Fibroma of tendon sheath

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SUMMARY A series of nine cases of fibroma of tendon sheath is described including details of the ultrastructural features of two cases. The series was composed of lesions from six males and three females with a mean age of 38 yr. The most common site of involvement was the hand (including fingers) and the mean greatest diameter was 19 mm. Typically the tumours were lobulated and microscopically there was a collagenous stroma with spindle and stellate cells in a moderate degree of cellularity. One recurrence was noted in the series. The lesion was distinguished from circumscribed fibromatosis, nodular fasciitis, neurofibroma, leiomyoma, scar tissue, giant cell tumour of tendon sheath (localised nodular tenosynovitis) and fibrous histiocytoma. Ultrastructural studies revealed that the large majority of cells present in the two cases studied were myofibroblasts and fibroma of tendon sheath is therefore the third instance of a benign tumour containing these cells (the other two being dermatofibroma and giant cell fibroma of the oral mucosa).

In his classification of tumours of tendon sheath Buxton¹ considered that benign tumours included (i) lipoma, (ii) fibroma, (iii) chondroma and (iv) ganglion. Obviously the latter would not be considered a true neoplasm and the other three at first sight would be considered rare. Buxton also referred to a "giant celled myeloma" which was benign but liable to recur locally. It is obvious that this entity equates to the so-called giant cell tumour of tendon sheath a lesion which is probably not a true neoplasm and which is best regarded as a nodular tenosynovitis being a reactive proliferation of both A and B tenosynoviocytes.² Buxton regarded tendon sheath fibroma as uncommon and described a specimen from near the ankle and which was in the Museum of the Royal College of Surgeons of England. Geschickter and Copeland³ described a fibroma of tendon sheath as an encapsulated tumour composed of tightly packed spindle cells surrounded by collagen fibres.

From 1936 to 1979 no additional cases of fibroma of tendon sheath were reported in the literature until Chung and Enzinger⁴ described the clinical, macroscopic and light microscopical characteristics of this tumour. They described a well circumscribed often lobulated tumour which was usually attached to tendon or tendon sheath. Microscopically there was dense fibrocollagenous tissue containing usually

spindle but sometimes stellate cells and frequently bearing dilated or slit-like vascular channels some of which resembled tenosynovial spaces. In the last few years we have had experience of nine cases of tendon sheath lesions which fulfil the diagnostic criteria of fibroma as laid down by Chung and Enzinger. Our experience with these lesions is reported here in view of the fact that the majority were not initially recognised as fibromas and that electron microscopical studies were performed in two cases.

Material and methods

All tendon sheath lesions accessioned to the Division of Tissue Pathology of the IMVS over the 10-year-period 1971-80 were reviewed and nine cases were identified which exhibited features consistent with a diagnosis of fibroma. Haematoxylin and eosin (HE) stained sections were available in each case. In addition the following special stains were applied (i) Perl's Prussian blue, (ii) Masson's trichrome, (iii) Gordon and Sweet's reticulin, (iv) PAS, (v) PAS after diastase digestion, (vi) MSB, (vii) alcian blue (pH 2.5), (viii) Miller's elastin stain and (ix) von Kossa. In two cases portions of formalin-fixed tissue were post-fixed in osmium tetroxide and processed into Spurr's resin. Survey sections were cut and representative blocks were thin-sectioned for electron microscopy.

A limited amount of clinical data was abstracted

Clinical and macroscopic data of nine cases of fibroma of tendon sheath

Case	Sex	Age	Site	Clinical history	Macroscopic appearance
1	M	41	Not known	Not known	An encapsulated white nodule 20 × 10 × 10 mm
2	M	48	Palm of left palm	Swelling after injury	Irregular, firm grey nodule 26 mm in maximum diameter
3	F	26	Palmar aspect of left middle finger	Not known	Fibrous nodule size unknown
4	F	68	Back of the left hand	Cystic swelling	Firm, white nodule 25 × 25 × 20 mm
5	M	23	Not known	Not known	Not known
6	M	18	Right middle finger	Not known	Ovoid white nodule 20 × 15 × 15 mm
7	F	29	Extensor tendon apparatus of left hand	Described as a ganglion	White nodule 7 × 5 × 2 mm
8	M	58	Recurrence of above Ulnar aspect of distal phalanx of left index finger	" " Swelling present for a few months. Not painful	Irregular greyish tissue 12 × 8 × 5 mm Firm, white nodule 12 × 18 × 10 mm Weight = 1.6 g
9	M	33	Left knee	Smooth rounded tumour attached to quadriceps mechanism	Firm, white nodule 30 × 20 × 10 mm

from the information submitted with each biopsy and this is shown in the Table.

