

A Diagnostic Challenge: A high-grade fibroblastic osteosarcoma of the mandible arising from a recurrent fibro-osseous lesion

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Background: Osteosarcoma (OS) of the jaws consists of three major histologic subtypes: chondroblastic OS, osteoblastic OS and fibroblastic OS (FOS). Ossifying fibroma of the jaws have three subtypes: conventional ossifying fibroma (COF), juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF).

Case Report: We present an interesting case of a mandibular tumor initially diagnosed as a COF with subsequent recurrences diagnosed as JTOF and FOS. A 62-year-old female presented in 2015 with an expansile radiolucent lesion of the right mandibular premolar region. Biopsy of this lesions was diagnosed as COF. Patient returned in 2016 complaining of a “bump” in her right mandibular buccal vestibule and recent onset of paresthesia of the right lip and chin. Excisional biopsy of the tumor rendered a diagnosis of JTOF. Patient was lost to follow up after 3 months, returning in 2019 with a rapidly growing tumor in her right mandible. Patient underwent right hemi-mandibulectomy with reconstruction, with the surgical specimen diagnosed as FOS. We reviewed the microscopic features of the primary tumor and subsequent recurrences, examining the expression of MDM2, CDK4 and p53 in these tumor samples. Surgical specimens from 2015 and 2016 revealed hypercellular proliferation of spindle and stellate cells with minimal collagen, with focal areas of osteoid and thin trabeculae of woven bone within the stroma. Tumor cells revealed no evidence of cytologic activity or increased mitotic activity. In contrast, the surgical specimen from 2019 revealed highly cellular plump spindle cells proliferation with cytologic atypia, increased mitotic activity, with focal and minimal osteoid. All three tumor samples revealed nuclear positivity for MDM2, CDK4 and p53.

Conclusion: This case represents a low-grade FOS undergoing de-differentiation to a high-grade FOS, illustrating the challenge and significance of distinguishing low-grade FOS from JTOF which share overlapping microscopic features.