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Maxillary Osteoblastoma in a Child Mimicking Intraosseous Hemangioma: A Case Report

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ABSTRACT

Maxillary osteoblastoma in children is exceptionally rare, accounting for fewer than 5% of all craniofacial cases. This benign bone tumor can closely mimic intraosseous hemangioma both clinically and radiographically due to its highly vascular stroma, leading to diagnostic pitfalls. We report a 5-year-old girl whose maxillary lesion exhibited soft, fluctuant swelling, reddish discoloration, bleeding on touch, and post-extraction hemorrhage—features strongly suggestive of hemangioma. A multidisciplinary approach involving maxillofacial surgery, radiology, and pathology was essential for definitive diagnosis. This case underscores that osteoblastoma must remain in the differential diagnosis of vascular-appearing pediatric maxillary masses, and multidisciplinary collaboration is critical to avoid misdiagnosis.

Keywords: Osteoblastoma; Maxilla; Hemangioma.

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Introduction

Osteoblastoma is a rare, benign osteogenic neoplasm that accounts for approximately 1% of primary bone tumors, predominantly affecting the spine and long bones in young males in their second to third decade of life [1]. Craniofacial involvement occurs in only 10–15% of cases, with maxillary localization being exceptionally uncommon, particularly in the pediatric population [2]. The diagnostic challenge intensifies when osteoblastoma mimics vascular lesions such as hemangioma, given their overlapping radiologic features including expansile radiolucency and prominent vascular channels [3]. We present a case of maxillary osteoblastoma in a child initially suspicious for hemangioma, highlighting diagnostic pitfalls and the importance of integrated clinical-radiological-pathological correlation in pediatric jaw tumors.

Case Presentation

A 5-year-old Afghan girl presented to a general dentist eight months before maxillofacial consultation with a chief complaint of mild right palatal swelling and pain. Clinical examination revealed a carious upper right deciduous first molar (tooth #E). The tooth was extracted under local anesthesia. Immediately following extraction, the socket exhibited notable and prolonged bleeding exceeding expected intraoperative oozing, requiring sustained manual pressure to achieve hemostasis. Although the socket healed uneventfully over subsequent weeks, the palatal swelling failed to resolve. Instead, it progressively enlarged over the following months, extending from the right palatal side to the right buccal vestibule, accompanied by ipsilateral facial swelling (Figure 1).

This clinical deterioration prompted referral to the maxillofacial surgery service at Al Zahra Hospital, Isfahan, in December 2025 for definitive evaluation. Intraoral examination revealed a soft, fluctuant swelling on the right hard palate measuring approximately 3 × 2.5 cm, extending from the palatal midline to the right alveolar ridge. The overlying mucosa demonstrated a reddish discoloration (Figure 1). Light probing induced spontaneous bleeding from the surface, which was controlled with gentle pressure. The extraction site of tooth #E was fully healed. No pulsation or bruit was detected on auscultation. General physical examination revealed syndactyly of the left foot, characterized by partial soft tissue fusion between the toes (Figure 1). The mother reported an identical finding on her own left foot. No other syndromic stigmata were identified. Panoramic radiography demonstrated a mixed-density expansile lesion in the right maxilla. The unerupted upper right first and second permanent premolars were displaced superiorly toward the maxillary sinus, with intact follicular spaces and no evidence of root resorption (Figure 2). Subsequent contrast-enhanced computed tomography (CT) revealed an expansile, blastic, sclerotic mass involving the right maxillary bone, measuring approximately 26 × 22 mm in greatest dimensions. The overlying cortex was expanded but intact, without cortical breach. Superiorly, the lesion extended toward the right maxillary sinus, which exhibited mild fluid opacification. No soft tissue component, periosteal reaction, bone destruction, or infiltration into adjacent structures was identified. Regional lymphadenopathy was absent (Figure 3). Fine needle aspiration (FNA) performed during incisional biopsy yielded frank blood, leading the surgeon to strongly suspect intraosseous hemangioma. Consequently, the patient was referred for cerebral angiography to further evaluate the vascular nature of the lesion. Angiography demonstrated evidence of tumoral blush supplied by the right facial artery.

