

# Odontoameloblastoma

## Case Report and Review of Literature

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### ABSTRACT

**The odontoameloblastoma is an extremely rare odontogenic tumor about which there are fewer than 50 reported cases in the English literature.<sup>2,9</sup> It is a neoplasm that contains elements of both proliferating odontogenic epithelium and mesenchymal tissue.<sup>1</sup> The lesion primarily affects young adults in the third decade of life, with a median age of 20.2 years of age.<sup>2</sup> There is a slight male predilection; and the lesion is known to occur almost equally in the mandible and maxilla.<sup>1</sup> This paper aims to review the current literature pertaining to this pathologic entity and describe a case that was treated at our institution.**

The odontoameloblastoma is an extremely rare odontogenic tumor about which there are fewer than 50 reported cases in the English literature.<sup>2,9</sup> A review of the literature by Dive, et al., in 2011, confirmed only 22 cases of unequivocal odontoameloblastoma<sup>1</sup> based on histologic criteria.

The World Health Organization adopted the term in 1971 to describe a neoplasm that “includes odontogenic ectomesenchyme, in addition to odontogenic epithelium, that resembles an ameloblastoma in both structure and behavior.”<sup>1</sup> Histologically, the entity consists of a tumor with a background of odontogenic epithelial neoplastic cells (ameloblastoma), with elements of ectomesenchyme that lead to the formation of dental hard tissues (enamel and dentin).<sup>1</sup> It is hypothesized that epithelial proliferation leading to the formation of island or cords of cells producing the histologic appearance of ameloblastoma secondarily induces

adjacent mesenchymal cells to produce hamartomatous mass of mineralized dental tissues.<sup>2</sup>

The first documented human occurrence of an odontoameloblastoma was described in 1944 by Thoma, et al. Since then, sporadic reports have been generated across the globe.<sup>1</sup> Prior to 1971, the odontoameloblastoma, ameloblastic fibroodontoma and developing odontomas were reported as ameloblastic odontoma, adamant-odontoma, and soft and calcified odontomas. At this time, the term ameloblastic odontoma was abandoned and new terminology, including odontoameloblastoma, was chosen given the fact that the aggressive behavior of this tumor more closely resembles an ameloblastoma than that of an odontoma.<sup>1</sup>

Given the rarity of the odontoameloblastoma and confusing nomenclature in the past, the true incidence is difficult to determine. However, the epidemiology suggests it is an entity that predominantly affects young adults (mean age of 20.2 years of age), with a slight male predilection.<sup>1</sup> The lesion affects both jaws nearly equally, with a slightly increased incidence in the mandible.<sup>3</sup> And while they can occur in any part of the jaws, the majorities occur distal to the canine region.<sup>2</sup>

The lesion most commonly presents with the following symptoms: swelling, dull pain, malocclusion or altered eruption pattern of teeth. Occasionally, it is identified on routine radiographic examination.<sup>3</sup> Radiographically, it typically presents as a unilocular or multilocular radiolucency, with varying amounts of radiopaque components resembling mature dental tissues.<sup>3,5</sup> The radiopaque material may be in the form of small, diffuse particles, or a large central mass, which may cause a mass effect on adjacent structures.<sup>2,5</sup> The margin is usually well-defined, and displacement of surrounding anatomical structures, such as teeth, is more likely to occur, rather than resorbing them.<sup>3</sup>