

## RECURRENT PIGMENTED VILLONODULAR SYNOVITIS OF KNEE TREATED WITH SURGERY AND ADJUVANT RADIOTHERAPY: A CASE REPORT

Yu-Chang Liu<sup>1</sup>, Yu-Jen Chen<sup>1</sup>, Yung-Chang Lu<sup>2,3</sup>, Chang-Hung Chung<sup>1</sup>

*Department of Radiation Oncology<sup>1</sup>, Department of Orthopaedic Surgery<sup>2</sup>, Mackay Memorial Hospital  
Mackay Junior College of Nursing<sup>3</sup>*

Pigmented villonodular synovitis (PVS) is a rare disease involving the synovial membrane and causing symptoms similar to arthritis. In diffuse PVS, recurrence is not uncommon after surgery. We report a male patient with PVS of the knee that recurred 1 year and 10 months after initial synovectomy. He was again operated on and then given adjuvant radiation therapy (RT) with a total dose of 34.5 Gy in 15 fractions beginning 39 days postoperatively. RT relieved his symptoms even though an MRI revealed persistent disease. RT may be beneficial for selected patients with diffuse PVS.

[ Therapeut Radiol Oncol 2004; 11(1): 55-58 ]

Key words: Pigmented villonodular synovitis, Radiotherapy, Case report

### INTRODUCTION

Pigmented villonodular synovitis (PVS) is a rare disease affecting the synovial membrane of joints or tendon sheaths. The etiology is unclear. Histologically, it is characterized by proliferation of synovial lining cells that may spread along synovial surface and invade surrounding subsynovial connective tissues [8]. Presenting symptoms include pain and swelling of the involved sites. The mainstay of treatment is surgical excision. In its diffuse form, however, recurrence after surgery alone is relatively high, ranging from 16% to 48% [8].

Attempting to reduce recurrence, external beam radiation therapy (EBRT) after partial synovectomy was suggested to be helpful [1]. In the circumstances of recurrent PVS, patients had been successfully salvaged by EBRT alone or combined with surgery [7].

We present a case of recurrent PVS of the knee managed by surgery plus adjuvant EBRT.

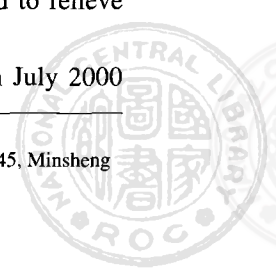
### CASE REPORT

A 24 year-old male came to our hospital in June 2000 complaining of right knee pain and swelling for one month. He had sustained trauma to the knee in February 2000. Initial physical examination showed prominent swelling over the right suprapatellar region with localized warmth. No tenderness was elicited during knee palpation. Right knee joint sonography revealed marked suprapatellar bursitis with synovial fluid accumulation. Joint aspiration yielded blood-tinged brown fluid. Physical therapy and non-steroidal anti-inflammatory drugs (NSAID) were prescribed but failed to relieve symptoms.

Arthroscopy was performed in July 2000

Received: 2003, 10, 27. Accepted: 2003, 12, 27.

Address reprint request to: Yu-Chang Liu M.D., Department of Radiation Oncology, Mackay Memorial Hospital, No.45, Minsheng Rd., Danshuei Township, Taipei County 251, Taiwan (R.O.C.)



and a fibrillated synovial surface was noted (Figure 1). Arthroscopic synovectomy was then performed.

After the synovectomy, he continued to have intermittent right knee pain ameliorated by NSAID and muscle relaxant. He stopped taking analgesic three months after the surgery and remained symptom-free until July 2001, when right knee pain recurred. Contracture of both knee joints (right knee range of motion: 0 to 100 degrees; left knee range of motion: 0 to 110 degrees) was found. The symptoms seemed to be improved by hydrotherapy and range of motion and strengthening exercises, and he remained symptom-free after completion of physical therapy.

However, in April 2002, right knee swelling and pain gradually developed. One week of NSAID treatment produced no relief, so arthroscopy was performed. As suspected, the PVS had recurred, and he was treated with a total synovectomy.

Pathology confirmed the diagnosis of recurrent PVS, and he was referred for postoperative adjuvant radiation. We used 6 MeV radiation for a target volume defined by computed tomography (CT) of the knee in the same position as it would assume on treatment table. Computer-assisted treatment planning (Figure 2) was used for dosimetry and calculating distribution of the dose to cover the target volume, which included overt disease seen on CT image plus a 5 cm longitudinal margin above and below it. The entire knee was covered in the radiation field by using anteroposterior parallel-opposed portals. Radiation was begun 39 days after surgery for a total dose of 34.5 Gy given with a daily fraction size of 2.3 Gy. No acute complications were observed during RT, which was given from May to June, 2002. After completing RT he needed no further NSAID and has remained symptom-free, despite evidence of persistent PVS on magnetic resonance imaging in December 2002.

