Virchows Arch [Pathol Anat] (1982) 397:93–101

Virchows Archiv A

Synovial Sarcoma Arising in an Anatomical Bursa

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Summary. In previous studies, the origin of synovial sarcoma directly from synovium has not been satisfactorily established. This case report describes the light and electron microscopic features of a biphasic synovial sarcoma occurring within the popliteal fossa. At surgery, a cystic mass was identified in relationship to the semitendinosus tendon at the anatomical site of the semitendinosus bursa. The tumour originated from the inner surface of the bursa as multiple papillary projections with no evidence of extension beyond the capsule of the bursa. Portions of the synovial surface were hyperplastic but otherwise normal. The findings indicate that biphasic synovial sarcoma can arise directly from synovium and support the hypothesis of a mesenchymal histogenesis for this tumour.

Key words: Biphasic synovial sarcoma – Semitendinosus bursa – Electron microscopy

In reporting a large series of tendosynovial sarcomas, Hajdu and associates (1977) summarized accumulated evidence that suggested an origin of this neoplasm from mesenchymal tissue with the potential to differentiate as synovium. However, the origin of tendosynovial sarcoma from synovium has not been substantiated. The majority of synovial sarcomas do not originate in synovial linings but are usually closely associated with large joints, tendons or aponeurotic fascia (Cadman et al. 1965; Crocker and Stout 1959; Fernandez and Hernandez 1976; Mackenzie 1966). Though some tumours are grossly and histologically in continuity with the synovium of joints or bursae at the time of surgical excision, this is not in itself conclusive evidence for origin from this membrane. Recent ultrastructural observations have, in fact, questioned the origin of synovial sarcomas from mesenchymal

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0340-1227/82/0397/0093/$01.80

tissues and have suggested that this tumour class may be derived from true glandular epithelium (Evans 1980; Mickelson et al. 1980). To provide further evidence for a mesenchymal derivation and more specifically, synovium, for tendosynovial sarcoma, we are reporting light and electron microscopic findings in a case of biphasic synovial sarcoma arising in the popliteal fossa within the semitendinosus bursa.

Case Report

The patient, a 33 year old registered nurse with no previous history of illness, first noted a small lump behind her left knee 3 years prior to surgery. A diagnosis of Baker's cyst was made and she was managed expectantly until approximately 2 months before operation. At this time she noted tenderness behind the knee on walking and also some rest pain, but no enlargement of the mass. Clinical examination confirmed the presence of a 3×2 cm cystic lesion palpable in the medial part of the left popliteal fossa, in association with the semitendinosus tendon. At surgery a tense, fluid filled, whitish blue, cystic mass arising from the medial aspect of the popliteal fossa was found in close conjuction with the semitendinosus tendon, to which it was adherent in one area. The mass was easily dissected free by blunt digital dissection, necessitating sharp dissection only from the semitendinosus tendon. During the process of removal it ruptured and a small quantity of brownish fluid with some whitish debris floating in it escaped. Macroscopic examination of the removed lesion showed a pearly white, partially opened, semicystic, smooth oval mass 3 cm in maximum dimension. In areas the lining was smooth but the bulk of the cavity was occupied by inward projections of papillary brownish tan friable tissue.

Materials and Methods

Portions of the excised lesion were initially fixed in 10% buffered formalin, routinely processed and embedded in paraffin. Special stains performed included reticulin, PAS (with and without diastase), alcian blue, mucicarmine and Hale's colloidal iron.

Subsequently, portions of tissue were retrieved from formalin, post-fixed in osmium tetroxide, dehydrated through a graded series of alcohols and embedded in Epon-Araldite. Toluidine blue-stained plastic sections were used for orientation and the selection of areas for ultrastructural studies. Uranyl acetate and lead citrate stained thin sections were examined in a Phillips 301 electron microscope.

