Spontaneous Osteochondroma Resolution in a Young Female: Imaging and Histopathological Findings

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Key Words

Osteochondroma · Spontaneous resolution · Imaging · Histopathology

Abstract

Objective: To report the first operated and histopathologically studied case of spontaneously resolving solitary osteochondroma. **Case Presentation and Identification:** An 18-year-old girl presented with leg pain of 2 months' duration. She had been diagnosed to have a proximal tibial osteochondroma since the age of 9 years. Excision biopsy of the tumor remnant and its surrounding tissue was done after CT and MRI imaging. **Conclusion:** The spontaneous resolution of this case could have been due to simultaneous interruption of blood supply to the tumor and its cartilaginous cap or hyperemia around the tumor.

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Introduction

Osteochondroma is the most common benign bone tumor, representing 15% of all bony tumors according to Pongkrietch and Sirikulchayanonta [1], and 45.3% of benign tumors according to Barbosa et al. [2]. It occurs frequently as a solitary osteocartilaginous exostosis and rarely as hereditary multiple lesions. The most common

sites of occurrence are the long bones of the lower extremity (50%), usually the lower end of the femur and upper end of the tibia [3]. Spontaneous regression of osteochondroma is an extremely rare event [4].

Case Report

An 18-year-old UAE female presented with a 2-month history of pain on the upper third of her right leg. She had been diagnosed to have osteochondroma of the upper right tibia 9 years earlier, but remained asymptomatic. She had no significant diseases in the past, except being detected to have G6PD deficiency. She had no trauma to the tumor and her legs.

Clinical examination revealed a healthy female with normal gait. There was diffuse swelling on the upper end of her right leg on the medial side, which was slightly tender but not warm. No other bony prominences or skeletal abnormalities were detectable elsewhere in her body. Routine blood examinations including biochemistry were normal. Her G6PD screening was normal.

X-ray examination of her knee in December 1999 (age 9 years 7 months) demonstrated a pedunculate long stalked osteochondroma distal to the medial tibial flare, far away from the epiphyseal line with the open tibial growth plate (fig. 1).

An X-ray in October 2000 (age 10 years 5 months) showed the osteochondroma had started to resolve as evidenced by shortening, narrowing tip, moth-eaten irregular borders and permeated stalk. A recent X-ray showed almost complete resolution. An irregular remnant of the tumor was detectable with foci of speckled calcification (fig. 2).

A CT scan revealed a very thin marrow cord with no cortical destruction, no marrow infiltration and no infiltration of the

Fig. 1. December 1999, right knee X-ray: long pedunculate osteochondroma distal to medial tibial flare.

Fig. 2. February 2008, right knee X-ray and close-up: near to complete resolution of the tumor with irregularity and permeation at the base of the tumor and foci of speckled calcification (arrows).

Fig. 3. Intraoperative findings. **a** Crural fascia showing vascular abnormality. **b** Opened crural fascia showing vascular soft tissue covering the tumor.

overlying subcutaneous fat layer. An MRI of the right leg showed focal irregularly shaped lesion on the surface of the posteromedial aspect of the right upper tibia. The lesion showed a low signal pattern at all pulsed sequences with streaks of bright signals of edema within its layers. The overlying subcutaneous fat looked clear and intact with no infiltrations.

In June 2008 (age 18 years) she was operated. The tumor was approached by vertical incision medially after its location was confirmed by fluoroscopic images. The crural fascia showed abnormal vascularity (fig. 3a). Below the crural fascia there was vascular soft tissue covering the tumor (fig. 3b). The tumor remnant did not show any cartilaginous cap and its surface was generally smooth. The soft tissue and the bony tumor were excised. Histopathological examination of the soft tissue covering the tumor showed fibrofatty tissue with areas of collagenized fibrosis and foci of increased vascularity with thick muscular small arteries, arterioles and veins, without evidence of interconnections or arteriovenous malformations. Smaller capillary vessels also were detected (fig. 4). The bony tumor revealed viable bony trabeculae with viable osteocytes without prominent osteoblastic rimming or osteoclastic activity. The intertrabecular spaces were filled with mature adipose

