Case Report

Bursitis with pseudosarcomatous fibroblastic proliferation; a case report

Chaipak Sithinatesakul, MD*., Vorachai Sirikulchayanonta, MD**

*Department of Surgery, Ramathibodi Hospital, Mahidol University, Bangkok, Thailand. ** Department of Pathology, Ramathibodi Hospital, Mahidol University, Bangkok, Thailand Received: 12 May 2014; Accepted 2 June 2014

ABSTRACT

Pseudosarcomatous fibroblastic proliferation (PSFP) is a quite common lesion found in various locations including soft tissue, bone and other organs and often raises a difficult differential diagnosis with malignancy. However, PSFP has not been described in tissue related to bursitis. We report a case of 31-year-old male who developed a cystic lesion in association with a bursitis. Histologically, this showed the presence of sheets of pleomorphic cells with mitosis and mimicked myxofibrosarcoma and angiosarcoma. The findings of ganglionlike cells, metaplastic bone formation and the clinical history of a short incubation period supported the diagnosis of pseudosarcomatous lesion. Consideration of PSFP as a mimic of soft tissue tumours is important for avoiding the pitfall of a malignant diagnosis.

Key words: Bursitis, pseudosarcomatous fibroblastic proliferation, PSFP

INTRODUCTION

Pseudosarcomatous fibroblastic proliferation (PSFP) usually mimics histological malignancy and creates one of the most common pitfalls in differential diagnosis of soft tissue lesions. This lesion has been reported in various organs but occurs most commonly in soft tissue. Such lesions are recognized under different names according the anatomic and clinical features, including nodular fasciitis, proliferative fasciitis and myositis, intravascular fasciitis, cranial fasciitis, postoperative spindle cell tumor (usually occurs in the genitourinary tract), inflammatory pseudotumor and atypical decubital fasciitis¹. Most PSFP lesions do not require aggressive treatment and some of them are self-limiting^{2,3}. Cases of frank malignancy have been reported developing in association with bursitis⁴. It is therefore important to be aware of this lesion, which can mimic malignancy in histologic appearance, and may lead to unnecessary aggressive treatment.

CASE REPORT

A 31-year-old male computer programmer, developed a mass beneath a bed-sore on the left buttock over the course of 5 months. Twenty years ago, he had a complete spinal cord injury due to a motor-cycle accident, causing fracture of thoracic spine at level T5-6. Owing to the fact he spent most of his daily life on wheel chair, he developed decubitus ulcer at left buttock sixteen years later and was admitted for debridement and muscle flap surgery. The ulcer had been persistent since then for almost four years. From five months prior to this admission, a bulging mass beneath the ulcer was noticed and prompted this admission.

Physical examination revealed a bed-sore ulcer on the left buttock, grade 4, with an associated bulging mass underneath measuring 4 cm. in diameter (fig. 1). The clinical diagnosis was bed-sore ulcer with chronic bursitis. Resection of the bursa and overlying skin-ulcer was performed following with muscle flap surgery. The removed specimen was a cystic mass measuring 4 cm. in diameter surrounded with fatty tissue. The inner surface showed bluish pink polypoid appearance (fig. 2). Microscopically, the overlying skin showed an ulcer-bed lined with granulation tissue (fig. 3). The underlying bursa cavity was lined with polymorphonuclear cell-rich fibrinous exudate and granulation tissue was present in the edematous fibrous stroma of the cyst wall (fig.4). Focal small foci of aggregated

large pleomorphic cells were seen, with enlarged vesicular nuclei, arranged in loosely packed sheets and admixed with extensive inflammatory cell infiltrates including polymorphonuclear leucocytes, lymphocytes, plasma cells and few eosinophils (figs. 5&6). Some foci revealed pleomorphic cells clinging along sinusoidal spaces (fig. 7). There were 4-5 mitotic figures per 10 high power-fields but no atypical mitoses were seen. Cells with vesicular nuclei, prominent nuclei and basophilic cytoplasm resembling ganglion cells seen were recognized (fig. 8). Adjacent to the edge of bursa several areas of metaplastic trabecular bone formation were seen, with no malignant osteoid identified (fig. 9). After surgery, the wound healed satisfactorily and no complication were seen during six months of follow-up.



Figure 1 the left buttock revealed a chronic ulcer with reddish granulation tissue at the center.

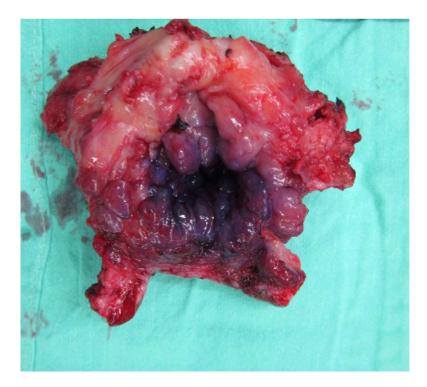


Figure 2 The removed bursa-cyst showed a cyst covering with fatty tissue; the inner surface of the cyst showed bluish red polypoid appearance.

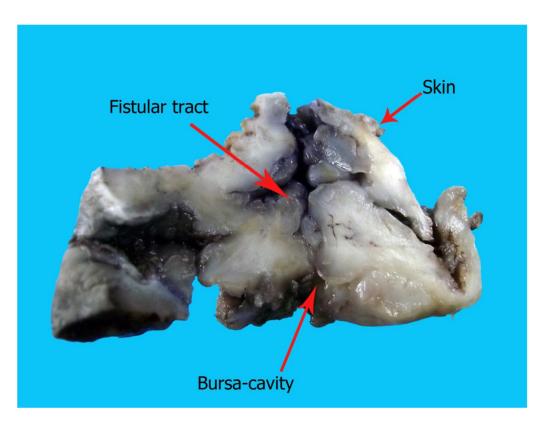
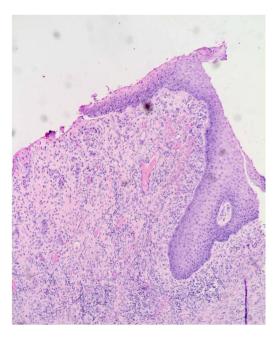


Figure 3 The cut surfaces of the formalin-fixed specimen revealed fistula-tract communicating between skin ulcer and underlying bursa-cavity.



