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Pigmented villonodular synovitis of the subarocmial space (case report)

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Abstract

Pigmented villonodular synovitis (PVNS) is a rare benign proliferative disorder affecting synovial membranes, bursae, tendons, skin and bone. **Materials** The article presents a rare clinical case treated for PVNS of the subacromial space. The patient complained of pain in the shoulder joint at the largest possible points, and a deformity developed 7 months prior to the visit could be visualized at the anterior aspect of the shoulder joint. The patient denied any history of trauma. Arthroscopy of the shoulder joint was performed to rule out the intra-articular nature of the lesion with an exploratory puncture performed to establish the diagnosis with instrumental methods. Delto-pectoral approach was used to remove the neoplasm that was histologically examined to confirm the diagnosis. The upper limb was postoperatively immobilized with a Gilchrist-bandage for 2 weeks. The sutures were removed 2 weeks after the operation. **Result** The patient underwent a course of rehabilitation to restore the function of the upper limb. The upper limb function was preoperatively and postoperatively evaluated with the ASES questionnaire. The patient showed the shoulder function completely regained at two years with ASES scored 100. MRI scan of the shoulder joint showed no signs of recurrence. Extra-articular location of PVNS is very rare and is described in few case reports. Open procedure or arthroscopic removal of the affected synovial membrane are used to treat the condition. **Conclusion** Removal of the affected synovial membrane allowed us to obtain a good clinical result.

Keywords: pigmented villonodular synovitis, synovitis, shoulder joint

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INTRODUCTION

Pigmented villonodular synovitis (PVNS) is uncommon. A benign proliferative process may involve synovial membranes, bursa, tendons, bone tissue, and skin [1, 2]. PVNS of the shoulder joint is very rare with the incidence of 1.4 % (4/272) reported by L. Van der Heijden [3]. Intraarticular types of the disease are more common. L.P. Müller et al. reported 17 intra-articular involvement out of 18 cases [4] and X. Mahieu et al. reported 16 intra-articular cases out of 18 [5]. T.Q. Serra reported PVNS of the shoulder joint and subacromial space involving the bone tissue of the articular surfaces and the acromioclavicular joint [6]. Major symptoms of intra-articular type of the disease are edema and pain and pain at palpation and impaired configuration are typical for extra-articular involvement [7–9]. Open techniques or arthroscopy are used to remove the involved synovium [3, 7, 10-13]. We present a very rare clinical case of extraarticular PVNS of the shoulder joint in a 48-year-old man.

MATERIAL AND METHODS

A 48-year-old patient I. presented with pain and swelling of the left shoulder. The patient reported a deformity at the anterior aspect of the left shoulder 7 months before his visit to the hospital. Physical examination revealed a deformity at the anterior aspect of the left shoulder joint. A positive fluctuation symptom was revealed on palpation. Palpation of the anterior surface of the left shoulder caused discomfort. ROM in the shoulder joint was full with pain at the extreme points. Muscle strength was not reduced. MRI of the left shoulder joint showed thickening of the synovial membrane, hypertrophied villi and hemosiderin inclusions (Fig. 1).

Puncturing of the mass performed during the first visit resulted in evacuation of 20 mL brown liquid without additional impurities and sediment (Fig. 2).

No changes in blood parameters (leukocytes, 6.97 10^9/L; erythrocyte sedimentation rate (ESR), 11 mm/h) were found preoperatively. The patient was offered an open surgical removal of the involved synovial membrane from the subacromial space and arthroscopic treatment of the shoulder to rule out intraarticular lesions. The cavity of the shoulder joint was explored from the posterior arthroscopic approach with the patient in a half-sitting position (Fig. 3).