Results

An examination of the clinical data shows that in the nine cases there was one recurrence making 10 specimens available for examination. There were six males and three females in the series. The mean age was 38 yr (range 18–68 yr). The lesion was related to the knee in one case and to the hand in six instances. Three of these were related to fingers and three to either the palmar or dorsal aspects of the hand.

The mean greatest diameter was 19 mm (range 7–30 mm) and in seven specimens there was a distinct lobulation. In one case mucoid areas were seen macroscopically and in one other microscopically. Microfoci of cystic change were noted in two specimens. Generally the stroma was markedly collagenous (confirmed by the trichrome stain) and the majority of constituent cells were spindle-shaped (Figs 1, 2). Blood vessels were evenly dispersed throughout in nine instances and only in one were slit-like spaces prominent. Limited areas of looser, less fibrous stroma were seen in six samples and the constituent cells were here stellate in shape (Fig. 3). Assessment of the overall degree of cellularity revealed it to be mild in three, moderate in five and marked in two specimens. Extravasation of erythrocytes (in small numbers) was seen in three cases. Occasional multinucleated giant cells (macrophage polykaryons) were seen in one case. Foam cells were not seen in any of the cases studied. There was no significant mitotic activity.

Small foci of mucoid change were seen in two cases and these areas were shown to contain an acid mucin. In two other cases there were small areas

exhibiting a storiform pattern. There was a fine reticulin pattern with fibres apparent around individual cells in all cases. A small amount of haemosiderin was present in three specimens but there was never any detectable fibrin. No elastin was seen in the lesions and the von Kossa stain was uniformly negative. True encapsulation was not seen. In several cases there was condensation of marginal connective tissue giving the appearance of a pseudocapsule.

Ultrastructural studies were performed on two of the cases and identical features were seen in both. Sections revealed spindle and stellate cells set in a collagen matrix. The nuclei varied from regular and elongated to highly convoluted the latter being much frequent (Fig. 4). Chromatin was margined and a prominent nucleolus was only occasionally seen. The cytoplasm contained large amounts of rough endoplasmic reticulum which was often dilated by an amorphous substance. Free ribosomes occurred in large numbers but only an occasional swollen mitochondrion was seen. There were also large vacuoles present which appeared to have contained lipid. Fine myofilaments were visible just beneath the cell surface and occasional larger bundles were seen between the organelles of some cells (Fig. 5).

At higher magnification these myofilament bundles exhibited periodic densities indicating their similarity to actin (Fig. 6). The cell surface tended to be indistinct but no cell-cell or cell-stroma (hemidesmosomes on a basal lamina) junctions were seen. Occasional groups of pinocytotic vesicles were visible however (Fig. 7). Clefts frequently occurred between the collagen and the cells and between neighbouring cells (Fig. 4). These clefts were not bounded by a basement membrane and cell cytoplasm rarely extended around their entire internal

Fig. 1 *Fibroma of tendon sheath with a collagenous stroma and constituent spindle and stellate cells. Haematoxylin and eosin $\times 100$.*

Fig. 2 *The majority of cells are spindle-shaped. Haematoxylin and eosin $\times 200$.*

Fig. 3 Less frequent stellate cells are illustrated. Haematoxylin and eosin $\times 400$.

circumference. No erythrocytes were seen in these structures.

Discussion

The clinical data were limited in this series but in agreement with the findings of Chung and Enzinger⁴

the lesion was more common in males than females and the fingers and hands were most commonly affected. The mean age in the present series was 38 yr compared to a median of 31 yr in that of Chung and Enzinger. The mean diameter was 19.0 mm compared to a median of 18.0 mm. Only in one case was there recurrence of the lesion (five months after initial excision). This is considerably less than the recurrence rate of 24% reported by Chung and Enzinger but their series was much larger and follow-up was for a considerably longer period of time.

In determining the typical features of fibroma of tendon sheath it is evident that a collagenous stroma generally containing spindle cells in a moderate degree of cellularity were the dominant features.

Blood vessels were fairly prominent in a majority and in 80% an attachment to tendon sheath was evident. Lobulation was observed in 70%. Foci of stellate cells were observed in 60% of cases. These represented the main diagnostic features and were similar to the criteria developed by Chung and Enzinger except that slit-like spaces were infrequent in the present series.