Results

The surgically resected lesion was a cystic structure with a well-defined but thin fibrous capsule (Fig. 1). Although, at the time of resection, the lesion was distinctly unilocular, as a result of distortion during fixation some sections appear to have loculated compartments (Fig. 1). A major portion of the inner surface and lumen of the cyst was occupied by papillary or polypoid proliferations of tumour (Figs. 1 and 4) with histologic features of synovial sarcoma of biphasic type (Fig. 2). Considerable portions of the tumour projecting into the central cavity of the bursa showed regions containing variably sized and irregularly shaped, but distinct, glandular structures lined by a relatively orderly arranged low columnar epithelium (Fig. 2). These gland-like spaces were surrounded by a spindle cell component which in some regions was continuous with adjacent areas composed solely of poorly defined interlocking fascicles of spindle cells (Fig. 2b). Between some

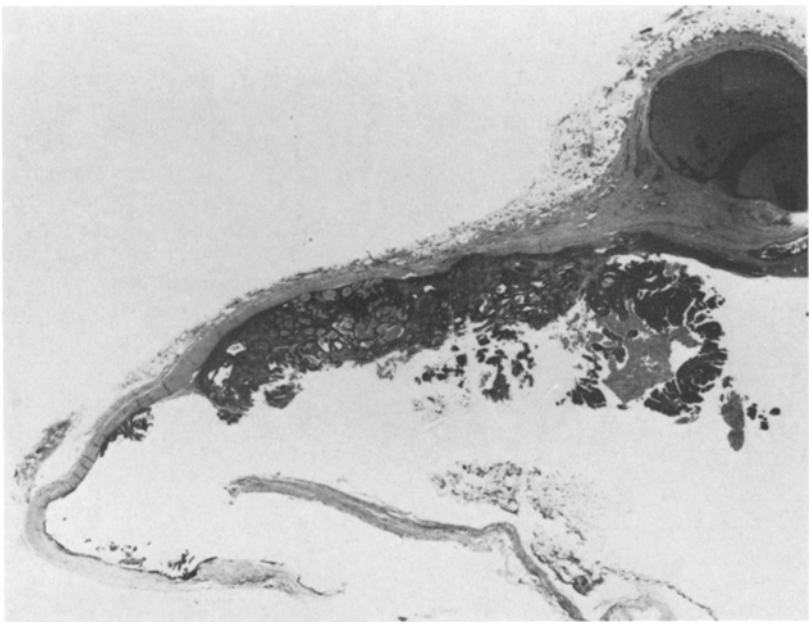

Fig. 1. Distinct, thin, fibrous bursal wall partially lined in this section by projecting masses of biphasic synovial sarcoma (H and E, × 48)

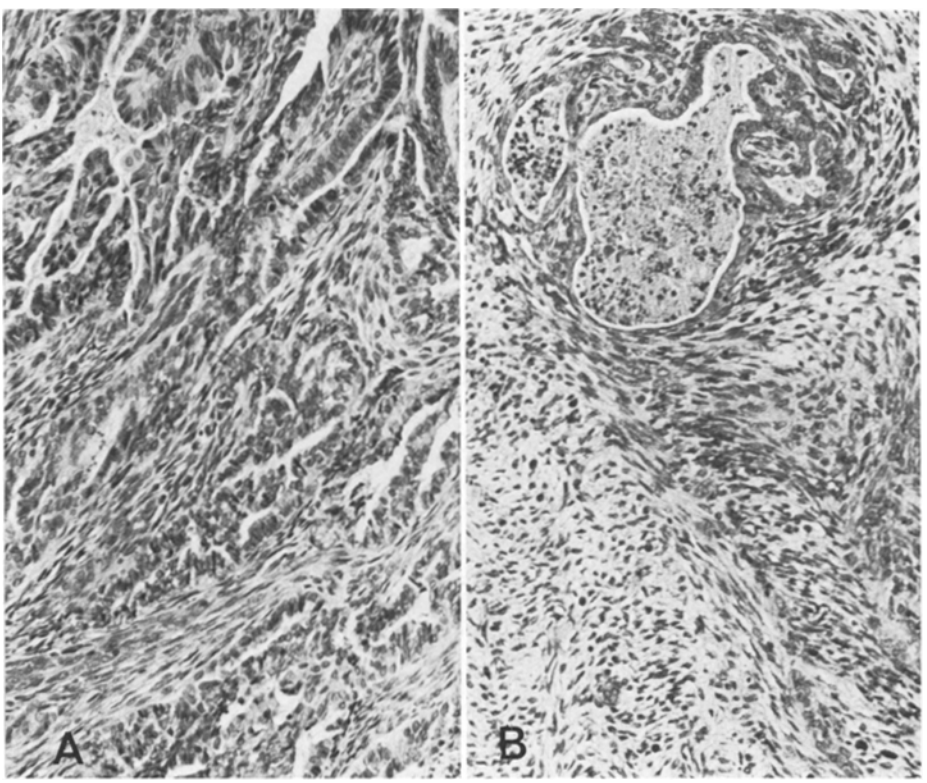

Fig. 2 (A) Biphasic portions of synovial sarcoma with slit-like epithelioid glandular spaces encompassed by the spindle cell phase. **(B)** Area of synovial sarcoma with a preponderance of fascicles of spindle cells and a fibrosarcomatous appearance surrounding epithelial lined gland-like spaces (H and E, × 125)

