Pigmented villonodular synovitis of the elbow in a 6-year-old girl: a case report

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ABSTRACT
Pigmented villonodular synovitis of the elbow is rare and even rarer in children. We report a case of pigmented villonodular synovitis of the elbow in a 6-year-old girl who underwent total synovectomy after the diagnosis was confirmed by biopsy. The osteochondral defect at the olecranon was filled with calcium phosphate bone paste. Two years after surgery, neither recurrence nor joint degeneration was found.

Key words: bone cements; calcium phosphates; child; elbow; synovitis, pigmented villonodular

INTRODUCTION
Pigmented villonodular synovitis (PVNS) was first reported by Jaffe et al.1 in 1941. The knee and hand are the most frequently involved sites, followed by the hip, wrist, ankle, joints of the hands and feet, and shoulder.2,3 Involvement of the elbow is rare4,5; only 21 cases of PVNS of the elbow have been reported.2,3,5-19 Its occurrence in children is even rarer10,13,20,21; only 2 cases have been reported.6,13 Details of the treatment and results of these cases are unknown. We report a 6-year-old girl with PVNS in the left elbow who underwent a total synovectomy. Calcium phosphate bone paste was used to fill the bone defect after curettage.

CASE REPORT
In April 2003, a 6-year-old girl presented with an 8-month history of occasional pain and limited range of movement in her left elbow. Her mother noted progressive stiffness and swelling of the elbow and reported no history of trauma or microtrauma. On physical examination, no local heat or tenderness was present, but apparent swelling was noted. The elbow's range of movement was 120° in flexion, -40° in extension, 70° in pronation, and 90° in supination. A plain radiograph and computed tomography scan showed a round radiolucent defect at the olecranon (Fig. 1). In T1-weighted magnetic resonance images, the lesion showed iso-intensity to the surrounding muscle, and high intensity in T2-weighted images (Fig. 2). The lesion was enhanced heterogeneously by the administration of Gd-DTPA contrast. An arthro-
scopic biopsy of the synovial tissue showed a hypertrophic brown synovium, which on histological evaluation showed haemosiderin-laden macrophages, neovascularisation, inflammatory cells, polynucleotic giant cells, and foam cells. A diagnosis of PVNS was therefore confirmed.

Through a posterolateral approach, the elbow was temporally dislocated, and a nearly total synovectomy of the elbow was performed. The entire elbow cavity was filled with the synovial proliferation. The lateral wall of the olecranon had been destroyed by synovial invasion; the medullary canal of the olecranon was replaced by brown synovial tissue. After a meticulous synovectomy and curettage, the articular surface of the olecranon had an osteochondral defect measuring 6x10 mm. Calcium phosphate bone paste (Biopex, Mitsubishi-Welpharm, Tokyo, Japan) was used to fill in the defect.

Postoperatively, a long-arm splint was applied for 2 weeks; range of movement exercises were allowed after splint removal. Two years after the surgery, the elbow’s range of movement was 120° in flexion, with 20° loss of extension. Radiographs and magneticresonance images showed no recurrence or any degenerative changes (Fig. 3).

**DISCUSSION**

PVNS is classified as a benign but locally progressive synovial proliferative lesion in the joint, bursae, and tendon sheath. PVNS involving the knee and hand is not uncommon but in the elbow it is rare. PVNS involving the elbow of a child is even rarer. Aydingoz et al. reported a 6-year-old girl with PVNS in the elbow joint where the synovial tissue had invaded the olecranon fossa and olecranon, as in our case. However, they did not mention the treatment used to correct the bone defect or any results of treatment.

The principle treatment for PVNS is a complete synovectomy. When PVNS occurs at the tendon sheath, it is relatively easy to remove the entire lesion. Nonetheless, because of the complexity of the joint structure in the knee and elbow joints, complete resection of the synovial tissue in these joints is often difficult. For patients severely affected by bony destruction and arthroplasty, use of radiation along with synovectomy has been reported.

In the present case, however, these treatments...
were difficult to apply because of the patient’s age. After the synovectomy and curettage, a bone defect adjacent to the joint was created. In Japan, allografts are usually not available. An autograft appeared to be an alternative choice for filling the defect, but we decided not to harvest from the iliac bone or knee joint, in order to avoid future deformity at the site of harvest. We instead used calcium phosphate bone paste to fill the defect. The paste has been widely used to fill bony defects in distal radius fractures of the elderly.25,26 There is a possibility that the paste may leak into the joint cavity. Using calcium phosphate on the surface of the joint poses a risk of synovitis, wear of the articular cartilage, and some other adverse reactions. There are few clinical reports concerning reactions against calcium phosphate used adjacent to the joint cavity. In a study on the use of calcium phosphate to fill bony defects in goat knee femoral condyles, the investigators achieved successful coverage of the surface of the defects without any adverse reaction in the joint structures for up to 6 months.27

Two years after surgery, our patient showed no apparent reaction to the calcium phosphate and no radiological changes were observed. Nonetheless, careful management of such a case is mandatory.

REFERENCES