

CASE REPORT

Single benign metastasising leiomyoma of an inguinal lymph node

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Accepted 26 July 2016

SUMMARY

Benign metastasising leiomyoma (BML) is a rare benign disease associated with uterine leiomyoma and history of uterine surgery. It most frequently occurs in premenopausal woman, with a pulmonary localisation, and consisting of multiple nodules. We present an uncommon case of a 69-year-old woman with a single BML of an inguinal lymph node. CT scans of thorax and abdomen excluded other metastasis localisation. The patient was cured with surgical excision of the mass. Lymph node involvement has been reported incidentally in BML literature. Lymphangitic spread can be considered a possible mechanism of BML metastasis.

BACKGROUND

Benign metastasising leiomyoma (BML) is a rare condition characterised by histological benign proliferation of smooth muscle cells, metastasising with a haematological spread.^{1 2} It has been associated with a history of uterine leiomyoma, and many patients presenting with BML symptoms have previously undergone hysterectomy or other lower abdominal surgery.^{3 4} BML not only occurs predominantly in women of reproductive age but can also occur in postmenopausal. In the majority of cases, the disease consists of multiple nodules in the lungs, although other locations, such as the abdomen, soft tissue, skeletal muscles and lymph nodes, have been reported.³⁻⁶ Single case reports of metastases in skin, bone and heart have also been reported.^{4 7 8} Lesions are usually asymptomatic and most nodules are discovered using routine imaging. In this report, we describe an extraordinary case of a single metastasis of BML in an inguinal lymph node. We also provide a literature overview of cases of BML metastases to lymph nodes.

CASE PRESENTATION

A 69-year-old woman presented with a painless subcutaneous mass in her left groin. The mass had slowly been growing over the course of 2 years. An enlarged lymph node was suspected. Her medical history consisted of an appendectomy at early age, a retroverted uterus correction at the age of 32 and uterine leiomyoma diagnosed at the age of 46, which was left untreated.

INVESTIGATIONS

On physical examination, a solid mass was found in the left groin, with a size of ~20 mm×20 mm. Ultrasound-guided fine needle aspiration detected the presence of non-malignant leiomyoma, with

positive desmins and actin. There was no mitotic activity and no cytonuclear atypia. To confirm the diagnosis, complete lymph node excision was performed. Histological analysis showed a lymph node with the presence of spindle-shaped cells that resembled the characteristics of those found in the fine needle aspiration (figure 1). EMA, S-100 and CD-117 staining were negative. After the diagnosis was confirmed, CT of thorax and abdomen was performed to investigate other metastases. This scan revealed no other suspected lesions.

DIFFERENTIAL DIAGNOSIS

Differential diagnosis of inguinal masses includes:

- ▶ reactive lymph node,
- ▶ lipoma,
- ▶ malignant metastasis,
- ▶ primary lymph node malignancy (eg, lymphoma),
- ▶ hernia (femoral, inguinal),
- ▶ femoral aneurysm,
- ▶ hydrocele of the canal of Nuck.

TREATMENT

With the surgical excision of the lymph node, the patient's symptoms disappeared and other treatments were deferred.

OUTCOME AND FOLLOW-UP

The patient has not returned with recurrence or formation of BML in other locations postoperatively. We advised her to return to clinics if

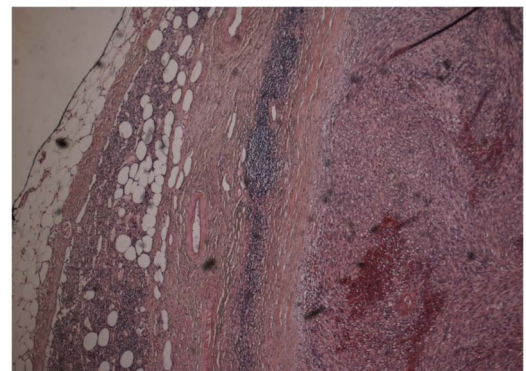


Figure 1 Histopathological examination. The lymph node is almost completely effaced by the lesion with just a small marginal rest of lymphoid cells. The lesion consists of non-atypical smooth muscle cells (H&E stain, ×200).