Selective catheterization of the feeding vessel was performed with a microcatheter, followed by embolization with a mixture of glue (n-butyl cyanoacrylate) and lipiodol to reduce intraoperative bleeding risk. Following embolization, the patient underwent incisional biopsy. Histopathological examination of the biopsy specimen revealed irregular trabeculae of osteoid and woven bone lined by plump, uniform osteoblasts without significant atypia, embedded in a highly vascular fibrovascular stroma and giant cell osteoclasts. (Figure 4). These findings established the diagnosis of osteoblastoma and definitively excluded hemangioma. Following histopathological confirmation, the patient underwent complete surgical excision of the right maxillary mass under general anesthesia (Figure 5). Intraoperative findings confirmed an expansile, vascular, bony lesion consistent with osteoblastoma. Postoperative histopathology of the resected specimen reconfirmed the diagnosis with negative surgical margins. The patient remained asymptomatic at a 6-month follow-up.

Table 1. Differential diagnosis of osteoblastoma vs. key mimics in pediatric maxillofacial region.

Feature	Osteoblastoma	Monostotic Fibrous Dysplasia	Intraosseous Hemangioma	Juvenile Ossifying Fibroma
Peak age	10–30 years [9]	5–15 years [10]	20–40 years [11]	5–15 years [7]
Gender predilection	Male > Female (2:1) [9]	Equal [10]	Female > Male [11]	Equal [7]
Common maxillofacial site	Mandible > Maxilla [9]	Maxilla > Mandible [10]	Mandible > Maxilla [8]	Maxilla > Mandible [7]
Growth pattern	Expansile, slow [9]	Expansile, painless [10]	Slow, expansile [11]	Locally aggressive [7]
Bleeding tendency	Moderate (vascular stroma) [9]	None [10]	Marked [8]	Minimal [7]
Radiographic appearance	Mixed lucent/sclerotic with well-defined margins [9]	«Ground-glass» matrix, ill-defined blending borders [10]	«Honeycomb» or «spoke-wheel» trabeculation [8]	Mixed radiolucent-radiopaque, thin cortical bone [7]
Cortical integrity	Expanded, intact or thinned [9]	Thinned but intact [10]	Expanded, may be perforated [11]	Thinned, may be perforated [7]
Calcification pattern	Irregular osteoid trabeculae [9]	Immature woven bone without osteoblastic rimming [10]	Phleboliths (rare in jaws) [8]	Psammoma-like ossicles or trabeculae [7]
Microscopic hallmark	Plump osteoblasts lining osteoid trabeculae [9]	Irregular curvilinear bone trabeculae («Chinese characters») [10]	Endothelial-lined vascular channels with red blood cells [8]	Cellular stroma with spherical ossicles (psammomatoid) or linear trabeculae (trabecular) [7]
Stroma	Highly vascular fibrovascular [9]	Hypocellular, fibrous [10]	Vascular channels, minimal stroma [11]	Cellular, fibroblastic [7]
Osteoblastic rimming	Prominent [9]	Absent [10]	Absent [11]	Variable, often absent [7]
IHC markers	Osteocalcin (+), SATB2(+), CD31/34/ERG (-) [9]	CD31/34(-), SATB2 variable [10]	CD31(+), CD34(+), ERG (+), podoplanin(+) [8]	BCL2(+), RUNX2(+), CD31/34(-) [7]

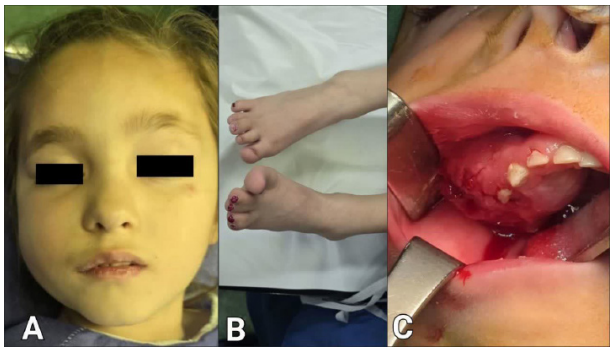

Figure 1. A) Extraoral photograph shows right facial swelling, B) Syndactyly of the left foot, C) Intraoral view reveals a soft, fluctuant, reddish palatal swelling.

