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CASE REPORT

Heterotopic ossification in the anterior maxilla: a case report and review of the literature

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A 13-year-old boy had complained of an asymptomatic swelling in the anterior maxilla for approximately 4 years. The patient reported no local trauma. The intra-oral examination revealed an exophytic lesion in the incisive papilla between the maxillary central and lateral incisor teeth. The radiographies detected no significant findings. Histopathologically, the lesion showed a dense fibrous tissue above the overlying mucosa. Bone ossification lay beneath a partially hypertrophic cartilage showing occasionally pleomorphic chondrocytes. Because of its microscopic aspects, heterotopic ossification may be mistaken for chondrosarcoma or other conditions involving periosteum, such as parosteal osteosarcoma. A case of heterotopic ossification in the anterior maxilla is presented, and clinicopathologic similarities with other osteochondromatous lesions are discussed.
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Case description

A 13-year-old boy had complained of an asymptomatic swelling in the anterior maxilla for approximately 4 years. On intra-oral examination, an exophytic lesion was seen in the incisive papilla between the maxillary central and right lateral incisor teeth. The lesion presented firm to palpation with a well-circumscribed border extending through the gingival insert, 2 cm in diameter. The overlying mucosa presented unremarkable. The patient reported no history of trauma. No significant radiographic findings were detected by panoramic and periapical radiographies. An excisional surgical approach was performed under local anesthesia. The underlying alveolar bone was compressed and no connection between the lesion and the underlying bone was reported. Grossly, the lesion exhibited a dense, solid mass after sectioning. Histopathologically, a thick, dense fibrous tissue with a cartilaginous cap (Fig. 1, arrow) was located just below the oral mucosa. A trabecular bone of normal appearance (Fig. 1, star) lay under the cartilaginous cap, in a well-vascularized spindle cell stroma. Cartilage caps were organized in lobules, separated by dense fibrous tissue (Fig. 2, square). A gradual transition from fibrous to osseous tissue (Fig. 3, star) and cartilaginous tissue (Fig. 3, arrow) was characteristic. Beneath the cartilage lay matured bone trabeculae of normal appearance (Fig. 4). The cartilaginous cap presented hypercellular with pleomorphic chondrocytes arranged irregularly (Fig. 5, arrow). The patient is free from disease after 2 years of follow up.

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Comments

An uncommon case of heterotopic ossification in the anterior maxilla is presented. The arrangement and maturation are important histopathologic features for differential diagnosis from other osteochondromatous processes (1). Bizarre parosteal osteochondromatous proliferation (BPOP) is considered to be a late stage of heterotopic ossification (1). A calcified cartilage matrix is usually identified at the interface of cartilage and bone, characterized by blue staining with hematoxylin ('blue bone'; 2). The bony trabeculae are apparently formed by a process of endochondral ossification. The cartilage is hypercellular and hypertrophic, with moderately enlarged nuclei (2). In the present case, the cartilage cap was hypocellular, without endochondral ossification, and no cartilage calcification ('blue bone') was found.

The fibro-osseous component shown in heterotopic ossification may resemble osteochondroma. The cartilage cap of osteochondroma grows from the periosteum of the underlying bone, becoming a well-formed trabeculae bone (3). The underlying bone trabeculae in

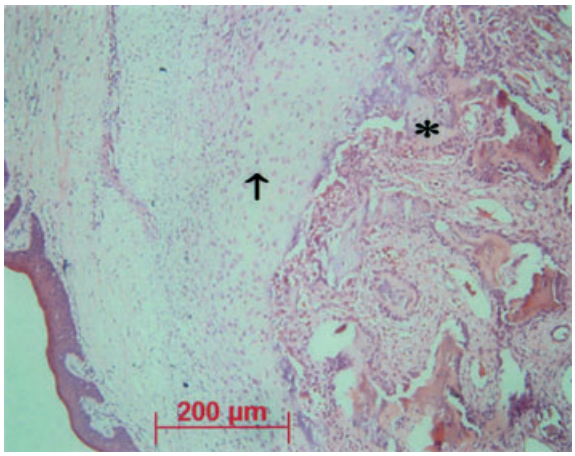

Figure 1 Osteochondromatous proliferation laying just below oral mucosa showing a cartilage cap (arrow) with enchondral ossification at edges of lesion (star) surrounding by a fibrous tissue (hematoxylin and eosin).

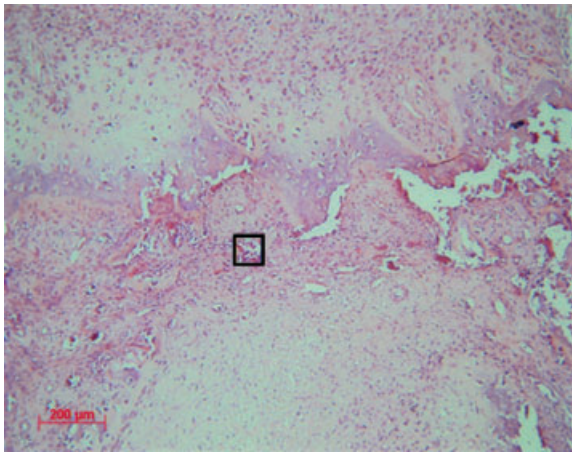

Figure 2 Osteochondroid tissue distributed in lobules and separated by dense fibrous tissue (square; hematoxylin and eosin).

