CASE REPORT

Osteoblastoma of the Hypopharynx: A Case Report

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Abstract

Context: Osteoblastoma is a benign, slow-growing, but locally aggressive bone tumor. It is usually localized in the cervical vertebra but may rarely involve the laryngeal cartilage.

Case description: We report a case of osteoblastoma hypopharynx in a 10-year-old boy who presented with snoring and mouth breathing. His video laryngoscopic examination showed a mass in the pyriform fossa, which could not be excised by endoscopic excision technique and hence, was referred to our center for external approach excision. The whole mass was excised en bloc via an external approach, and a Ryles tube was inserted for 10 days. Histopathological examination (HPE) report came as Osteoblastoma. Ryles tube was removed after doing a fluoroscopy to confirm normal swallowing.

Conclusion: Osteoblastoma hypopharynx which has never been reported before in the literature can be treated completely by surgical excision via an open approach.

Keywords: Case report, Histopathology, Hypopharynx, Larynx, Osteoblastoma, Osteoid osteoma, Pyriform fossa.

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Introduction

Osteoblastoma is a benign bone tumor that is closely related to osteoid osteoma. Henry Jaffe and Leo Mayer first described osteoblastoma as an "osteoblastic osteoid tissue-forming tumor" in 1932.[1,2] Lichtenstein later termed the lesion as an "osteogenic fibroma of bone" in his original description of osteoblastoma.[3,4] It was initially described as a "giant osteoid osteoma" by Dahlin and Johnson Jr due to its histologic similarity to osteoid osteoma.[5] The term osteoblastoma was later coined by Lichtenstein and Jaffe in their detailed independent reports of this neoplasm[3,6] in 1956. Osteoblastoma differs from osteoid osteoma in being larger than 2 cm in diameter and its aggressive behavior in bone (Fig. 1).

We report a case of osteoblastoma hypopharynx and describe the clinical presentation, radiological findings, and successful outcome after surgical excision of the lesion.

Case Description

A 10-year-old boy presented with complaints of snoring and mouth breathing for the duration of the past 11 months. There was no history of difficulty in swallowing or voice change. On examination, the voice was normal, with no noisy breathing.

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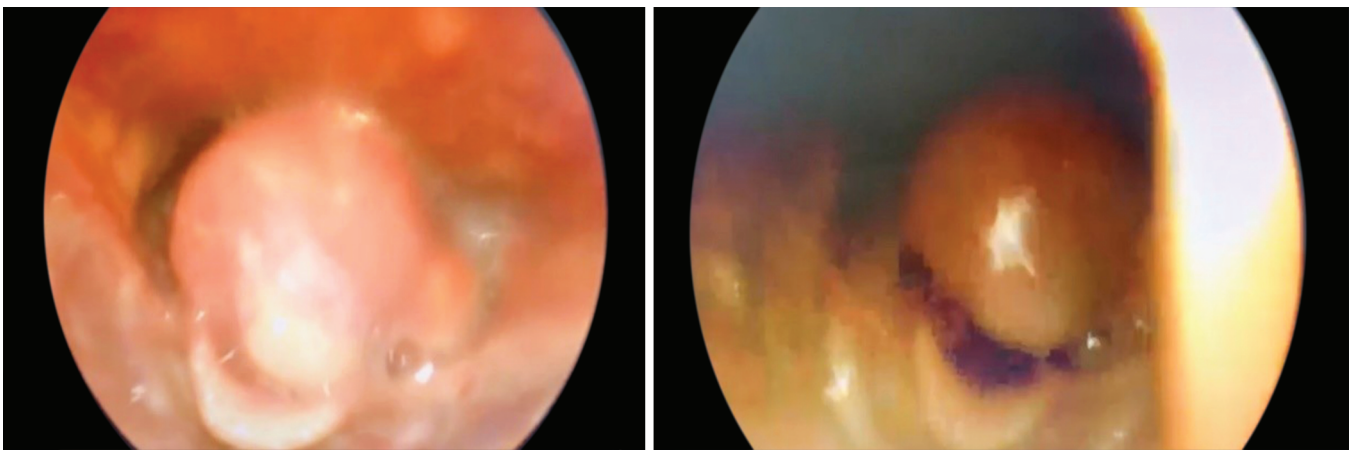

Fig. 1: Videolaryngoscopy (VLS) showing hard stony mass arising from the left pyriform fossa

On auscultation, no stridor and the act of swallowing was normal. The neck examination was normal. Videolaryngoscopy (VLS) examination revealed a smooth protruding round-shaped mass occupying the left pyriform fossa extending to the left side of the larynx, obscuring the view of the larynx except for the epiglottis and anterior part of the right aryepiglottic fold. Hence, the vocal cord mobility could not be appreciated. However, since the voice was normal, mobility was assumed to be normal. There was no significant pooling. Contrast-enhanced computed tomography (CECT) neck was done, which showed a heterogeneous lesion with an area of dense calcification and fat density arising from the left posterolateral wall of the hypopharynx. Endoscopic excision of the mass was attempted elsewhere. However, it was found to be tightly adherent and could not be removed. A biopsy was attempted, but the mass was found to be bony and hard on palpation. Hence, adequate specimens could not be obtained for histopathological examination. The patient was referred for complete excision of the mass (Fig. 2).

