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A case of benign cystic mesothelioma arising from the lesser omentum in a 13-year-old girl treated through a laparoscope excision

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Abstract

Benign cystic mesotheliomas (BCMs) are extremely rare tumors usually occurring in females of reproductive age. A 13-year-old Japanese girl presented with abdominal pain, and an adnexal cystic tumor was suggested by images. At laparoscopy, the cystic mass was found arising from the lesser omentum. Histological examination and calretinin-immunohistochemistry revealed the lesion as a BCM. Cytology was initially useful for the diagnosis. This is a benign pathology, but recurrences are not uncommon. BCM should be included in the differential diagnosis, when investigating pelvic and abdominal masses associated with adnexal tumors in women.

Keywords: Benign cystic mesothelioma, omental tumor, cytology, calretinin

Background

Benign cystic mesotheliomas (BCMs) are rare abdominal tumors usually occurring in women of reproductive age [1,2]. Approximate 140 cases have been reported to date [3,4]. Surgical removal is the main form of treatment. Follow-up is important because of the high rate of recurrence [5,6] and malignant transformation [7,8]. We present a rare case of BCM developed in a 13-year-old girl treated through a laparoscope excision.

Case presentation

A 13-year-old Japanese girl complained of abdominal discomfort and visited to a local doctor. Menarche was 12-year-old. She had no surgical history, and her medical history was not significant. She denied sexual intercourses, and was not taking regular medications and had no allergies. Plain computed tomography (CT) revealed a huge pelvic mass, and she referred to the Department of Obstetrics & Gynecology, Gujo City Hospital. While a trans-rectal ultra-sonography showed a uterus without any changes and normal sized ovaries, magnetic resonance imaging (MRI) revealed a huge pelvic mass: the lesion was a single cyst, measuring 16 × 6 × 13 cm (**Figure 1**). Clinical laboratory data, including tumor markers (CA125 17.7 U/ml, CA19-9 9.4 U/ml, CEA 0.5 ng/ml) showed within normal limitations.

Under a working diagnosis of a right para-tubal cyst, she underwent a laparoscopic surgery one month after her first visit. A huge cystic tumor occupied the abdomen, and the content, serous fluid (830 ml), was aspirated from the cyst for cytological diagnosis. The cystic tumor was originated from the lesser omentum, and twisted (**Figure 2**). Although cellularity on the cytological specimen was few, we detected activated mesothelial cells (**Figure 3**). After the release of torsion, the cystic tumor was removed. The cystic tumor was consisted of monolobular cyst (**Figure 4**). The lesion was fixed in 4% buffered formalin, routinely processed, and embedded in paraffin. After which 3-4 μm-thick sections were stained with haematoxylin and eosin for pathological diagnosis. In addition, immunohistochemistry using Ventana monoclonal antibodies, such as calretinin (1:2000 dilution), CD31 (1:100

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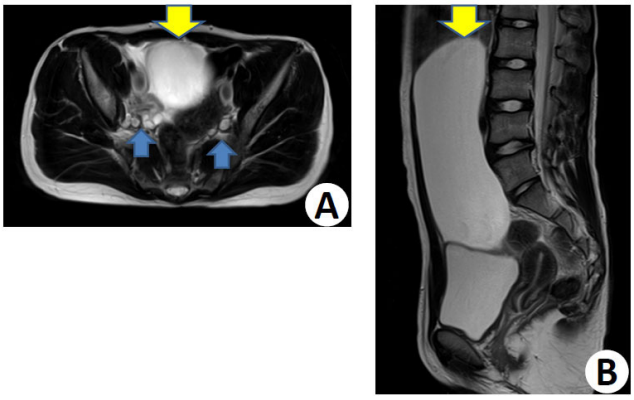

Figure 1. (**A**) T2-weighted MRI. Cystic lesion is present (yellow allow) and normal ovaries are observed (blue allows). (**B**) T1-weighted MRI. A huge cystic lesion is present.

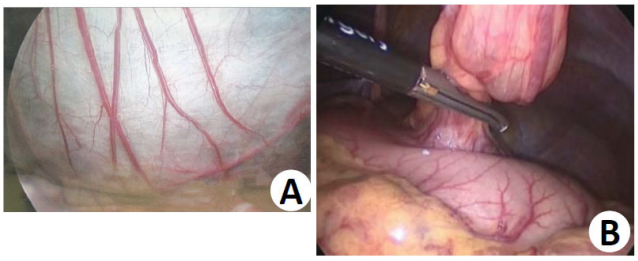

Figure 2. (**A**) By a laparoscopic view, a huge cystic lesion is present. (**B**) The lesion arises from the lesser omentum after aspiration of the fluid, and twisted.

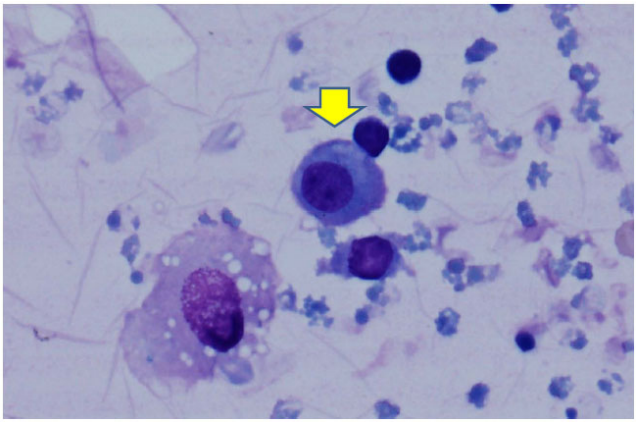

Figure 3. The cytology showed activated mesothelial cells and histiocyte (May-Geimza stain, x 400).