To cite: Laban KG, Tobon-Morales RE, Hodge JAL, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-216546

Table 1 Review of cases with lymph node benign metastasising leiomyoma

Author	Year	Age	Localisation	Amount	Treatment	Other localisation
Andreeva <i>et al</i> ¹⁰	2012	54	Retroperitoneal	–	–	
Yoon <i>et al</i> ¹¹	2011	34	Retroperitoneal	Multiple	Hysterectomy, resection and aromatase inhibitor	Lung
Tori <i>et al</i> ¹²	2008	47	Retroperitoneal	Multiple	GnRH agonist	Skeletal muscle (biceps)
Obana <i>et al</i> ¹³	1996	39	Retroperitoneal	Multiple	–	
Barter <i>et al</i> ¹⁴	1987	46	Retroperitoneal	Multiple	Resection+chemotherapy+radiation treatment	
Horie <i>et al</i> ¹⁵	1984	50	Retroperitoneal	Single	Hysterectomy+resection	LPD
Rigaud and Bogomoletz ¹⁶	1983	57	Retroperitoneal	Multiple	Hysterectomy+hormonal ablation+resection	
Hsu <i>et al</i> ¹⁷	1981	40	Retroperitoneal	Multiple	Hysterectomy+resection	LPD
Deppe <i>et al</i> ¹⁸	1980	32	Retroperitoneal	Multiple	Hysterectomy+resection	
Abell and Littler ¹⁹	1975	27	Retroperitoneal	Multiple	Hysterectomy+resection	

LPD, leiomyoma peritonealis disseminata.

symptoms recurred or new masses were noticed. No further routine follow-up was advised.

DISCUSSION

BML usually occurs in premenopausal women with a history of uterine leiomyoma and lower abdominal surgery, and mainly consists of multiple nodules affecting the lungs. To the best of our knowledge, our case is the first case of BML in an inguinal lymph node. Only a few cases describe single metastasis, and the occurrence of BML in postmenopausal women is rare.⁹

We conducted a MEDLINE literature search, covering the period from 1960 up to 2016 for lymph node metastasis of BML. In table 1, articles describing cases of lymph node BML are presented.^{10–19}

Lymph node involvement is reported retroperitoneal. Only one case report found a single lymph node at presentation.¹⁵ In the cases reporting lymph node involvement, BML was found in lungs, skeletal muscles, chest and abdomen. Leiomyoma peritonealis disseminata has also been identified to coexist with lymph node manifestation.^{15 17}

The pathogenesis of BML is still largely unknown. Previous reports have described a mainly haematological spread of metastasis.² However, our case, as well as several previous case reports, demonstrate the involvement of the lymphatic system in the disease.^{10–19} In several cases, there was no evidence of haematological spread, as most metastases were only present in lymph nodes. Therefore, lymphangitic spread could be a plausible explanation of metastasis. Clinical implications, including prognosis and specific treatment, require further investigation. CT should be performed to exclude other nodules in thorax and abdomen, since BML mostly occurs with multiple nodules.⁶ Other localisations, such as lymph nodes, should be investigated with clinical suspicion of tumour metastasis.

There is no standard treatment for BML. Possible treatment strategies include expectative follow-up, surgical resection, hormonal ablation (oophorectomy) and hormonal medication (GnRH agonist, aromatase inhibitors, selective oestrogen receptor modulators and progesterone).^{1 2} Because of hormonal association, postmenopausal patients with BML should be treated with either expectative follow-up or primary surgical resection.²⁰

We present the first case of a single BML located in a lymph node in the groin. Lymph node localisation has previously been described to be located mainly retroperitoneal. This case and previous case reports of lymph node BML, without other

metastases localisations, indicate the possibility of the lymphangitic spread of the disease.

Learning points

- ▶ Single benign metastasising leiomyoma is extremely rare.
- ▶ Benign metastasising leiomyoma can occur in inguinal lymph nodes.
- ▶ Lymphangitic spread is a possible route of metastasis.

Contributors This case report was designed by KGL and RET-M, The manuscript was written by KGL, RET-M and HWRS. The pathological images and pathological sections of the manuscript were provided and written by JALH.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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