Informed consent was taken for mass excision by an external approach. Consent was also obtained for postoperative nasogastric tube feeding. The possibility of the need for a tracheostomy was also explained. The whole mass was excised by an external approach. Awake intubation was not attempted, considering the patient's age. It was decided to do the orotracheal intubation under video guidance with the help of a C-MAC laryngoscope, and the surgeon was ready to do an emergency cricothyrotomy in case of failed intubation. After successful orotracheal intubation under GA, the patient was positioned with neck extension, and the head was stabilized by a head ring. No. 14 F size Ryles tube was inserted, and the position was confirmed. A horizontal skin crease incision was placed at the level of the lower border of the thyroid cartilage toward the left of the midline. Subplatysmal flaps elevated. The left sternohyoid and sternothyroid muscles are cut to expose the left thyroid. The thyropharyngeus stripped off the oblique line of the thyroid cartilage, and the lateral border of the thyroid cartilage was exposed. The lateral border of the thyroid cartilage was retracted with a double hook to expose the left pyriform fossa. A bony mass was palpated in the left pyriform fossa, and dissection was carried out to expose the bony mass. A small area of mucosa was found to adhere to the mass, and sharp dissection was used to deliver the mass. As a result, on excision, a small rent occurred in this area of adherence, which was sutured in layers, after which tissue glue was applied. The excised whole mass was then sent for histopathological examination (HPE). Ryles tube was retained for postoperative feeding. Minivac suction drain placed. Postoperatively, the patient was treated with antibiotics, analgesics, antiemetics, and proton pump inhibitors. Ryles tube feeding was started on the next day. The drain was removed after 48 hours. Fluoroscopy was done after 10 days, and there was no leak or fistula at the site of repair. Hence, the Ryles tube was removed, and oral feeds were started (Fig. 3).

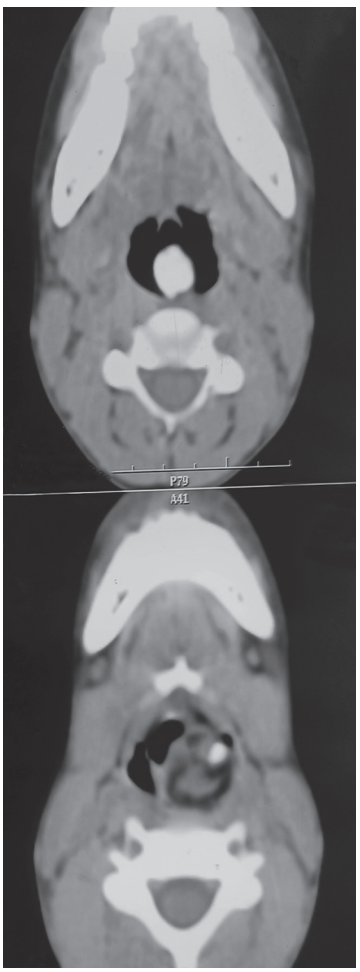

Fig. 2: Contrast-enhanced computed tomography (CECT) showing heterogeneous lesion with an area of dense calcification and fat density arising from the left posterolateral wall of the hypopharynx

Discussion

Osteoid osteomas and osteoblastomas are benign bone-producing tumors with identical histologic features but differ in size, sites of origin and symptoms.[7]

Histopathologically, osteoid osteoma and osteoblastoma are round-to-oval masses of hemorrhagic gritty tan tissue that are well circumscribed and composed of randomly interconnecting trabeculae of woven bone that are prominently rimmed by a single layer of osteoblasts. The stroma surrounding the neoplastic bone consists of loose connective tissue that contains many dilated and congested capillaries. The relatively small size, well-defined margins, and benign cytologic features of neoplastic osteoblasts help distinguish these tumors from osteosarcomas. Osteoid osteomas elicit the formation of a lot of reactive bone, which encircles the lesion. The nidus (which is the actual neoplasm) manifests radiographically as a small round lucency that may be centrally mineralized[7] (Fig. 4).

