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Case Report

Pigmented Villo-Nodular Bursitis of Pes Anserinus Bursa: A Rare Case Report and Review of Literature

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Abstract

16 yrs old female present with insidious onset swelling for last 6 weeks with intermittent pain without any functional deficit. MRI with contrast revealed abnormal cystic swelling with characterstic finding of hetrogenous contrast enhancement due to haemosiderin deposition pointing towards Pigmented villonodular bursitis. After preanaesthetic clearance and informed consent, Incisional biopsy with marginal resection was done. Histopathology revealed Pigmented villonodular bursitis which is a rare entity at rare site of presentation. In our study, we have compared it with old reported cases and reviewed the literature for better understanding the pathology and recogonizing need for further investigation to understand its etiology.

Keywords: Pigmented villinodular synovitis and bursitis, Pes anserinus bursa

Introduction

Localized - Pigmented villonodular Synovitis (L-PVNS) is rare, locally active, benign lesion involving either tendon sheath, synovial tissue of bursa rarely intraarticular. Pigmented villonodular bursitis (PVNB) is least common [1,2]. It is second most common benign soft tissue tumor of fingers [1,2]. PVNB of Pes anserinus bursa is rarely documented diagnosis. Our purpose is reporting a rare case and review of literature for early diagnosis and better management.

Case Report

16yrs old female presented with pain and swelling of left inner side of upper leg since 6weeks. Swelling was insidious onset, gradually progressing, localized to left medial aspect of upper leg, associated with pain, no fever. Left knee was asymptomatic. On examination single oval swelling of 5*4 cm localized on medial aspect upper leg, soft to firm, slightly warm and tender, non-compressible, immobile, fixed to underling tissue. There were no dilated veins, pulsation, fluctuation transelumination. No increase in size on walking. Tinel's sign was negative. Knee and distal neurovascular examination was within normal limits. X-ray was normal. USG left knee swelling revealed Cystic lobulated swelling of 2.7*6.0 cm with front projection visible with mild vascularity. MRI left knee revealed cystic swelling in between normal pes anserinus tendons of 3.8*2.6*6.0 cm extending till medial knee joint without any communication with synovial thickening and papillary projection with gadolinium enhancement (Figure 1-3). Blood test was within normal limits.

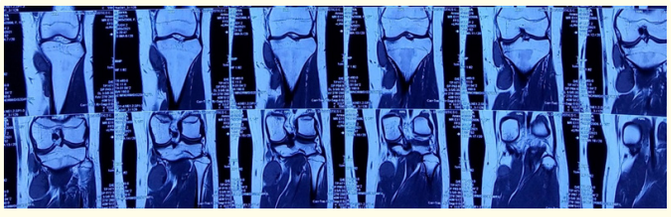

Figure 1: MRI T1W image showing coronal cut section from anterior to posterior with abnormal mass with Hypo to isotense signal over pes insertion extending anterior to posterior as visible in sequential sections shown with black arrows.

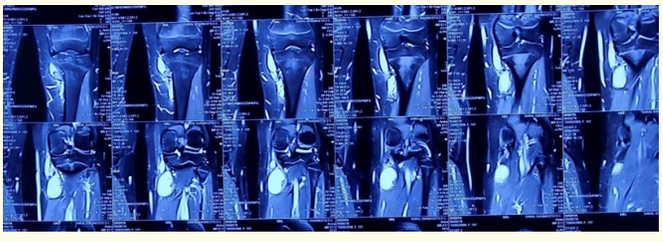

Figure 2: MRI T2W image showing coronal cut section from anterior to posterior with intermediate to high signal cystic swelling over pes tendons extending till medial knee joint without involving joint capsule as visible in sequential section shown by black arrow.

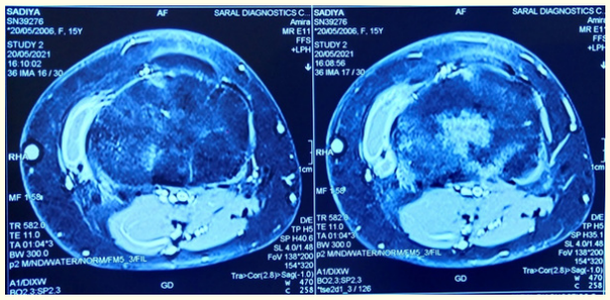

Figure 3: Contrast MRI axial section at proximal tibia level showing encapsulated swelling with enhancement not involving bone shown by black arrow.

