Osteochondroma of the Hyoid Bone

Blake S Raggio, MD[1]; Samuel Ficenec, B.S.[2]; Thomas C. Flowers, B.S.[2]; Claire Lawlor, MD[1]; Kimsey Rodriguez, MD[3]

[1]Tulane University Medical Center, Department of Otolaryngology; [2]Tulane University School of Medicine;
[3]Ochsner Health System, New Orleans, Department of Pediatric Otolaryngology

ABSTRACT

Background: Osteochondroma is the most common benign bone tumor but rarely occurs in the head and neck.
We report the second case of an osteochondroma arising from the hyoid bone, and the first hyoid osteochondroma in a patient with a history of multiple osteochondroma (MO).

Method: Describe the clinical presentation, diagnosis, and management of a patient with osteochondroma of the hyoid and review the literature.

Results: A 17-year-old-male with a history of MO presented with a 1-month history of a non-enlarging neck mass with associated neck pain and dysphagia. The patient reported a history of multiple surgeries to remove osteochondromas from various locations of the axial skeleton but denied prior involvement of the head and neck. Physical exam revealed a 1.5 cm firm, non-tender, midline neck mass fixed to the hyoid. Lateral plain film showed a smoothly corticated lesion on the hyoid's anterior surface. A diagnosis of osteochondroma was presumed, and removal was planned. Intraoperatively, a 1 cm calcified lesion pedicled to the hyoid body was excised en bloc with a 1.5 cm segment of adjacent bone. Histopathology confirmed the diagnosis of osteochondroma. The patient exhibited complete resolution of symptoms on follow-up. Review of the English literature revealed one reported case of solitary osteochondroma of the hyoid.

Conclusion: Osteochondroma is a benign bony tumor that rarely occurs in the head and neck. We present the second reported case of osteochondroma arising from the hyoid and discuss the characteristic clinical and radiologic findings which should alert the physician to its successful diagnosis and management.

CONTACT

Blake S. Raggio, MD
1415 Tulane Ave, SL-59
New Orleans, LA 70112
Phone: (504) 988-5454
Fax: (504) 988-1694
braggio@Tulane.edu

INTRODUCTION

Osteochondromas, the most common benign bone tumors, are cartilaginous neoplasms of unknown origin with rare malignant potential.[1,2] Osteochondromas present as single (solitary osteochondroma) or multiple lesions (multiple osteochondroma) and mostly affect adolescents and young adults with disputed male sex predilection.[1,3] Osteochondromas commonly arise from the lower limbs of the axial skeleton but rarely occur in the head and neck. Diagnosis relies on a combination of clinical, radiological, and histological criteria, and excision is often curative.[1-4] Osteochondroma represents a rare diagnosis to include in the differential of any midline neck mass.

We describe the first reported case of hyoid osteochondroma in a patient with multiple osteochondroma (MO), discuss its surgical management, and perform a brief review of the literature.

CASE REPORT

A 17-year-old African-American male with a history of MO presented with a 1-month history of a non-enlarging neck mass with associated neck pain and dysphagia. The patient reported a history of multiple surgeries to remove osteochondromas from various locations of the axial skeleton, but he denied prior involvement of the head and neck. Physical exam revealed a 1.5 cm firm, non-tender, midline neck mass fixed to the hyoid. Lateral plain film showed a smoothly corticated and flat lesion on the hyoid's anterior surface (Figure 1). Given the patient's history of MO, a diagnosis of osteochondroma was presumed and surgical excision was planned. Intraoperatively, a 1 cm calcified lesion pedicled to the hyoid body was excised en bloc with a 1.5 cm segment of adjacent bone (Figure 2). Histopathology confirmed the diagnosis of osteochondroma. The patient exhibited complete resolution of symptoms on follow-up without signs of recurrence.

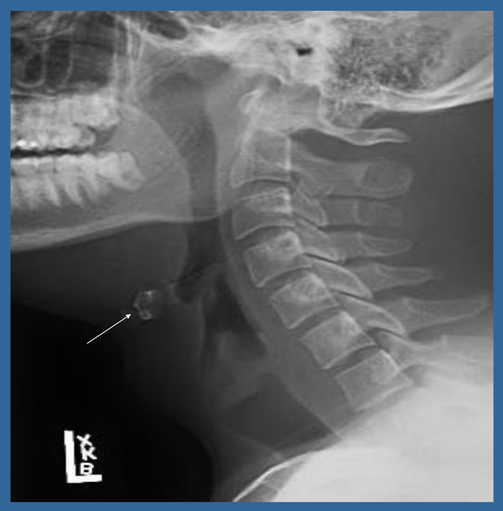

Figure 1. Lateral plain film depicting a smoothly corticated lesion on the hyoid's anterior surface (arrow).