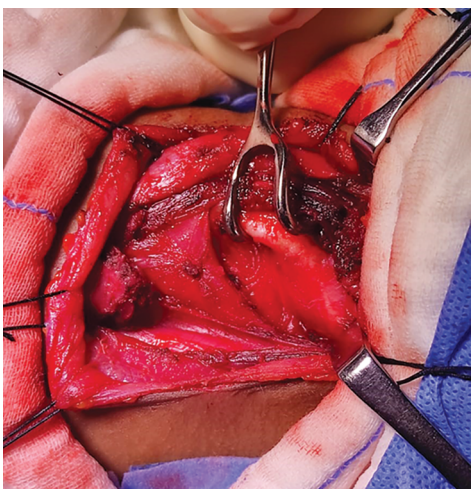

Fig. 3: Double hook placed over the thyroid cartilage ala to retract and expose the left pyriform fossa; the cut end of the strap muscles can be seen

The current case fulfilled the histologic criteria for osteoid osteoma/osteoblastoma. However, the lesion being >2 cm in size, the absence of severe pain and the minor reactive bone component favored the diagnosis of osteoblastoma.

Osteoblastomas are bone-forming tumors that account for less than 1% of all primary bone tumors.[1,8–10] Patients typically present around the second to third decades of life. Male: female ratio is 2 : 1.[6] Diagnostic criteria are according to the WHO classification of soft tissue and bone tumors (5th edition)[11]:

Essential features include:

- Lytic bone tumor >2 cm in size on imaging.
- Well-defined tumor borders.
- No evidence of permeation of the host bone.
- Histological evidence of a bone-forming tumor consisting of trabeculae of remodeled woven bone framed by plump osteoblasts in a vascularized background.

Pathogenesis

Rearrangement of the *FOS* gene is seen in 90% of the cases (like osteoid osteoma).[8]

Location

The spine, especially the neural arch (posterior elements), is the most frequently affected site in more than one-third of cases. Other sites include the pelvis, the limbs (femur and tibia), the jaws, and the other craniofacial bones.

Osteoblastoma hypopharynx has not been reported in the literature. However, osteoblastoma larynx has been reported in thyroid cartilage and arytenoid cartilage. In this case, the mass was seen in the lateral aspect of the aryepiglottic fold and extended to the pyriform fossa. It is possible that it had its origin from the arytenoid cartilage because no other cartilage was seen nearby. (Fig. 5)

In contrast, the lower extremities are the most common site of osteoid osteoma. The intertrochanteric or intracapsular region of the hip is affected in two-thirds of cases.[12,13] It is also seen in the diaphyseal part of the tibia and the humerus. In 50–60% of the cases, osteoid osteoma is seen in the femur and tibia.[14,15]

Around 7–20% of osteoid osteoma is seen to involve the spine. It may manifest as painful scoliosis, though painless conditions also can occur. About 50% of lesions occur in the cervical spine, and about 78% of osteoid osteomas in the lumbar spine are associated with scoliosis[16–19] (Fig. 6).

Clinical Features

Osteoblastomas present with insidious onset of dull pain, worse at night, with minimal response to salicylates, unlike osteoid osteoma.[8] Neurological symptoms may be seen in spinal lesions. Systemic symptoms, including fever, weight loss and hyperdynamic circulation, may be present. This is attributed to an exaggerated immune response to the tumor (Fig. 7).

Radiological Features

- Lesions are predominantly lytic with a rim of reactive sclerosis.
- Expansive with a bubbly appearance.
- Internal calcifications present.
- Associated soft tissue mass may be present.
- A rapid increase in size with associated cortical expansion in the majority of patients, sometimes with cortical destruction.
- Surrounding sclerosis or periostitis in up to 50%.
- Secondary aneurysmal bone cyst-like changes are in 20%.

Histopathology

Macroscopically, osteoblastomas are well-defined tumors with osseous expansion with thinning of the cortex rimmed by sclerotic host bone.[8,9]