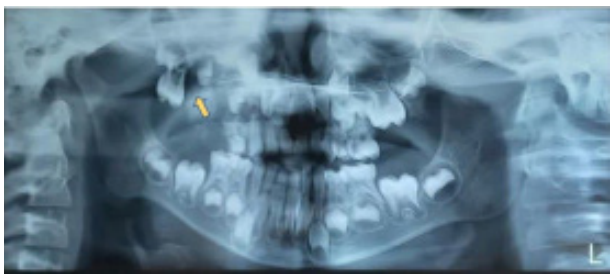

Figure 2. Radiographic Panoramic View: permanent premolars displacement toward maxillary sinus.

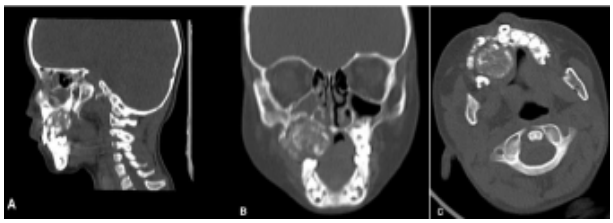

Figure 3. Computed Tomography: A) Sagittal View, B) Coronal View, C) Axial View: Expansile blastic sclerotic mass involving the right maxillary bone with intact expanded cortex and mild ipsilateral sinus opacification.

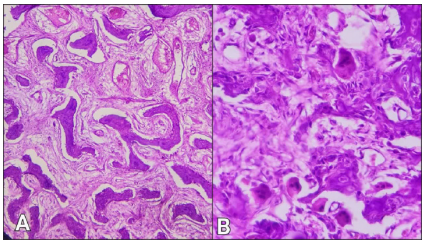

Figure 4. Microscopic View: A) (H&Ex4), B) (H&Ex100): Irregular osteoid trabeculae lined by osteoblasts with vascular stroma and giant cell osteoclasts.

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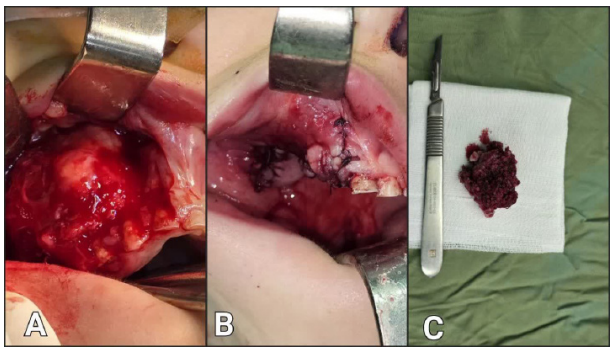

Figure 5. Surgical findings: A) Intraoperative view of the right maxillary vascular mass, B) Postoperative defect after complete excision, C) Excised gross specimen with mixed tan-white and hemorrhagic cut surface.

Discussion

Osteoblastoma of the craniofacial skeleton in children, particularly involving the maxilla and adjacent sinuses, remains an exceptionally rare entity with variable clinical presentations. Vella et al. reported a case of maxillary sinus osteoblastoma in a child who presented with progressive exophthalmos, highlighting that sinonasal osteoblastomas can mimic malignant tumors or mucoceles due to orbital involvement [4]. Unlike our patient, who exhibited soft palatal swelling with vascular features mimicking hemangioma, Vella's case lacked the striking bleeding tendency and post-extraction hemorrhage that characterized our diagnostic dilemma. Bacot et al. described an osteoblastoma of the ethmoid sinus presenting with nasal obstruction and chronic headache, emphasizing the diagnostic challenge posed by non-specific sinonasal symptoms [5]. Similarly, Young et al. reported a nasal septal osteoblastoma in a patient with recurrent epistaxis and nasal obstruction, which interestingly shared the vascular presentation with our case, as epistaxis suggested a hypervascular lesion [6].