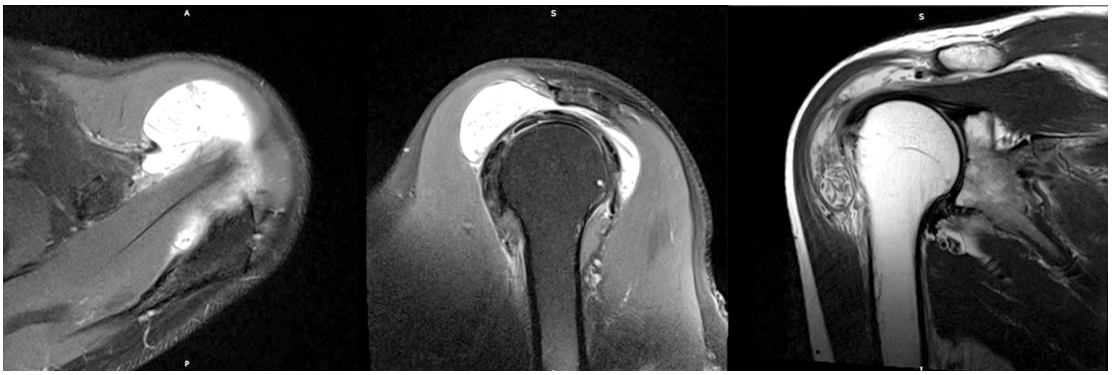

Fig. 1 MRI of the left shoulder joint of patient I. Sections in the coronal, axial and sagittal planes

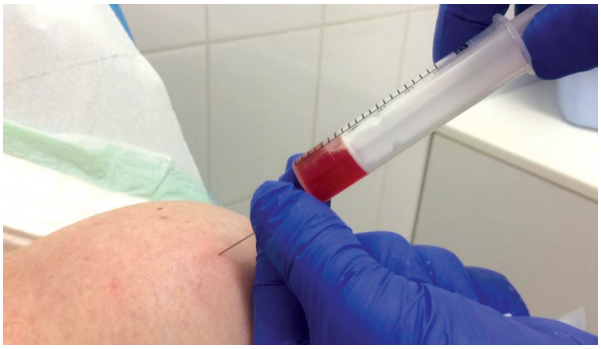

Fig. 2 Performing puncturing of a subcutaneous mass

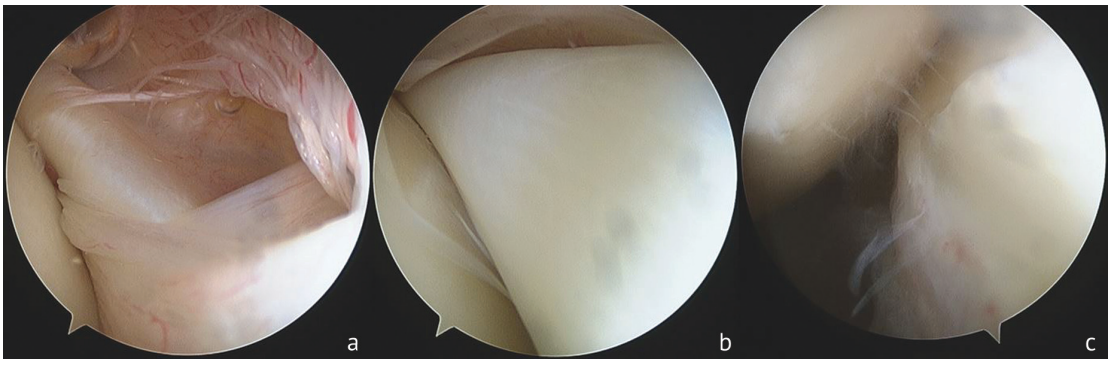

Fig. 3 Arthroscopic exploration of the shoulder joint: (a) subscapularis tendon area; (b) tendon of the long head of the biceps; (c) inferior aspects of the joint. No findings of involved synovial membrane revealed

The delto-pectoral approach was used to visualize the mass. The subacromial sac was filled with brown liquid (Fig. 4). The bursa walls were fused with the anterior portions of the humerus medially and at the acromion. The bursal sac was exposed and removed in a blunt way. The wound was explored and sutured in layers, aseptic bandages applied. The involved synovial membrane was taken for histological examination.