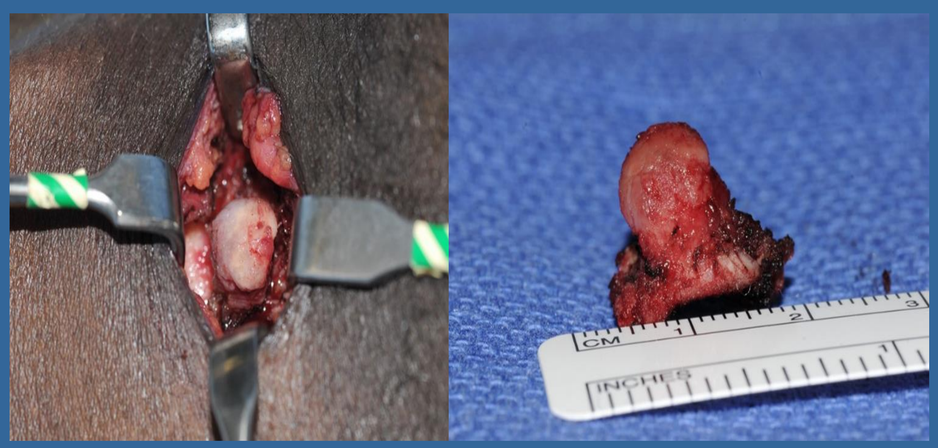

Figure 2. Intraoperative depiction of osteochondroma attached to hyoid bone (Left). Gross depiction of osteochondroma excised en bloc with surrounding hyoid bone (Right).

DISCUSSION

Osteochondromas are cartilage capped bony projections continuous with that of the underlying bone.[1] They present two distinct forms including solitary osteochondroma (SO) and multiple osteochondroma (MO), also known as hereditary multiple exostoses.[1,2]

Epidemiology and Pathogenesis
Osteochondroma may be the most common bone tumor[1,2]; however, the reported incidence of SO is likely underestimated as the majority go clinically undetected.[5] MO is an autosomal dominant disorder that occurs in approximately 15% of patients with osteochondroma. Both SO and MO usually present before age 30 and may be more common in males.[1-3] The cause of osteochondroma formation is unknown, though several theories exist.[1]

Genetics
Both SO and MO are true neoplasms derived from mutation in a cartilaginous cell of the growth plate.[1] Mutations in either of three genes involved in heparin sulfate biosynthesis, *EXT1-3*, contribute to tumorigenesis by altering endochondral ossification.[1-4,6]

Sites of Involvement
Osteochondromas generally arise in bones pre-formed by cartilage, the most common being the bones of the lower limbs.[1,3] Osteochondroma rarely occurs in the head and neck region, with only one other case of hyoid osteochondroma reported in the English literature.[3] Our case is unique in that it is the first osteochondroma of the hyoid in a patient with MO.

Clinical Presentation
Osteochondroma grows slowly until skeletal maturity is reached.[2] When lesions become clinically evident, they most commonly present as a chronic firm mass with symptoms reflecting the size and location of the lesion.[1] Malignant transformation is rare (1-5%)[1,2] and should be suspected in lesions with increasing pain or size.[1] With that being said, most osteochondromas are asymptomatic and discovered incidentally.[2]

Diagnosis
SO and MO clinically may mimic a number of lesions, and the differential diagnosis varies from benign to malignant lesions. A combination of clinical, radiological, and histological criteria should help distinguish osteochondroma from other entities.[3]

*Macroscopy: O*steochondromas appear sessile or pedunculated with a characteristic cartilaginous cap which is usually thin and covered by a thin fibrous outer layer continuous with the periosteum of the underlying bone.[1-3]

Microscopy: Osteochondromas have three distinct layers: a thin fibrous perichondrium, hyaline cartilage cap, and mature lamellar bone with marrow elements.[1,2] Eosinophilic, periodic acid-Schiff stain positive inclusions can be seen in the cytoplasm of the cartilaginous cells.[3]

Imaging: Osteochondromas typically appear as pedunculated or sessile lesions with or without cartilaginous calcifications. Characteristically, the cortex of the bony protuberance is continuous with the cortex of the underlying bone. This relationship may not be evident on simple radiographs alone; therefore, computed tomography (CT) scan or magnetic resonance imaging (MRI) are used to show the characteristic continuity of cortical and spongy bone inside the lesion and helps predict the thickness of the cartilage cap.[3,4]

Treatment and Prognosis
Definitive treatment for symptomatic or cosmetically unappealing osteochondromas is complete surgical excision. Asymptomatic osteochondromas should be surveilled with imaging and clinical exams and do not require treatment unless one suspects malignant transformation. [1,4] Features concerning for sarcomatous transformation include pain, increasing size, cartilaginous cap with a thickness equal to or greater than 1.5 to 2 cm, growth disturbance, and new onset of symptoms.[2,3] Malignant degeneration to chondrosarcoma generally is treated by means of broad surgical resection, with radiotherapy and chemotherapy reserved for cases of poorly differentiated tumors.[4,5]

Excision of the osteochondroma is usually curative. Recurrence is rare (2%-5.3%)[5] and seen in cases of incomplete removal; however, recurrence in a well excised lesion should raise the suspicion of malignancy.[4]

CONCLUSION

We report the second of an osteochondroma arising from the hyoid bone, and the first hyoid osteochondroma in a patient with MO. Although osteochondromas rarely present in the head and neck, otolaryngologists should be aware of their characteristic clinical, radiological, and histological features and their appropriate management.

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