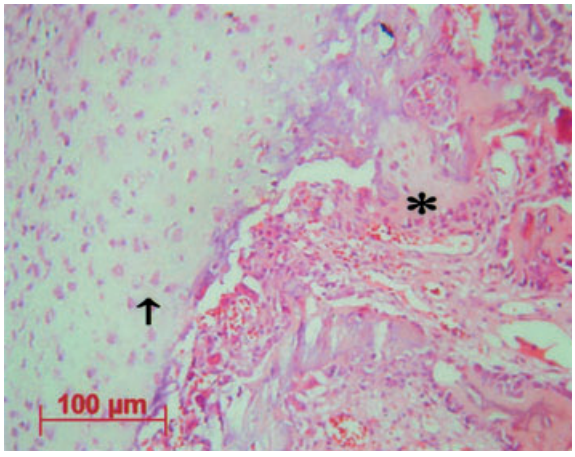

Figure 3 Chondroid cap and cancellous bone in a vascularized spindle cell stroma. Irregularly arranged, pleomorphic chondrocytes resembling chondrosarcoma (arrow). At bottom, bony trabeculae in a gradual transition to cartilage with a benign nature of maturation (star; hematoxylin and eosin).

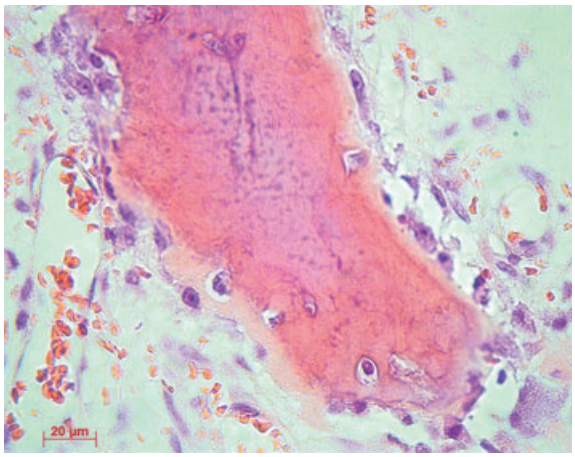

Figure 4 Well-organized bony trabeculae with well-vascularized bone and spindle cell stroma (hematoxylin–eosin).

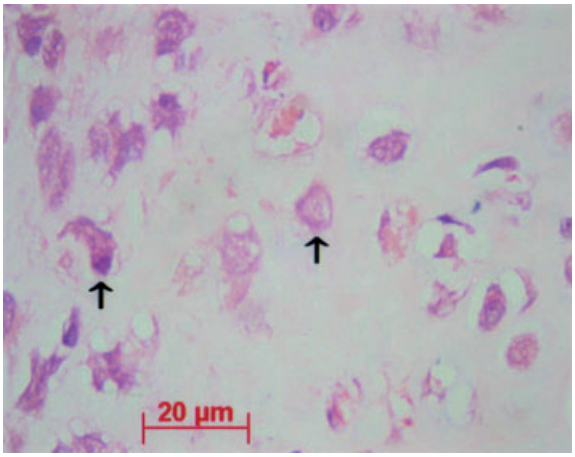

Figure 5 High-power view of chondroid proliferation showing hypercellularity and chondrocytes with moderately enlarged nuclei (arrow; hematoxylin and eosin).

heterotopic ossification are much more regular. Unlike that of heterotopic ossification, the cartilage component of osteochondroma has an orderly arrangement, is hypocellular, and the chondrocytes are usually arranged in parallel rows (3). In addition, the cancellous bone and hematopoietic marrow in osteochondroma are shared with the bone host (3).

Hypercellularity and pleomorphism in the cartilage cap, as well as a loose cell arrangement and organization, may simulate periosteal osteosarcoma, chondroblastic osteosarcoma, and chondrosarcoma. Unlike heterotopic ossification, parosteal osteosarcoma shows attachment to the cortex. Microscopically, it presents by forming a cartilaginous cap with well-formed bony trabeculae (3). The cartilage is hypercellular, organized in lobules with endochondral ossification (3). The narrow islands of osteoid may undergo maturation accompanied by a 'columnar' arrangement toward the base (1).

In osteosarcoma-producing cartilage, a fine connection between bony trabeculae and cartilage is present. Unlike heterotopic ossification, there is no gradual transition from fibrous to osseous and cartilaginous tissue. Indeed, atypical bone formation with binucleated osteocytes is seen. The chondroid lobules show bone formation in the center (3).

The slight atypia seen in low-grade chondrosarcoma is not sufficient for differential diagnosis, as cellularity and cytologic atypia are also seen in heterotopic ossification. Chondrosarcoma tends to permeate the surrounding bony trabeculae (4). The loose arrangement of chondrocytes seen in heterotopic ossification is also characteristic in chondrosarcoma (4).

An uncommon case of heterotopic ossification in the anterior maxilla is presented. Because of microscopic similarities to other osteochondromatous lesions, a meticulous clinicopathologic study is essential. The differential diagnosis includes clinical presentation and roentgenographic findings. Microscopic peculiarities, such as calcified cartilage matrix ('blue bone'), cellularity and/or atypia of chondrocytes, bone maturation and the stroma, are important points of distinction.

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