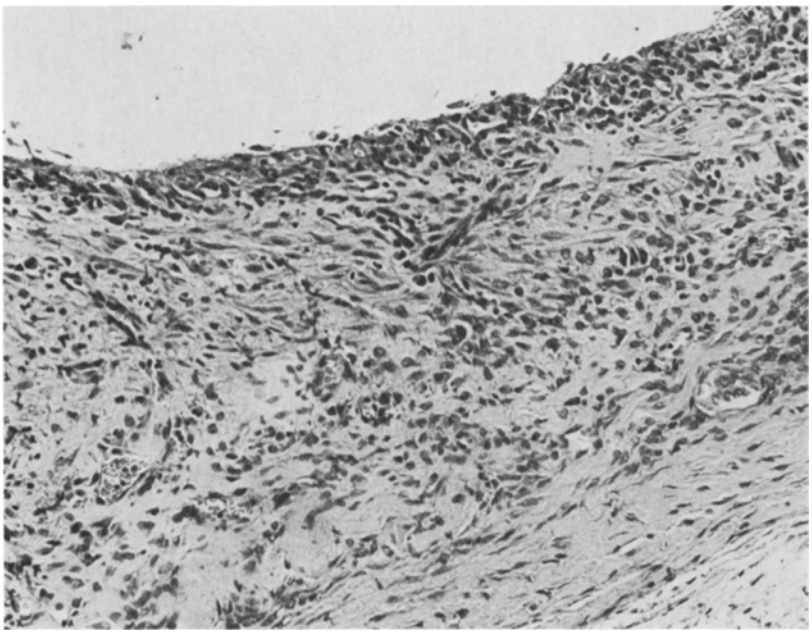

Fig. 3. Bursal capsule (*lower right*) lined on the inner surface by hyperplastic but otherwise normal synovial tissue (H and E, × 375)

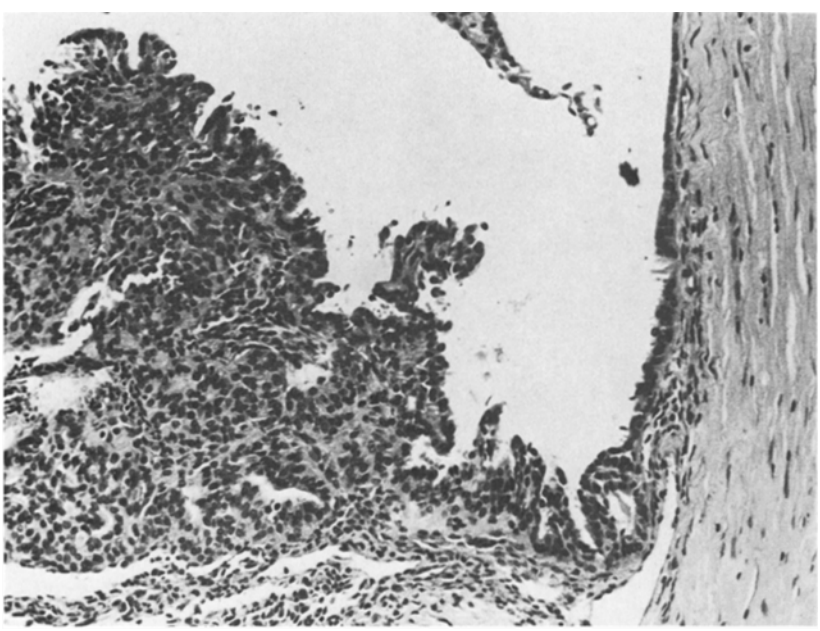

Fig. 4. Dysplastic epithelial-like lining of bursal wall (*right side*) continuous with surface cells of polypoid portion of sarcomatous tissue. Note similarity of surface cells to those within the tumour tissue (H and E, × 375)

