of the ovary in the English language literature.^{2,3} We report here a case of primary lipoleiomyoma of the ovary because of its rarity. It can also be occasionally misdiagnosed as sarcoma on clinical and radiological examination.

Case history

A woman in her fifties presented with a gradually progressive abdominal mass of 8 months duration. Abdominal CT and ultrasound scan revealed a large mass suspected to be a liposarcoma. The CT scan demonstrated a huge fat density in the right adnexa with few solid enhancing components and minimal fluid. The right ovary was not visualised separately but the right ovarian pedicle was feeding the mass. The ultrasound scan showed a large intraperitoneal echogenic mass with hypoechoic areas and fluid pockets. The uterus was normal in size and had no fibroids in both the scans.

Figure 1 A cut section of the tumour shows yellow and grey-white whorled areas.

Figure 2 Tumour composed of predominantly smooth muscle cells in fascicles and adipose tissue (H&E ×100).

Histopathological findings

The mass weighed 4.5 kg and comprised of four well-encapsulated nodules attached to one another. Three nodules were large and each measured approximately 18×15×12 cm. A fourth central small grey-white nodular mass measured 4×2×2 cm. Cut sections of the two large nodules showed uniform yellow lobules; the third large nodule was also yellowish but had a central grey-white whorled mass that measured 6 cm across (fig 1). The cut section of the small nodule had a whorled appearance. One of the nodules had a pearly white area on the external surface suggestive of a compressed ovary.

Multiple representative sections from the smaller central nodule and the grey-white area in one of the larger nodules showed a tumour comprising predominantly smooth muscle cells arranged in interlacing fascicles admixed with small foci of mature adipose cells (fig 2). The sections from the large yellowish masses showed extensive mature adipose tissue in the form of lobules separated by thin fibrovascular septae. Intimately admixed with these adipose lobules were multiple leiomyomatous areas (fig 3). Stroma showed scattered lymphocytes, plasma cells and eosinophils. Also seen were focal areas of hyaline degeneration. Sections from the pearly white area showed compressed ovarian stroma with few adipose cells. To confirm that spindle cells were smooth muscle cells, immunohistochemistry was performed using desmin, which showed a diffuse positive reaction (fig 4).

Discussion

Primary leiomyoma of the ovary is uncommon and they account for about 1% of the benign neoplasms.⁴ Lipoleiomyoma is even rarer. To the best of our knowledge, this is the third case reported.

We made a diagnosis of lipoleiomyoma of the ovary based on the presence of intimately

Figure 3 Tumour showing predominantly mature adipose tissue admixed with smooth muscle cells (H&E ×100).

Figure 4 Immunohistochemical staining for desmin showing diffuse positive reaction (×100).

Take-home messages

- Lipoleiomyoma is a very rare benign tumour of the ovary.
- The tumour is identified by the presence of mature adipose tissue intimately admixed with smooth muscle cells.
- Lipoleiomyoma may mimic sarcoma clinically.

admixed mature fat and smooth muscle cells. It was considered to be of ovarian origin based on 1) the presence of ovarian tissue closely adherent to the excised mass, 2) presence of adipose tissue within the ovary and 3) CT scan showing the right ovarian pedicle feeding the mass and non-visualisation of the right ovary.

Microscopic features are similar to leiomyoma, but in addition they have mature adipose tissue. The amount of fat may range from being a minor component in what is otherwise leiomyoma to being a neoplasm composed entirely of mature adipocytes.⁵ Primary ovarian leiomyoma most likely originates from smooth muscle cells in the walls of the arterial vessels within the cortical stroma, corpus luteum and in the ovarian ligaments at their point of attachment to the ovary.⁶ Its precise histogenesis is, however, uncertain.⁷ The theories on histogenesis of adipose tissue are unresolved even in the case of the uterus where these tumours are relatively more common than in the ovary. In uterine origin, they most probably represent tumour metaplasia within a leiomyoma.^{5,8} In ovary too, presence of adipose tissue might represent metaplasia.

Fatty tumours of the ovary are very rare and almost invariably benign.⁴ The tumours are usually found in menopausal and post-menopausal women. Clinically, the tumours are asymptomatic and discovered either incidentally or when they attain huge sizes with attendant complications. The only puzzle for the pathologists could be the unexpected presence of fat in the ovary. Occasionally, the clinical presentation and macroscopic appearance may mimic sarcoma, as in this case, and create diagnostic confusion.³ Primary leiomyoma must also be differentiated from pedunculated subserosal parasitic uterine leiomyoma, which has lost its attachment from the uterus and is instead attached to the ovary.⁹ Lastly, it should be differentiated from ovarian fibroma, which is more common.⁹

M Hemalata, V Kusuma, P Sruthi

Department of Pathology, Kempegowda Institute of Medical Sciences (KIMS), VV Puram, Bangalore 560004, India

Correspondence to: M Hemalata, 44, Type 5, Block 2, BRC Campus, NIMHANS Quarters (Dairy Circle), Bangalore 560029, India; ycjreddy@yahoo.com

doi: 10.1136/jcp.2007.047183

Accepted 5 March 2007

Competing interests: None.