tissue. No hematopoietic activity was seen. There were foci of dystrophic calcification with no evidence of hemosiderin deposition. Also there was no mature cartilage in the examined specimen. There was no sign of tumoral lesions or malignant transformation in the examined bony specimen (fig. 5). Routine cultures of the excised bone and soft tissue were negative and no acid-fast bacilli were detected. The patient's pain and swelling disappeared after operation and her 15 months' postoperative follow-up did not reveal any more detectable abnormalities.

Discussion

Osteochondromas are primary bone tumors, which are usually located in the distal femur, upper tibia or upper humerus. Although the exact etiology of the tumor is not known, a peripheral portion of the physis is thought to herniate from the growth plate. This metaplastic cartilage grows to form the exostosis, which is connected to

Fig. 4. Photomicrograph of the soft tissue covering the tumor showing fibrofatty tissue and areas of collagenized fibrosis (arrow) and foci of increased vascularity (black arrowhead). Inset: Thick muscular artery (white arrowhead). HE. Original magnification $\times 40$.

Fig. 5. Microscopic view of the bony tumor showing pink bony trabeculae with viable osteocytes with no cartilaginous cap, no hemosiderin deposits and no osteoblastic rimming or osteoclastic activity. Intertrabecular spaces filled with mature adipose tissue (arrow) showing no hematopoietic activity. Foci of dark blue dystrophic calcification (arrowhead) seen. HE. Original magnification $\times 100$.

the bone by a thin stalk [5]. Spontaneous regression of osteochondroma is very rare. Although the first description of spontaneous regression of the osteochondroma was in 1835 by Hunter [6], Song [7] reported the 12th case in 2000. Five more cases have been reported from 2000 to 2007 [4, 8, 9] and our case is the 18th. Claikens et al. [10] reviewed 10 cases of reported resolving osteochondromas, 7 of sessile type and 3 pedunculate, all regressed before skeletal maturity.

Five hypotheses have been proposed to explain the spontaneous regression of osteochondroma. Copeland et al. [11] suggested that spontaneous regression of skeletal osteochondromas may be due to cessation of cap growth followed by active resorption. They argued that a traumatic lesion such as a fracture may stop the growth of the osteochondroma by injury to the cartilaginous cap, vascular supply disruption, or stimulus of the reparative process in the adjacent bone. Paling [12] reported spontaneous regression of a solitary osteochondroma that apparently resulted from cessation of growth of the tumor prior to skeletal maturation, with subsequent incorporation of the lesion into the enlarging bony metaphysis. Song [7] reported a case of osteochondromatosis of the radius and ulna regressing after ulnar lengthening. He hypothesized the change of mechanical strain or stress after lengthening as the possible mechanism for resolution. Choi et al. [8] reported a case of osteochondroma disappearance re-

sulting from an accompanying progressively enlarging pseudoaneurism. They attributed the resorption to pressure erosion of the pseudoaneurism on the tumor.

Serial X-rays of our case showed spontaneous resolution of the osteochondroma starting at age 10 with no signs of incorporation of the tumor in the growing metaphysis. The tibial growth plate was still open after resolution of the tumor with no accompanying pseudoaneurism, no history of trauma and no prior operations. Lack of a cartilaginous cap, permeation of the stalk, foci of dystrophic calcification, and hypervascularity of the covering soft tissue all suggest an active resolution-repair mechanism of the tumor. While the initiating factor is not clear to us, simultaneous interruption of the blood supply to the tumor and its cartilaginous cap seems to be a probable cause for the resolution. Another possibility is an unknown mechanism triggering hyperemia around the tumor, resulting in spontaneous resolution.

Conclusion

A case of spontaneously resolved proximal tibial osteochondroma is presented. The resolution could have been due to simultaneous interruption of blood supply to the tumor and its cartilaginous cap or hyperemia around the tumor.

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