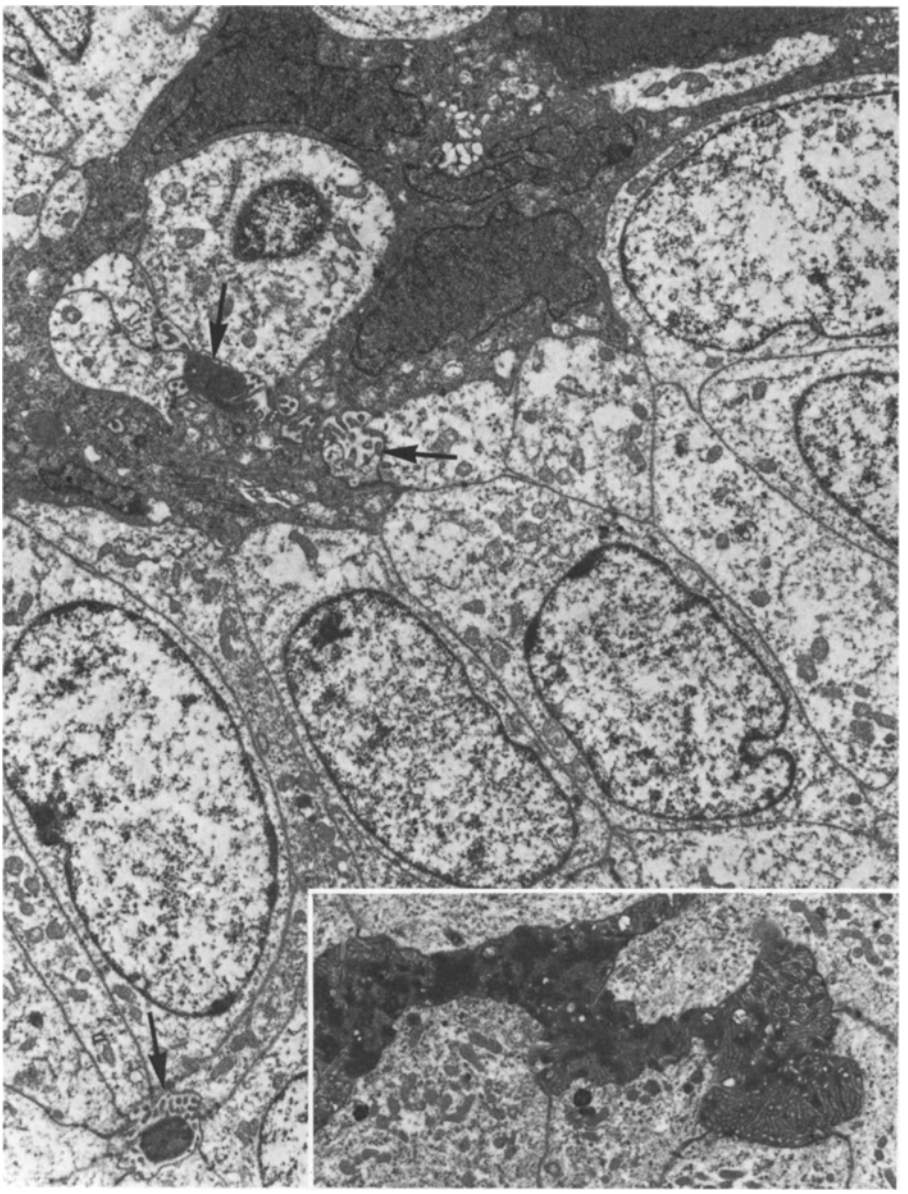

Fig. 5. Closely associated tumour cells of "light" and "dark" type involved in the formation of small, intercellular, microvilli-containing lumens (*arrows*) with zonula occludens-like junctions. Both cell types have relatively organelle-poor cytoplasm (× 6,000). *Inset.* Well-formed extracellular luminal space packed with narrow microvilli and electron-dense granules probably representing secretory products (× 4,000)