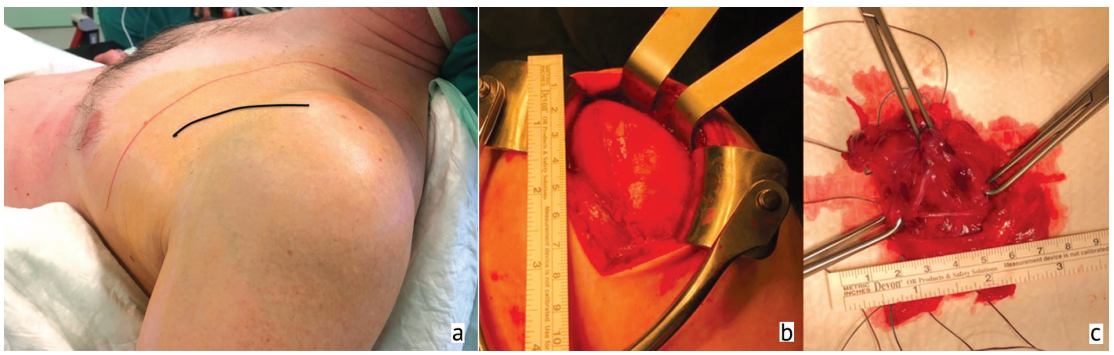

Fig. 4 Stages of the operation: (a) position of the patient on the operating table, the incision indicated with the black line; (b) surgical approach to the mass; (c) a fragment of the mass removed

The patient was discharged from the hospital for outpatient treatment after two days. The upper limb was immobilized with Gilchrist bandage for 2 weeks for better healing of postoperative wounds. The bandage was removed for exercising the shoulder joint. Histological findings confirmed the diagnosis (Fig. 5).

Follow-up examinations were performed at 1.5 and 6 months after the operation with the last produced at 2 years. The ASES grading system was used to evaluate the shoulder joint preoperatively and at the last follow-up. Preoperatively ASES score was 85 points.

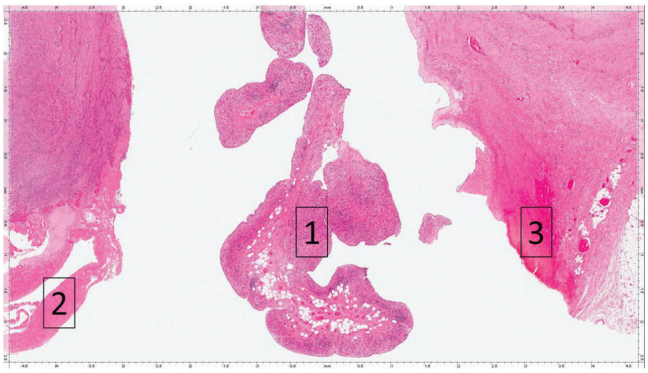

Fig. 5 Histological preparation of the mass removed showing uneven fibrous synovial membrane with papillary hyperplasia (1), foci of fibrinoid necrosis (2) and elements of granulation tissue (3). Stained with hematoxylin and eosin; magnified ×200

RESULTS

The patient reported complete recovery at 2 years. Postoperative scars showed no signs of inflammation and fusion with underlying tissues. The patient had full range of motion and muscle strength was not reduced (Fig. 6). MRI scan showed no evidence of recurrence (Fig. 7). The ASES score was 100 points at 2 years.

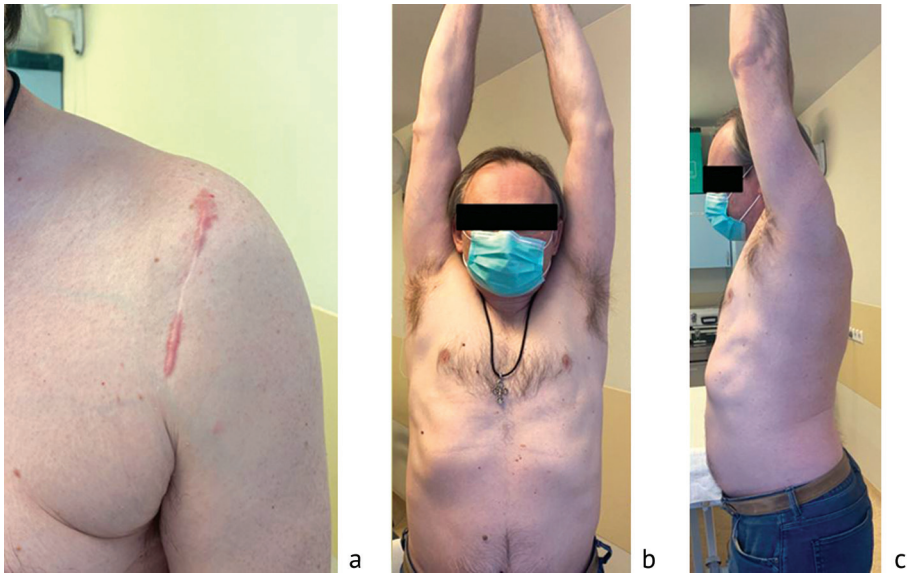

Fig. 6 Photographs of patient I. at two years after the operation showing (a) postoperative scars; (b, c) functional outcome