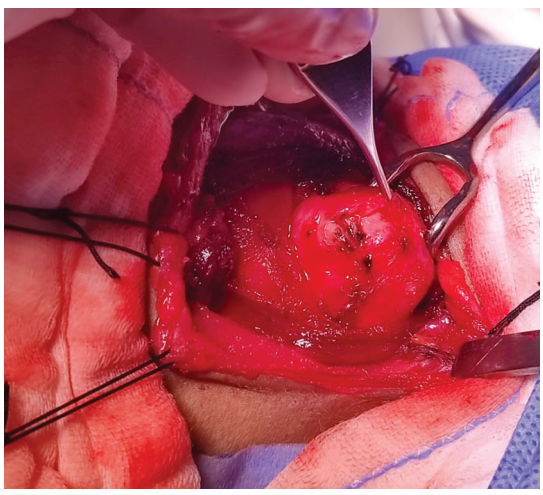

Fig. 4: Mass in the left pyriform fossa

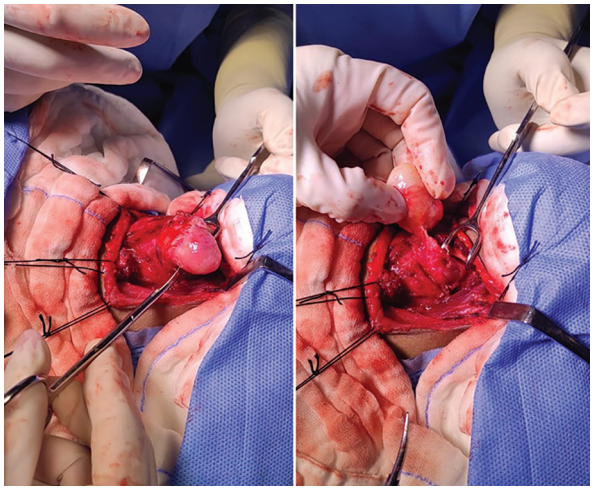

Fig. 5: Mass was seen as tightly adherent to the left pyriform fossa mucosa

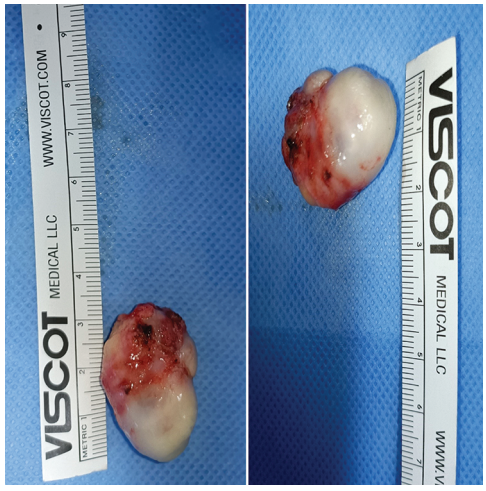

Fig. 6: The excised mass was about 3.4 × 2.3 cm in size

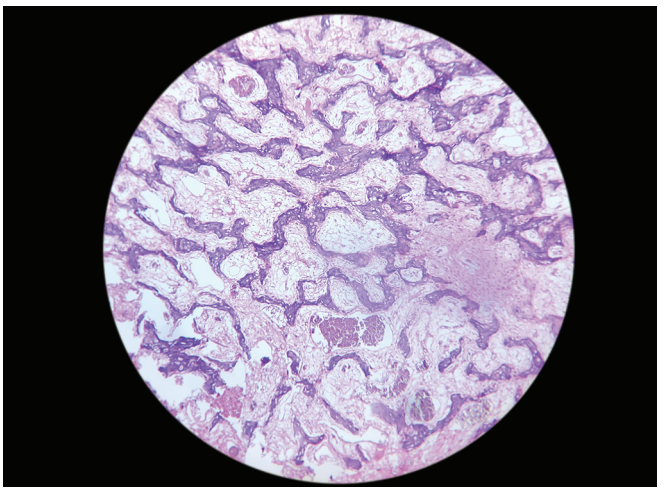

Fig. 7: Histopathological examination (HPE) showing randomly interconnecting trabeculae of woven bone rimmed by a single layer of osteoblasts

Microscopically, osteoblastomas are like osteoid osteoma and are characterized by.[8,9]

- Interconnecting trabeculae of woven bone rimmed by a single layer of osteoblasts.
- Trabeculae with different degrees of mineralization (from osteoid to pagetoid appearance) connecting to the bony edge in the periphery.
- Richly vascularized loose stroma.
- Possible central sclerotic nidus.
- Scattered osteoclastic giant cells.
- Well-defined borders without destructive bone permeation and no soft tissue extension.
- No atypical mitotic figures.
- Possibly aneurysmal bone cyst-like changes.

Treatment

Complete excision of the tumor is the surgical treatment goal for osteoblastoma. The surgery chosen depends on the location and aggressiveness of the tumor. En bloc surgical excision is associated with a lower recurrence rate.

Conclusion

Osteoblastoma of the hypopharynx has never been reported before in the literature, though there have been rare reports of osteoblastoma larynx. Though it was seen in the pyriform fossa, the possibility of it arising in the lateral aspect of the arytenoid cannot be ruled out. It is a diagnosis that can closely mimic osteoid osteoma and requires careful radiological and pathological correlation for a definite diagnosis. It can be treated completely by surgical excision. Open surgery is warranted since it is difficult to excise en bloc by endoscopic approach.

Clinical Significance

Rare presentations of common benign bone tumors like osteoblastoma can arise from extraosseous sites such as the larynx and hyoid, as described in various case reports. It is important to do a CECT neck when a rare diagnosis is suspected. Endoscopic excision can be difficult in such cases where osteoblastoma presents as a hypopharyngeal mass, and it is important to do open surgery in such cases.

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