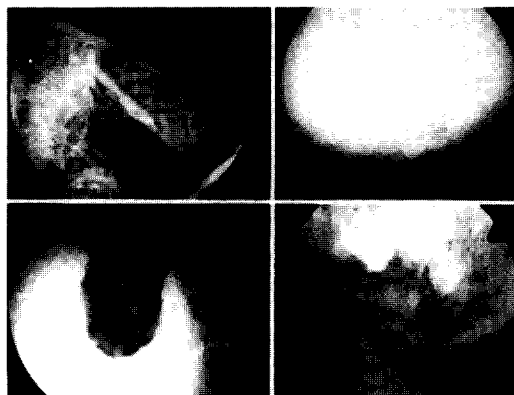


Fig 1. Arthroscopic findings during synovectomy.

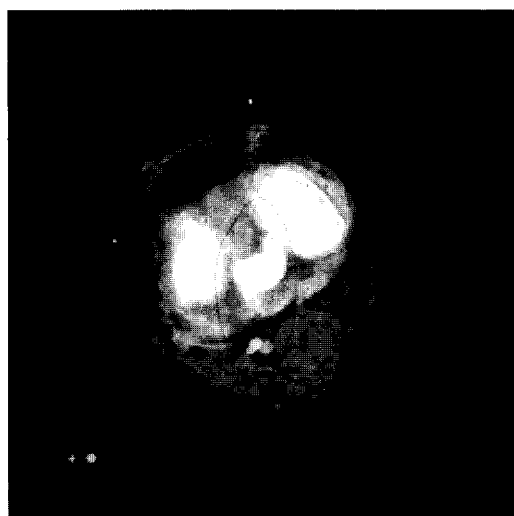


Fig 2. Computer-assisted treatment planning.

## DISCUSSION

The role of RT in treatment of PVS remains uncertain because of the extremely low incidence (1.8 cases per million) [1] of this disease. With such a rare condition, prospective randomized trials are very difficult to organize. Both EBRT [1, 7] and intra-articular instillation of radionuclides, such as yttrium-90 [4, 9], has been used alone or in combination with surgery to treat PVS.

Although available data in the literature is not sufficient to support the routine use of postoperative adjuvant RT for patients with PVS, it seems that this treatment modality may be beneficial in selected patients. In localized PVS,

surgery may be all that is needed due to the disorder's low aggressiveness and recurrence rate [5, 6]. However, diffuse PVS, with or without extraarticular invasion, frequently recurs after synovectomy alone [3, 10]. This is why adjuvant RT has been tried and seems to be beneficial in some cases [1, 4, 7, 9].

In a recent study of diffuse PVS of the knee, O'Sullivan et al. suggested that RT should be considered for patients who experienced extensive local relapse after prior adequate surgical treatment or who have a large burden of residual disease after surgery. In these circumstances, RT would be a very attractive option to salvage surgery, such as amputation, with its significant effect on quality of life [7].

Blanco et al. reported another series of diffuse PVS of the knee. They used partial synovectomy plus adjuvant RT with good functional results and without significant complications. They note that various factors may make complete removal of affected synovial tissue difficult, so that RT may provide an advantage in reducing recurrence of the disease by eliminating residual affected tissue [1]. It would be very helpful if a multicenter clinical trial could be designed to assess the benefits and toxicity of this therapy.

For our patient, treatment portals consisted of anteroposterior parallel-opposed fields. We used a fractionation schedule of 34.5 Gy in 15 daily fractions and CT-assisted treatment planning as suggested by O'Sullivan et al. [7]. A low anti-inflammatory dose of 26 Gy in 1.5 Gy daily fractions has also been used [1]. We used a radiation field margin 5 cm beyond the overtumor or surgical bed, the same as for low-grade sarcoma, again, according to the recommendation from O'Sullivan et al. [7].

MRI is essential for follow-up because the presence of residual disease does not reliably correlate with the clinical findings [2], as was the case with our patient. O'Sullivan et al. found in their series that complete response may be

observed 12 months after completion of RT, with no evidence of disease found on subsequent imaging studies [7]. Therefore, continued follow-up with MRI to observe treatment effect would be wise.