Figur 4 (Hematoxylin & Eosin: original magnification x40) Photomicrograph of the skin-ulcer showed ulcerated squamous epithelium; the ulcer-bed is made up of granulation tissue moderately infiltrated with acute and chronic inflammatory cells.

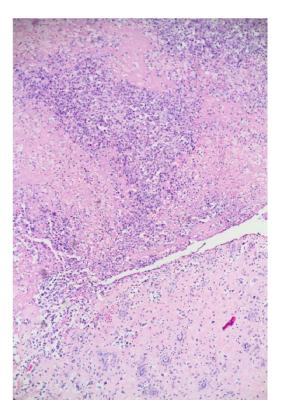
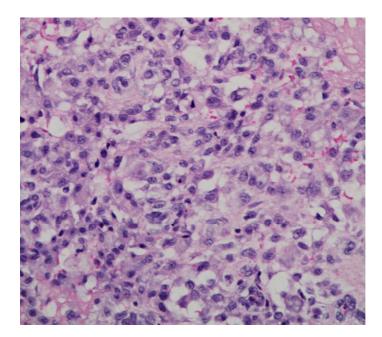


Figure 5 (Hematoxylin & Eosin; original magnification x40) Photomicrograph of bursa revealed presence of sheets of loosely packed cells surrounded with fibrinous material and fibrous stroma.



Figur 6 (Hematoxylin & Eosin; original magnification x200) High power of the loosely packed sheet disclosed large pleomorphic cells having vesicular nuclei and interspersing with acute and chronic inflammatory cells.

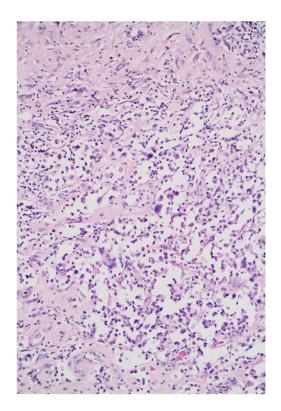
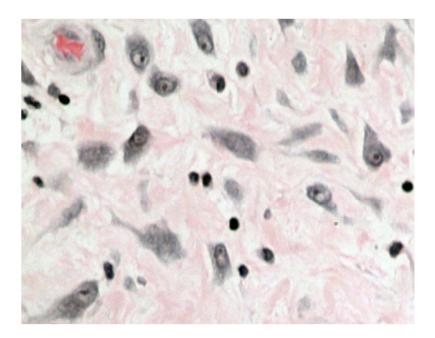


Figure 7 (Hematoxylin & Eosin; original magnification x100) Some foci revealed pleomorphic cells clinging along the sinusoidal spaces.



Figur 8 (Hematoxylin & Eosin; original magnification x400) There are cells having vesicular nuclei, prominent nucleoli and basophilic cytoplasm resembling ganglion-like cells in the lesion.

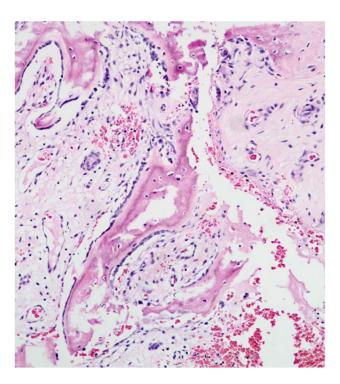


Figure 9 (Hematoxylin-Eosin; original magnification x100) Photomicrograph reveals reactive trabecular bones among the granulation tissue.

DISCUSSION

There are two types of bursitis in terms of pathogenesis. The first type is caused by inflammation in naturally preexistent bursa between bone and tendon, whereas the other one, so-called adventitious bursitis, is caused by repeated unusual shearing stresses of soft tissue overlying a bony prominence^{5,6}. The cyst in the present case lay in the anatomic site where the ischio-gluteal bursa is normally located. It is likely that it developed in the pre-existing bursa; however, the possibility of developing as adventitious bursitis could not be excluded since this type of bursitis often found associated with bed-sore (5). It is interesting that the gross-specimen demonstrated a fistula tract between the bursitis cavity and the overlying skin ulcer (fig. 3). The lesion in this case is quite similar to two reported cases caused by perforated ischiogluteal bursitis forming chronic sinus drainage7. This might be an explanation as to why the ulcer did not heal readily following the previous debridement and muscle flap surgery.

Microscopically there were a few small scattered islands comprising pleomorphic cells, having enlarged pale-stained round to oval shaped nuclei. Such features may arouse the suspicion of malignancy especially myxofibrosarcoma. Some cells arranged in sinusoidal pattern might alternatively suggest angiosarcoma. The presence of ganglion-like cells and metaplastic bone intermingled with substantial inflammatory cell infiltrates and fibrinous material are usually seen in pseudosarcomatous lesions, and should favor a reactive diagnosis rather than a neoplasm^{1,8}. More over, malignancy following chronic bursitis has only rarely been described, and the average period for developing cancer in this setting was not less than 15 years⁴. The bursitis in our case lasted about 4 years and the incubation period appears to be too early to develop malignancy,

compared with these previously reported cases.

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