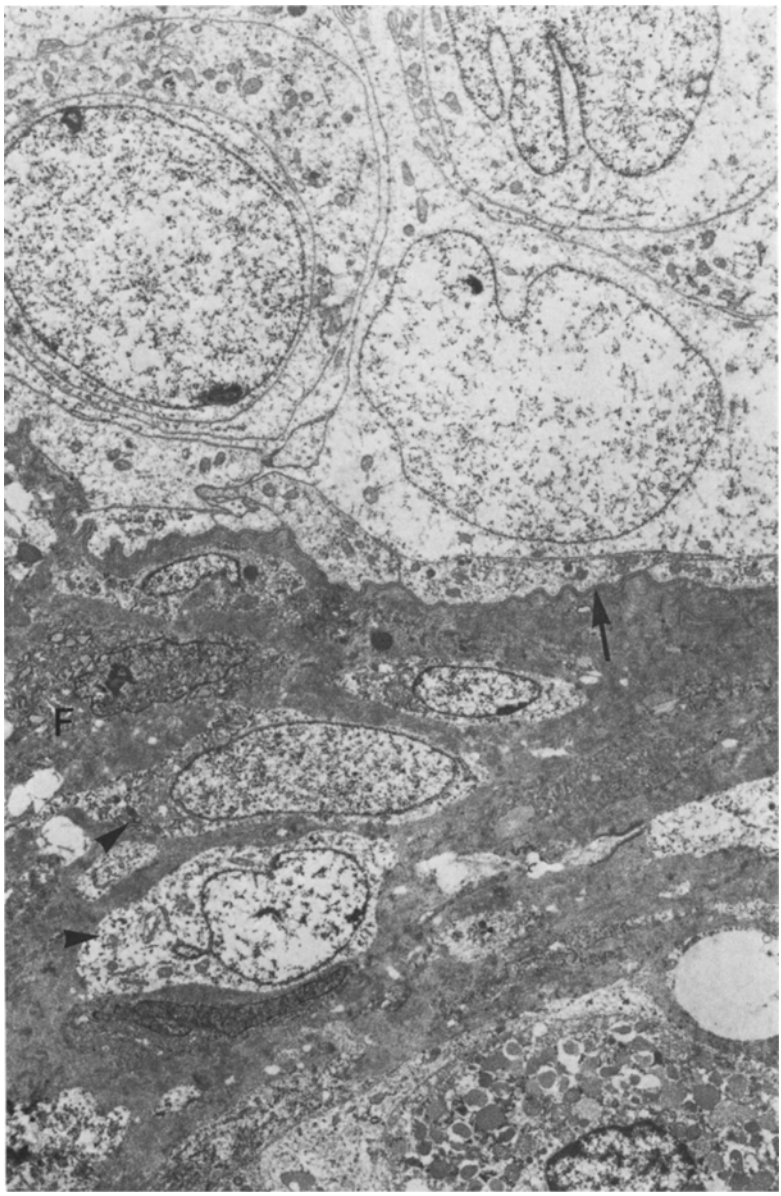

Fig. 6. Epithelioid tumour cells (*above*) separated from adjacent stromal tissue by a distinct narrow basement membrane (*arrow*). Stromal tissue contains spindle-shaped cells (*arrowheads*) similar in appearance to the tumour cells and occasional fibroblasts (*F*) (×4,500)

projecting masses of tumour, the cyst wall was lined by hyperplastic synovium (Fig. 3). Other portions of the inner surface were covered by a single layer of hyperplastic epithelioid cells continuous with similar cells on the surface of projecting tumour masses (Fig. 4) and closely resembled the epithelial-type cells lining the glands.

Alcian blue, PAS (after diastase), mucicarmine and colloidal iron (after hyaluronidase) stains were all positive. Mucopolysaccharides identifed were present in glandular lumens and as intracytoplasmic vacuoles in many of the epithelioid cells of biphasic areas. Frequently, both neutral and acid mucopolysaccharides were identifiable in the same gland-like lumens.

Ultrastructurally, the biphasic area was composed of polygonal to slightly spindle shaped, closely associated cells with a small to moderate amount of relatively organelle-poor electronlucent cytoplasm (Figs. 5 and 6). These cells had ovoid shaped nuclei with slight contour irregularities, a small peripherally situated nucleolus and a disaggregated chromatin pattern. Interspersed amongst them were occasional small groups of more irregularly shaped and darker staining cells, also with few cytoplasmic organelles; these showed greater nuclear irregularites and denser staining but the chromatin and nucleolar distribution was similar (Fig. 5). Although desmosome-like junctions were infrequently seen and generally small they were occasionally better developed and associated with tonofilament bundles; this was noted particularly in relationship to zonulae occludentes-like junctions that were associated with numerous variably-sized intercellular gland-like lumens (Fig. 5). These lumens contained moderate to prolific numbers of slender microvilli and densely staining secretory products. The darker staining cells also took part in the formation of intercellular lumens and junctional cell membrane specializations (Fig. 5). Epithelioid tumour cells are separated from the stromal component by a distinct narrow basal lamina (Fig. 6).