Following informed consent and pre-anaesthetic clearance, Incisional open biopsy with marginal resection was done under general anaesthesia and tourniquet control. Enbloc swelling sample sent for histopathology and tissue culture and sensitivity. Gross appearance was irregular grey brown tan soft tissue. Frozen section showed diffuse, papillary, villous, nodular hyperplastic synovium with stroma of lymphomononuclear cells, foamy cell, multinucleate giant cells, haemosiderin laden macrophages with no anaplasia, atypical mitosis and necrosis inferring PVNB. Postoperative period was uneventful. Sutures were removed at 2 weeks. Patient was asymptomatic till 2 years of follow up.

Discussion

Pigmented Villonodular Synovitis is benign, proliferative, locally aggressive, intra or extraarticular synovial pathology [1,2,10]. PVNS family of disease broadly devided into Localized and diffuse [1,2,10]. (Figure 4). L-PVNS involve either synovial sheath of tendons or bursa [1,2,10]. Incidence of rare entity is 1.8 per million of populations [2,3]. Most of literature is written of PVNS, PVNB literature is sparse. There were only twelve case reports in literature at pes anserinus site of L-PVNS [2,3]. Few of them documented in our case report [2-8]. (Table 1). It can present at any age with peak age d). Itbution is 3-5th decade with female more than male population [2]. Review of old case reports ranging from 2-5th decade with high in female sex (F: M – 2:1) [9].

Most common site is interphalangeal joints of fingers (85%) followed by knee, ankle and foot (large joints - 12%) [9,10]. Commonest bursae are infrapatellar followed by suprapatellar and pes anserinus bursa [9]. Abdelwahab., *et al.* explained that there are two kind of bursa in our body communicating or non communicating (to joint) [4]. PVNB of non communicating bursa like pes anserinus is extremely rare [4]. Its etiology is uncertain like inflammation, allergic, chromosomal abnormalities and trauma [9]. Discomfort,

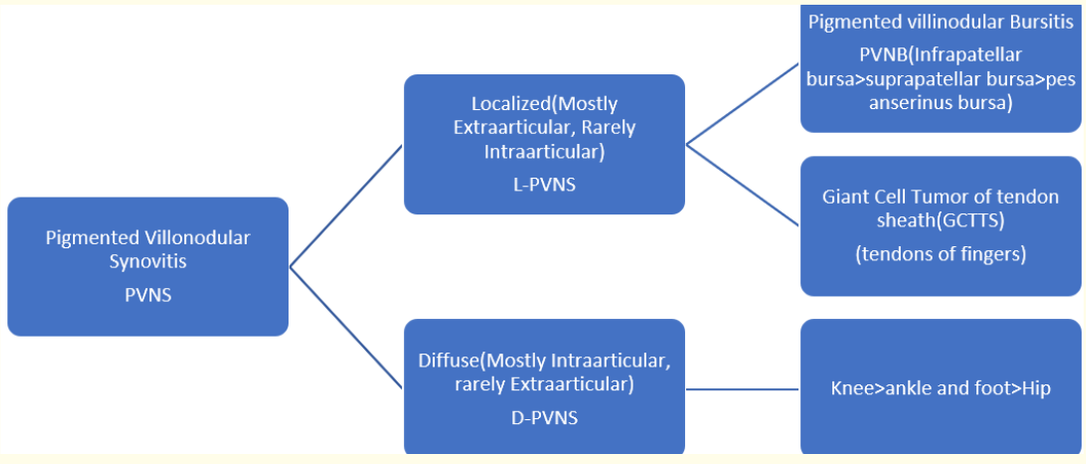

Figure 4: Classification of Pigmented villonodular synovitis as described by Jaffe., et al.