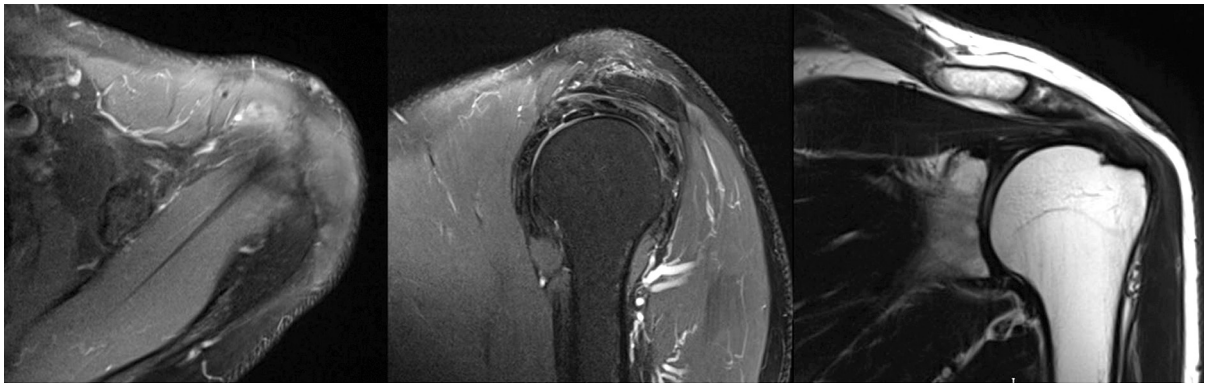

Fig. 7 MRI of the left shoulder of patient I. at two years of surgery. Sections in the coronal, axial and sagittal planes

DISCUSSION

PVNS is a rare benign proliferative disease and uncommon for the shoulder with the incidence of 0.8 % (2/237) reported in a systematic multicenter retrospective analysis of G. Xie et al. [14]. Although the disease is not associated with trauma L.P. Müller et al. reported a history of trauma in 4 out of 18 reviews [4].

The disease developed without an episode of trauma in the history of our case.

PVNS is mostly treated by surgical removal of the involved synovial membrane [10]. The PVNS was localized extraarticularly in our case and removed in an open manner. The shoulder was arthroscopically explored to rule out intraarticular lesions. The recurrence rate of PVNS varies and largely depends on the location and type of the disease. X.Mahieu et al. reported the recurrence of the disease for the knee joint up to 50 % [5]. Lizz van der Heijden et al. performed a crowdsourcing study reporting recurrence rate of 58 % (69/118) after arthroscopic, 36 % (35/97) after open and 50 % (5/10) after combined synovectomy. Local recurrence risk ($p < 0.05$) was higher for diffuse type disease [3].

The recurrence of PVNS of the shoulder joint was analyzed in the study by X. Mahieu et al. The recurrence was reported in 17 % of cases (5/30) in 18 references [5]. Major symptoms of PVNS were pain and swelling with stiffness and limited ROM being less common [3, 11, 13, 14]. Pain and subsequently developed deformity were major complaints in our case. Differential diagnosis of the disease is performed with chondromatosis [16], monoarthritis [17], and other proliferative diseases. G. Xie et al. reported elevated preoperative erythrocyte sedimentation rate (ESR) in 45.83 %, elevated C-reactive protein (CRP) rate in 38.41 % and elevated leukocyte count in 38.4 % of PVNS cases [14]. Blood counts were normal in our clinical case (leukocytes, 6.97 10^9/L; erythrocyte sedimentation rate (ESR), 11 mm/h).

We could find only three published cases with extraarticular PVNS of the shoulder. In 1997 C.J. Sawmiller et al. reported the condition in a 57-year-old woman. The patient was also diagnosed with a rotator cuff tear [18]. In 2008, CH Cho et al. reported the condition in a 21-year-old man [19], and in 2013 the case was described by Portuguese colleagues [20]. As of 2013, this was the first published case of extraarticular PVNS in the Portuguese population and the third in the world.

No additional intraarticular lesions were identified in our clinical instance.

CONCLUSION

PVNS is a rare benign proliferative disease and uncommon for the shoulder. The extraarticular form of the disease is extremely rare and is represented by several clinical cases in the modern literature. PVNS can be treated surgically with open or arthroscopic removal of the involved synovial membrane.

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