Discussion

In the tumour described in this report, both the histopathology and histochemical findings are typical of biphasic synovial sarcoma (Cadman et al. 1965; Evans 1980; Hajdu et al. 1977).

Although of limited number, the reported electron microscopic studies of biphasic synovial sarcoma have described histological and cytological features very similar to those detailed in the present report (Dische et al. 1978; Gabbiani et al. 1971; Golomb et al. 1975; Hirohata and Morimoto 1971; Klein and Huth 1974; Kubo 1974; Mickelson et al. 1980; Nunez-Alonso et al. 1979; Okagaki et al. 1976; Roth et al. 1975). Both in light microscopic and ultrastructural reports, these tumours had arisen in a variety of locations including soft tissues frequently in relation to joints but in none of them was a continuity with synovium actually demonstrated, grossly or microscopically. This is in keeping with the well recognized observation that the majority of synovial sarcomas do not appear to originate in synovial linings (Fernandez and Hernandez 1976).

The anatomic location of the lesion deep in the popliteal fossa intimately associated with the semitendinosus tendon, the gross description of a well-defined cystic structure and the histological appearance of the cyst wall enclosing the tumour with residual areas of hyperplastic synovium are all consistent with a bursa. Although multicystic synovial sarcomas have been

described (Hajdu 1979), grossly the lesion was unilocular with the loculated appearance in Fig. 1 due to distortion of the opened bursa during fixation. The cystic nature of the lesion did not appear to have arisen as a result of tumour degeneration or sloughing of tumour tissue during preparation for histological examination, since the dysplastic epithelium lining areas of the bursa was frequently continuous with the surface of the papillary tumour.

The absence of cell membrane junctional specializations and basement membrane formation in normal human synovium (Barland et al. 1962; Ghadially 1980) and their presence in synovial sarcomas (Gabbiani et al. 1971; Golomb et al. 1975; Hirohata and Morimoto 1971; Klein and Huth 1974; Kubo 1974; Mickelson et al. 1980; Nunez-Alonso et al. 1979; Okagaki et al. 1976; Roth et al. 1975) has been difficult to reconcile. These differences, together with the ability to form glands and the secretory capacity of the tumour cells have given rise to speculation that synovial sarcomas may not arise from synovial membranes, or even be of mesenchymal derivation (Dische et al. 1978; Evans 1980; Mickelson et al. 1980). However, ultrastructural studies of synovial tissue from a variety of animals (Groth 1975; Langer and Huth 1960; Roy and Ghadially 1967), and from human joints involved with rheumatoid arthritis (Grimley and Sokoloff 1966), clearly demonstrate desmosomes between synovial cells, and also basement membrane formation (Langer and Huth 1960). The potential to differentiate these types of cell membrane specializations appears to be expressed by the tumour cells of synovial sarcomas generally. The ultrastructural features of hyperplastic synovial cells (Grimley and Sokoloff 1966; Langer and Huth 1960) are similar to the germinal cell layer lining the surface of the ovary (Papadaki and Beilby 1971), a cell layer well known for its potential to give rise to a wide range of tumours with epithelial characteristics. It has been similarly speculated (Mazur and Kraus 1980) that uterine myometrial plexiform tumours and leiomyomas with tubular formation reflect the potential of the mesenchyme of the Müllerian ducts to form epithelial-like structures. Interestingly, both the light and electron micrographs of the plexiform tumour illustrated by Mazur and Kraus (1980) have close similarities to biphasic synovial sarcoma. The observations reported here indicate that synovium has a similar potential to differentiate as a true epithelium, thus supporting the thesis of a mesenchymal histogenesis for biphasic synovial sarcoma. This implies that mesenchyme in a variety of locations, including synovium, has the potential to give rise to synovial sarcomas.

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Accepted May 24, 1982