Author	Age	Sex	Site	Symtoms	MRI finding	Management	Recurrence and Follow up
Aditya V Maheshwari., *et al*.	17yrs	F	Right leg	Insidious and progressive pain, swelling over pes anserinus region since 3 yrs, tender, firm in consistency No H/o trauma	5.6*3.8*3.5cm mass T1W-hypo/isotense to muscle tissue T2W- intermediate to high compare to fat Gradiant echo - hetrogenous enhancement to to haemosiderin deposition No knee joint involvement	Marginal Excision open biopsy Part of semitendinosus is sacrificed Histopathology- Multinucleate giant cell in stroma of mononuclear cell, foamy cells, haemosiderin deposition	No recurrence upto 2 yrs of followup.
Aditya V Maheshwari., *et al*. 2007	18yrs	M	Left leg	Insidous onset and progressive pain, swelling over pes region since 2 yrs, tender, firm, noncompressible, 7*4 cm mass No H/o trauma	6.1*3.5*3.4cm T1W-hypo/isotense to muscle tissue T2W- intermediate to high compare to fat Gradiant echo - hetrogenous enhancement to to haemosiderin deposition No knee joint involvement	Marginal Excision open biopsy Pes tendons were normal Histopathology- Multinucleate giant cell in stroma of mononuclear cell, foamy cells, haemosiderin deposition	No recurrence upto 18 yrs of followup.
Haitao Zhao., *et al*. 2011	28 yrs	F	Right leg	Insidous onset and progressive pain, swelling over pes region since 2 yrs, tender, firm, noncompressible, 7*4 cm mass No H/o trauma	4.5*3.0*2.0cm T1W-hypo/isotense to muscle tissue T2W- intermediate to high compare to fat Gradiant echo - hetrogenous enhancement to to haemosiderin deposition No knee joint involvement	Marginal Excision open biopsy Part of semitendinosus is sacrificed Histopathology- Multinucleate giant cell in stroma of mononuclear cell, foamy cells, haemosiderin deposition	No recurrence upto 2 yrs of follow up.
Abdelwahab., *et al*. 2002	63yrs	F	Right Leg	Insidous Onset and progressive pain, swelling with limitation of knee movement since 2 yrs with associated Sjogren's syndrome No H/o trauma	5*3.5*2.0 cm	Wide excision open biopsy with resection of pes tendons and repair of intact fibres	No recurrence upto 18 yrs of followup.
Sami., *et al*. 2003	26yrs	F	Left leg	Swelling and insidious onset pain since 2 yrs Knee range of motion normal No H/o trauma	4.5*3.0*2.5cm	Marginal Excision open biopsy	No recurrence upto 2 and 3 months of follow up
Anthony I. Riccio., *et al*. 2007	26yrs	F	Left Leg	Insidious onset pain and swelling left leg Soft, nonpulsatile swelling No H/o trauma	CT - cystic swelling of 3.5*6.5cm MRI - fluid-fluid level present, T1W- hypotense T2W- hetrogenic on contrast enhancement No knee joint and upper tibia ivolvement.	Marginal Excision open biopsy	No recurrence upto 2 and 6 months of follow up

Sungwook Choi., *et al*., 2017	21yrs	M	Left leg	H/o trauma 2 weeks back H/o pain and swelling left knee and leg Anterior drawer positive Swelling and pain over upper medial leg since 2 weeks	MRI - Medial meniscus tear, anterior cruciate injury, mass seen over pes insertion area T2W - hetrogenous mass on contrast enhancement due to haemosiderin deposition	1st surgery - marginal excision with partial excision of pes tendons Medial menisectomy of irrepairable tear 2nd surgery - 12 months after 1st surgery Arthroscopic Acl reconstruction	No recurrence after 3 years of first surgery
Aikaterini Solomou., *et al.*	56yrs	F	Left leg	H/o Palpable lump on medial side of left knee	MRI -T1W isotense to muscle T2W - hypertense signal Contrast - Haemosiderin intermediate enhancement with solid visible mass	Resection of mass	Not available

Table 1: Varoius Old case reports in literature similar to our case report.