O'Sullivan's group didn't note any long-term complications of RT [7]. Radiation-induced sarcoma has been a concern, particularly in younger patients [6]. However, the risk cannot be quantified since so few patients have been treated with RT.

In conclusion, RT is an effective treatment modality for PVS. With only one patient having persistent disease, all other cases manifested complete response after RT (3 postoperative, 1 preoperative, 9 definitive) in one study of recurrent cases [7]. In patients receiving partial synovectomy and postoperative RT, recurrence rate is similar as those treated with complete synovectomy [1]. Although there is insufficient data to support its routine use in all patients with PVS, it is worth trying in patients with diffuse disease who have not been cured by initial surgery. It remains to be seen if it should be used as adjuvant therapy in all cases of diffuse disease to prevent recurrence.

## REFERENCES

1. Blanco CR, Leon HO, Guthrie TB: Combined partial arthroscopic synovectomy and radiation therapy for diffuse pigmented villonodular synovitis of the knee. *Arthroscopy* 2001; 17: 527-531.
2. Chin KR, Barr SJ, Winalski C, Zurakowski D, Brick GW: Treatment of advanced primary and recurrent diffuse pigmented villonodular synovitis of the knee. *J Bone Joint Surg Am* 2002; 84-A: 2192-2202.
3. Flandry FC, Hughston JC, Jacobson KE, Barrack RL, McCann SB, Kurtz DM: Surgical treatment of diffuse villonodular synovitis of the knee. *Clin Orthop* 1994; 300: 183-192.

4. Kat S, Kutz R, Elbracht T, Weseloh G, Kuwert T: Radiosynovectomy in pigmented villonodular synovitis. *Nuklearmedizin* 2000; 39: 209-213.
5. Kim SJ, Shin SJ, Choi NH, Choo ET: Arthroscopic treatment for localized pigmented villonodular synovitis of the knee. *Clin Orthop* 2000; 379: 224-230.
6. Ogilvie-Harris DJ, McLean J, Zarnett ME: Pigmented villonodular synovitis of the knee. The results of total arthroscopic synovectomy, partial arthroscopic synovectomy, and arthroscopic local excision. *J Bone Joint Surg Am* 1992; 74: 119-123.
7. O'Sullivan B, Cummings B, Catton C, et al.: Outcome following radiation treatment for high-risk pigmented villonodular synovitis. *Int J Radiat Oncol Biol Phys* 1995; 32: 777-786.
8. Rosenberg A: Tumor and tumor-like lesions of joints and related structures. Ruddy S, Harris Jr. ED, Sledge CB, Budd RC, Sergent JS: *Kelly's Textbook of Rheumatology*, 6th Ed., U.S., W.B. Saunders, 2001; 1677-1680.
9. Shabat S, Kollender Y, Merimsky O, et al.: The use of surgery and yttrium 90 in the management of extensive and diffuse pigmented villonodular synovitis of large joints. *Rheumatology* 2002; 41: 1113-1118.
10. Zvijac JE, Lau AC, Hechtman KS, Uribe JW, Tjin-A-Tsoi EW: Arthroscopic treatment of pigmented villonodular synovitis of the knee. *Arthroscopy* 1999; 15: 613-617.

## 手術及輔助性放射治療用於膝關節復發色素絨毛結節狀滑膜炎之治療： 病例報告

劉育昌<sup>1</sup> 陳裕仁<sup>1</sup> 盧永昌<sup>2,3</sup> 鍾昌宏<sup>1</sup>

馬偕紀念醫院 放射腫瘤科<sup>1</sup> 骨科<sup>2</sup> 馬偕護理專科學校<sup>3</sup>

色素絨毛結節狀滑膜炎為一稀有疾病，主要影響關節滑膜並產生類似關節炎的症狀。瀰漫性色素絨毛結節狀滑膜炎在接受手術治療之後，復發的情況並不少見。我們報告一位在初次滑膜切除術後一年又十個月復發的男性病患。他在第二次手術後 39 天開始接受術後的放射線治療，總劑量為 34.5 Gy，以 15 次分次照射給予。放射治療緩解了疾病相關的症狀，但追蹤的核磁共振影像仍有疾病存在的證據。放射治療對某些瀰漫性色素絨毛結節狀滑膜炎病人可能會有治療上的益處。

[ 放射治療與腫瘤學 2004; 11(1): 55-58 ]

關鍵詞：色素絨毛結、節狀滑膜炎、放射治療、病例報告