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Ectopic lobular breast cancer on the anterior chest wall: a rare entity

A 46-year-old woman with a rare case of infiltrating lobular carcinoma in ectopic breast tissue on the anterior chest wall is presented. Wide local excision and lymphatic mapping with, consequently, a sentinel node biopsy, seem adequate therapy for ectopic breast cancer.

Ectopic breast tissue arises from the primitive milk line, with an incidence of 0.6–6%.¹ Malignant transformation is rare.² The most frequently occurring manifestation is infiltrating ductal carcinoma, comprising 79% of all ectopic breast malignancies.³ Lobular-type and medullary-type carcinomas are seen in <10% of cases.⁴ Furthermore, most carcinomas in the ectopic breast tissue are located in the vulva and axilla (ie, 55–65%).⁵ The chest wall is the most unusual location.⁶

We present a patient with lobular carcinoma in ectopic breast tissue located on the anterior chest wall.

Case report

A 46-year-old Caucasian woman presented with a subcutaneous mass in a cicatrice of the anterior chest wall approximately 1–2 cm below the caudal rim of the mammary gland. At that location, she had undergone surgical excision of ectopic breast tissue without signs of malignancy, 22 years ago. She had noticed the present swelling 2 weeks previously, without further symptoms. She was premenopausal and had no family history of breast cancer.

Figure 1 Scar on the anterior chest wall showing a firm subcutaneous mass of approximately 4 mm diameter (arrow).

Clinical examination and x rays of both breasts and axillae were found to be normal. Clinical examination of the scar on the anterior chest wall showed a firm subcutaneous mass of approximately 4 mm in diameter (fig 1). Ultrasonography of the mass showed only scar tissue.

Excisional biopsy was performed, and pathology showed a pleomorphic lobular carcinoma in situ arising from ectopic breast parenchyma. The cells of the pleomorphic lobular carcinoma form a “single file” pattern (ie, aligned in a row) (fig 2). Immunostaining for E-cadherin was negative. The tumour was radically excised, and showed a mitotic activity index of 13 per 2 mm², Bloom Richardson grade III, negative Her-2-neu receptor status and positive oestrogen and progesterone receptors.

Thus, re-excision of the accessory breast tissue tumour was performed after lymphatic mapping and sentinel node biopsy (fig 3). One sentinel node was identified in the ipsilateral axilla.

Final pathology showed mammary gland tissue with remnants of the aforementioned lobular carcinoma, now with negative margins. Pathology of the sentinel node biopsy showed metastasis with extranodal growth. Therefore, an axillary lymph node dissection was performed. Final pathology showed no metastases in the second echelon axillary lymph nodes.

To exclude a primary breast cancer, MRI of the mammary glands and axillary regions was

Figure 2 Pleomorphic lobular carcinoma cells forming single rows, commonly termed “Indian file”.

Figure 3 Lymphatic mapping: one sentinel node was identified in the ipsilateral axilla (arrow).

performed, showing no pathological features. The patient received adjuvant radiotherapy of the chest wall, chemotherapy (5-fluorouracil, epirubicin and cyclophosphamide), followed by hormonal treatment.

Discussion

In a review study on all published cases of ectopic breast cancer from 1865 to 1994, only three cases of ectopic breast cancer arising in the chest wall were found.⁶ Combining this with the fact that lobular carcinoma in ectopic breast cancer is seen in <10% of cases makes our patient with infiltrating lobular carcinoma in ectopic breast tissue on the anterior chest wall an exceedingly rare case.⁵

In a study on 90 cases of carcinoma in ectopic breast tissue, radical or modified mastectomy offered no advantage in outcome over wide local excision combined with regional node dissection.⁷ In our patient, local excision showed radically excised infiltrating lobular carcinoma, therefore wide local re-excision was performed with negative margins.

Lymph node status is an important prognostic factor in breast cancer.⁸ Some authors have even reported an increased incidence of metastasis to regional lymph nodes with earlier involvement in ectopic breast cancer compared with standard breast cancer.^{9–10} However, the anatomy of lymphatic drainage of the ectopic breast is unclear. The use of lymphatic mapping may show these lymphatic pathways. In a recent study, the sentinel node technique was successful in isolating an unusual group of lymph nodes. This allowed more accurate staging of the disease and more appropriate adjuvant treatment.¹¹ In our patient, the sentinel node was located in the axilla and was positive for metastases. Completion axillary lymph node dissection was therefore performed.

In summary, we present a case of infiltrating lobular carcinoma in ectopic breast tissue on the anterior chest wall. As ectopic breast cancer is a rare entity and literature on the subject is sparse, drawing conclusions about the appropriate treatment is daunting. However, combining the data from our case with previous studies, wide local excision and lymphatic mapping with, consequently, a sentinel node