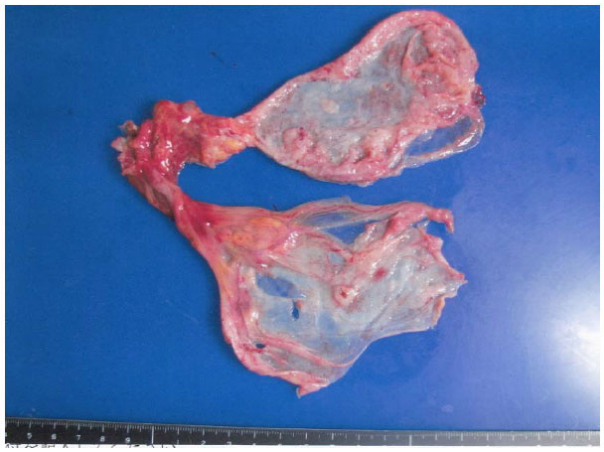

Figure 4. The resected tumor is monocystic, and papillary growth cannot be observed.

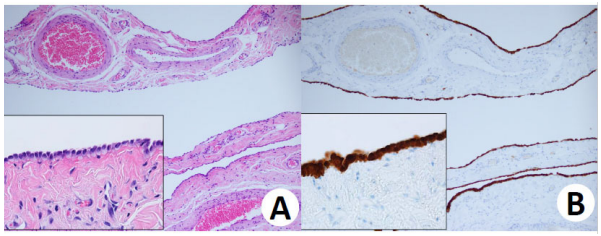

Figure 5. (**A**) The cystic spaces were lined by flattened endothelial-like cells. (H & E, x20) Inset, Cystic walls are lined by a single layer of cells. (x100).
(**B**) Strong immunoreactivity of the lining cells with calretinin (x20). Inset (x100).

dilution), CD34 (1:50 dilution), estrogen receptor (ER, 1:100 dilution), and progesterone receptor (PR, 1:100 dilution) was performed to determine the type of cells that covered the cyst wall. Monolayer cells forming cystic walls (**Figure 5A**) were immunohistochemically positive for calretinin (**Figure 5B**), but negative for CD31, CD34, ER, and PR. Cellular atypia was not found. Thus, the patient was diagnosed as a BCM arising from the lesser omentum. The recurrence was not detected after two years and 6 months after the surgery.

Discussion

BCM is a rare medical entity and was first described in 1979 by Mennemeyer and Smith [9]. Approximate 140 cases have been reported currently [3,4]. BCM occurs predominately in females of reproductive age [1,2], suggesting possibility of an estrogen exposure. No relationship has been found between BCM and asbestos exposure, unlike pleural mesothelioma [10].

As to pathogenesis of BCM, neoplastic or reactive change has been postulated [5,10-13]. Several common features, such as history of previous surgery, endometriosis, uterine leiomyoma, and inflammation, were reported in the BCM patients, suggesting that the lesion results from reactive changes to chronic irritation. In contrast, the neoplastic theory is based on features, such as the slow but progressive growth of the lesion when untreated, a few cases of recurrence of the lesion and low incidence of previous infection [6,10], as was our case.

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Pre-operative fine-needle aspiration biopsy or cytology of cystic lesions has been reported to help to formulate accurate and differential diagnosis [13-15]. However, immunohistochemical and electron microscopic investigations were recommended to confirm the diagnosis [16]. In our case, the cytology from the fluid during the operation revealed activated mesothelial cells, suggesting the cystic tumor as BCM. The differential diagnoses should be carefully done from several benign and malignant lesions, which present as cystic abdominal masses. Benign lesions include cystic lymphangioma [10,17], cystic adenomatoid tumors [18]. Since cells lined the cyst wall of BCM are positive for calretinin and negative for endothelial markers as CD31, CD34 and Factor-VIII, immunohistochemistry is useful for differenrtial diagnosis. Cystic lymphangioma is more common in young males rather than females: the cystic spaces are lined by a single layer of flattened endothelial cells, which are immunoreactive against vascular antibodies, such as CD31 and CD34 [19,20]. In a few cases of BCM, nuclei of the activated mesothelial cells lined along the cyst wall were positive for ER and PR [19,21], our case was negative against both antibodies.

Conclusion

An extremely rare case of BCM arising from a lesser omentum in a 13-year-old girl is presented with the histopathological, cytological, and immunohistochemical findings, which were useful for the diagnosis. This is a benign pathology, but recurrences are not uncommon. BCM should be included in the differential diagnosis, when investigating pelvic and abdominal masses associated with adnexal tumors in women.

Competing interests

The authors declare that they have no competing interests.

Authors' contributions

Authors' contributions	KN	RY	YU	MT	SM	YY	TI	TT
Research concept and design	✓	--	--	--	--	--	--	--
Collection and/or assembly of data	✓	✓	✓	✓	✓	✓	✓	--
Data analysis and interpretation	✓	✓	✓	✓	✓	✓	✓	--
Writing the article	✓	--	--	--	--	--	--	--
Critical revision of the article	✓	--	--	--	--	--	--	✓
Final approval of article	✓	✓	✓	✓	✓	✓	✓	✓

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