atraumatic swelling and intermittent pain without associated trauma are main sysmtoms with normal knee range of motion [2-10]. History of locking knee was reported in a case report of PVNS of medial patellar expension [11]. Common Swelling findings in most case reports are slightly warm, firm or soft, non compressible, non-pulsatile, nonfluctuent, immobile swelling [2-10]. Size of range 4-8cm was reported in literature [8]. Its pathology is very similar to fibroma [2]. It is believe that both are two points of common pathology continuum [8]. Radiogragh and ultrasonography are not very helpful in narrowing diagnosis. Radiograph with calcification can help in excluding other differential diagnoses [1] (Table 2). MRI with contrast is main investigation of choice to narrow differential diagnosis (Table 2). MRI is also helpful in determining site, knee involvement, size, extent, tendon engulment for planning of surgery. Besides Bursitis, traumatic haematoma, common benign tumor/tumor like lesion synovial chondromatosis, synovial haemangioma, lipoma arborecens, fibroma and synovial sarcoma are common differential diagnoses (Table 2). Hetrogenous intermediate signal due to haemosiderin deposition on contrast enhancement T2W image is characteristic finding on MRI [1,2,4,10]. It could be solid or cystic swelling on USG [1]. Fluid fluid level can be present in cystic swelling [1]. In our case, it was cystic swelling. Main stay of management is marginal excision open biopsy with or without pes tendon resection depending on involvement [2-10]. Radiotherapy is controversial in L-PVNS form [1,10]. Hillary W. Garner., *et al.* reported recurrence rate of PVNS is 8-56% depending on site of lesion, 8% in extraarticular localized form to 56% in intraarticular diffuse form [1]. Recurrence is mainly due to incomplete resection of synovium and involved tissue [1,4,10,12]. There was one case of recurrence reported in old literature [12]. Ohnuma., *et al*. reported a case of PVNB of pes anserinus bursa with knee extension. Lesion was recurred after 1.3 yrs from time of surgery because synovectomy of knee was not done initially. Following which second surgery of synovectomy and bursectomy was done. There was no recurrence till 2 years of follow up [12]. PVNB is rare pathology involving bursa needs further investigation to understand its etiology, prognosis.

Conclusion

PVNB is rare pathology of synovium, scarsely understood than broad term PVNS. Although rare but knee bursae are second common site after interphalangeal joints of finger. PVNB is benign proliferative synovial tumor like pathology common in all ages from 2-5th decade with female preponderance [1]. MRI with contrast is important for early diagnosis and planning for management. But Radiographs are helpful in excluding other deferential diagnoses. Although anecdotal but size can't be used as prognostic marker for recurrence. Recurrence is rare after marginal excision. Resection of parts of pes tendons engulfed by swelling can help in reduction of recurrence. Knee joint involvement can be one of the cause recurrence even after resection.

Differential Diagnosis of Neoplastic lesions	Age/Sex	Symptoms and Signs	Investigation Findings	Histopathology features
Pigmented Villinodular Synovitis	Diffuse-3-4th decade, equal sex distribution, 4 times comman than localized Localized-3-5th decade, female >males	Diffuse-Recurrent synovitis, monoarticular mostly involve knee, can lead to degenerative arthritis Localized-extraarticular mostly discomfort, firm or cystic swelling Most common site-interphalangeal joints finger, around knee, ankle and foot	Diffuse- Xray-normal joint space to arthritic in late cases MRI-Frond like synovial projection, joint effusion, hypervascular Localized- X ray-soft tissue swelling, sometimes bone erosion due to pressure, USG-well defined solid (sometimes cystic) vascular hypoechoic, homogenous engulfing tendons MRI-T1w-hypo/isotense to muscles T2W- hetrgenous due to haemosiderin deposition	Multinucleate giant cell in stroma of sheets of mononuclear cells, foamy macrophages, haemosiderin deposition
Synovial Chondromatosis	Middle age Equal sex distribution	Intraarticular>extraarticular Knee most common Recurrent effusion, locking, giving away	X ray-ring and arc type chondroid mineralization, equal sizes All lose bodies can combine to form synovial chondroma (upto 20cms)	Nodular cartilageous proliferation with atypia, necrosis Osteochondral loss body-enchondral ossification
Synovial Haemagioma	Children>adults Female >male	Recurrent synovitis(haemarthrosis) Quadriceps atrophy Can lead to arthropathy	X- ray-osseous erosion, phlebolith MRI-T1W- intermediate signal T2W-high signal with low signal of sepatation, fluid fluid level due to sluggish flow	Cavernous type haemangioma, pedunclated
Lipoma Arborescens	9-66yrs Equal sex	Nonspecific fullness Sometimes effusion	MRI- T1W-lobular frond like projection T2W-effusion, projection with fat like consistency mostly in suprapatellar fossa	Lobular frond like projection, fatty tissue and lipoma cells
Synovial Sarcoma				

Table 2: Differential Diagnosis of common benign synovial tumor/ tumor like swellings around knee in our case.

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