However, their lesion originated from the nasal septum rather than the maxilla, and preoperative angiography was not employed. Salmen et al. documented an aggressive osteoblastoma of the maxilla with rapid growth and local destruction, requiring extensive surgical resection [7]. Unlike our case, which demonstrated intact cortical bone without aggressive features on CT, the aggressive variant exhibited cortical breach and infiltration, underscoring the importance of distinguishing conventional from aggressive osteoblastoma based on imaging and histology. Badran et al., in a systematic review of pediatric benign paranasal sinus osteogenic tumors, noted that osteoblastomas are frequently misdiagnosed preoperatively due to their rarity and overlapping features with fibrous dysplasia, ossifying fibroma, and hemangioma [8]. Their review supports our observation that vascular mimicry, while uncommon, can lead to diagnostic confusion and potentially unnecessary angiography if clinical suspicion is not carefully correlated with histopathology. Osteoblastoma, monostotic fibrous dysplasia, intraosseous hemangioma, and juvenile ossifying fibroma represent the principal differential diagnoses for expansile maxillary lesions in children, each with distinguishing clinical, radiographic, and histopathologic features (Table 1). Radiographically, osteoblastoma typically appears as a well-defined mixed lucent-sclerotic expansile lesion with intact or thinned cortex [9]. Monostotic fibrous dysplasia demonstrates a characteristic "ground-glass" matrix with ill-defined borders blending into surrounding bone [10].

Intraosseous hemangioma shows a lucent "honeycomb" or "spoke-wheel" trabecular pattern, sometimes with phleboliths [8]. Juvenile ossifying fibroma presents as a mixed radiolucent-radiopaque mass with cortical thinning and potential perforation [7]. Histopathologically, osteoblastoma is defined by irregular osteoid and woven bone trabeculae lined by plump, uniform osteoblasts within a highly vascular fibrovascular stroma. Immunohistochemistry shows positivity for osteocalcin and SATB2 with negativity for CD31, CD34, and ERG [9]. Fibrous dysplasia displays curvilinear "Chinese character" bone trabeculae embedded in hypocellular fibrous stroma without osteoblastic rimming [10]. Hemangioma reveals endothelial-lined vascular channels with intraluminal red blood cells, staining positive for CD31, CD34, and ERG [8]. Juvenile ossifying fibroma exhibits psammoma-like ossicles (psammomatoid variant) or linear trabeculae (trabecular variant) within a cellular fibroblastic stroma, expressing BCL2 and RUNX2 [7].

Clinical bleeding tendency is marked in hemangioma and moderate in osteoblastoma due to their vascular stroma, but absent in fibrous dysplasia. Crucially, none of the above cases reported preoperative angiography and embolization based on an initial misdiagnosis of hemangioma, making our case unique in demonstrating the diagnostic cascade from FNA (frank blood) → angiography (tumoral blush) → embolization → biopsy → histopathologic diagnosis. This approach, while reflective of safe surgical planning, highlights a key learning point: osteoblastoma should remain in the differential diagnosis of vascular pediatric maxillary masses, as its highly vascular stroma can convincingly mimic hemangioma both clinically and

radiographically [7,8]. The critical diagnostic distinction requires histopathologic examination, sometimes with confirmatory immunohistochemistry, as imaging alone cannot reliably differentiate these entities [8,9].

Conclusion

This case underscores that osteoblastoma should be considered when evaluating vascular-appearing maxillary masses in children, even when angiography points toward hemangioma. A stepwise diagnostic approach from clinical assessment to imaging and ultimately histopathology is critical to prevent diagnostic error.

Conflict of Interest

There is no conflict of interest to declare.

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