In a consideration of the differential diagnosis of these lesions one must exclude circumscribed fibromatosis, nodular fasciitis, neurofibroma, leiomyoma, scar tissue, giant cell tumour of tendon sheath (localised nodular tenosynovitis) and fibrous histiocytoma. Distinction from nodular fasciitis and

Fig. 4 Spindle cells in a collagenous matrix (M) showing ovoid and convoluted nuclei (N). Clefts are visible between the cells and the collagen (C). $\times 1900$.

Fig. 5 Spindle cell with a convoluted nucleus (N), rough endoplasmic reticulum (RER), vacuoles (V), and a thin band of myofilaments just beneath the plasma membrane (arrows). $\times 5400$.

fibrous histiocytoma was relatively straightforward since myxoid change and extravasations of erythrocytes (features of the former) and a storiform pattern (a feature of the latter) were inconspicuous findings. In addition lesions were lobulated and not nodular. The circumscribed margins of the lesions tended to exclude fibromatosis. None of the lesions was considered to exhibit sufficient evidence to support the diagnosis of localised nodular tenosynovitis. Foam cells were never seen, siderophages were only noted in three cases and then infrequently and only a few multinucleated giant cells were seen in one

case. However the possibility must still be admitted that the lesions denoted as fibroma of tendon sheath might represent an endstage of a nodular tenosynovitis.

The ultrastructural characteristics of fibroma of tendon sheath have not been previously described. A study of two cases in the present series revealed identical features. Ultrastructurally open clefts occurred between the collagen and cells and between cells. These were considered probably to represent the dilated slit-like vascular channels likened by Chung and Enzinger to tenosynovial spaces.

Fig. 6 Higher power view of myofilament bundles (M) showing periodic densities (arrows). $\times 26\,200$.

Fig. 7 Portion of spindle cell showing rough endoplasmic reticulum (RER), myofilaments (M) and pinocytotic vesicles (arrows). $\times 12\,500$.

From our studies it is evident that these spaces were not vascular channels. There was no basement membrane and often there was no cell cytoplasm bounding the edge of the collagen. The spaces were devoid of red blood cells.

Of particular interest was the nature of the cells observed intrastructurally. Fibroblasts were seen rarely. These were elongated cells characterised by a smooth elongated nucleus, moderate to large amounts of rough endoplasmic reticulum (often dilated) and small numbers of other organelles including fine cytoplasmic filaments. The majority of cells seen were myofibroblasts. These cells combine the features of fibroblasts and smooth muscle cells. They exhibit nuclear indentations and folds, bundles of intracytoplasmic microfilaments arranged parallel to the long axis of the cell and dense bodies similar to those in smooth muscle cells, surface differentiations with desmosomes and hemidesmosomes, abundant rough endoplasmic reticulum and pinocytotic vesicles.⁵

Myofibroblasts has been described in a variety of lesions. These include granulation tissue,⁵ atheromatous plaques,⁶ ganglia,⁷ hepatic cirrhosis,⁸ triggering tenosynovitis,⁹ Dupuytren's contracture,¹⁰ desmoid fibromatosis,¹¹ circumscribed fibromatosis¹² and nodular fasciitis.¹³

As has been pointed out by Vasudev and Harris¹⁴ few reports exist of myofibroblasts occurring in true neoplasms. They refer to six documented cases of malignant tumours which contained myofibroblasts and which had been classified as fibrosarcomas. With regard to benign tumours nine cases of typical dermatofibroma have been shown to contain myofibroblasts.¹⁵ A giant cell fibroma of the oral mucosa has also been reported to contain myofibroblasts.¹⁶ If it is accepted that fibroma of

tendon sheath is a true benign neoplasm then it is an example of a tumour of myofibroblasts.

The nature and significance of these cells have been discussed by Ryan *et al*⁵ and Vasudev and Harris.¹⁴ The former have suggested that myofibroblasts arise locally from more conventional fibroblasts or from primitive mesenchymal cells. Fibroblasts cultured *in vitro* develop intracytoplasmic fibrillar systems similar to those of myofibroblasts.¹⁷ Vasudev and Harris suggest that myofibroblasts represent the expression of a limited potential in fibroblasts for differentiation along myoid lines and that the development of microfilaments is the normal reaction to a variety of proliferative stimuli.

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