

# Ameloblastoma arising in Calcifying Odontogenic Cyst: A Rare Histologic Variant

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## ABSTRACT

Calcifying odontogenic cyst (COC) has shown to be of extensive diversity in its clinical and histological features, as well as in its biologic behavior.<sup>1</sup> In this report, a rare case of ameloblastoma arising in COC is presented.

**Keywords:** Calcifying odontogenic cyst, Ameloblastoma, Rare histologic variant.

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## INTRODUCTION

The calcifying odontogenic cyst (COC) was first described as a distinct clinicopathologic entity by Gorlin et al in 1962.<sup>2</sup> It is a rare, well-circumscribed, solid or cystic lesions derived from odontogenic epithelium that resembles follicular ameloblastoma but contains ghost cells and spherical calcifications.<sup>3</sup> It is an uncommon lesion which was classified as a cyst<sup>4</sup> or as a neoplasm.<sup>5</sup>

## CASE REPORT

A 14-year-old male reported to the Department of Oral and Maxillofacial Surgery, GDC, Thiruvananthapuram, Kerala, India, with a swelling and occasional pain in posterior region of the right side of mandible. Panoramic radiograph showed a well-demarcated, multilocular radiolucent lesion extending from 33 to 46 regions (Fig. 1). The lesion was surgically enucleated and the specimen was sent for histopathological examination. Histopathology showed cystic epithelium overlying a connective tissue capsule. Epithelium was thick with a prominent layer of columnar basal cells with hyperchromatic nuclei, which were polarized away from the basement membrane. The superficial layers showed loosely arranged cells resembling stellate reticulum-like cells and ghost cell keratinization (Fig. 2). Based on these findings, the lesion was diagnosed as COC type 1A (simple unicystic type).

After surgical enucleation, the lesion recurred in the ensuing 2 years. Enucleation was done again and the specimen was sent for histopathological examination. Hematoxylin and eosin (H and E)-stained section showed



Fig. 1: Multilocular radiolucency extending from 33 to 46

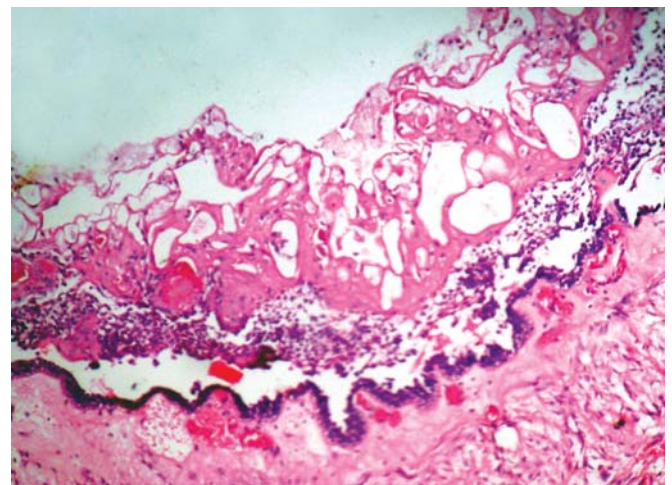


Fig. 2: H & E section shows cystic epithelium having ameloblastic cells and ghost cell keratinization

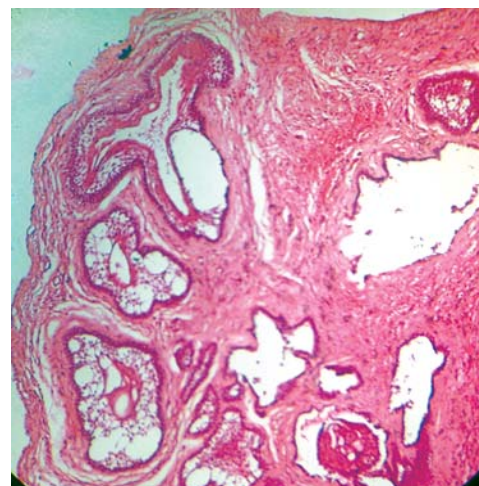


Fig. 3: H & E section shows ameloblastic tumor islands

thick fibrous wall with many small and large follicles of ameloblastoma. The follicular tumor islands consisted of peripheral palisading tall columnar hyperchromatic cells and stellate reticulum-like areas that fulfilled the ameloblastic histologic criteria (Fig. 3).

## DISCUSSION

Calcifying odontogenic cyst (COC) was described first by Gorlin, Pindborg, Practorius-clases and Vickers<sup>4,5</sup> and later by Gold.<sup>6</sup> Since then, several large series and isolated case reports have been published. It is an uncommon lesion which was classified as cyst<sup>7,8</sup> or as a neoplasm.<sup>8,9</sup> Since the first description by Gorlin et al,<sup>4,5</sup> it has become clear that two basic entities may be present: A cystic tumor and a solid neoplasm. Some authors considered that the original names, proposed previously, were inappropriate and suggested different names. Calcifying ghost cell odontogenic tumor,<sup>10</sup> cystic calcifying odontogenic tumor,<sup>11</sup> keratinizing ameloblastoma<sup>12</sup> and peripheral odontogenic tumor with ghost cell keratinization.<sup>13</sup>

In 1981, Practorius et al<sup>14</sup> proposed a widely used classification which tried to resolve the question on the cystic or neoplastic nature of the COC.

## Classification

*Type IA:* Simple unicystic type

*Type IB:* Odontome-producing type

*Type IC:* Ameloblastomatous proliferating type

*Type II:* Neoplastic type possesses some of the histological features of COC and has been called dentinogenic ghost cell tumor.

Several reports in the literature have documented the combined microscopic features of COC and ameloblastoma, merging from one to the other. The existence of an ameloblastoma, in association with COC, has been accepted since 1991, when Hong et al described two cases of plexiform ameloblastoma occurring in neoplastic variant of COC and strengthened a distinct difference between ameloblastoma ex COC and ameloblastomatous COC.

COC is most commonly found in second decade of life with almost equal sex distribution and similar incidence in maxilla and mandible. The most common site of occurrence is in the anterior part of jaws. They are usually unilocular but a few are multilocular as seen in the present lesion.

In the present lesion, the diagnosis of ameloblastoma is straightforward, being the first well-described case of COC.

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