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# Conservative Management of a Case of Plexiform Ameloblastoma

**Abstract:** Ameloblastomas are locally aggressive, benign odontogenic neoplasms having a wide variety of histologic patterns. It is essential to distinguish between the three clinical types of ameloblastomas – the intra-osseous solid lesion, the unicystic type and the extra-osseous lesion, as they differ in their biological behaviour and rate of recurrence and therefore require different forms of treatment. The case presented here is of a 9-year-old boy who reported to the department with pain and swelling along the right side of the mandible of 3 months' duration. Previous histopathological examination of the lesion, performed at a local hospital, produced a picture consistent with unicystic ameloblastoma. The lesion was managed by enucleation and the patient has been followed up for the past 5 years. Radiographic and clinical examinations reveal signs of healing without recurrence.

**Clinical Relevance:** Conservative management of unicystic ameloblastomas may be justified in children provided that the patient can be followed up at regular intervals.

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Ameloblastoma is a benign and locally aggressive neoplasm of odontogenic epithelium that has a wide spectrum of histologic patterns resembling early odontogenesis. It accounts for 1% of all tumours and cysts<sup>1</sup> of the jaws and is the most common of all odontogenic tumours.<sup>2</sup> It may arise from the enamel organ, epithelium of odontogenic cysts or from basal cells of the oral mucosa.<sup>3</sup> Most cases affect mandibular molar and ramus regions.<sup>3</sup> The tumour is usually asymptomatic and presents itself as a slowly enlarging facial swelling. It is a destructive tumour with a propensity for recurrence if not properly excised.<sup>4</sup>

## Case report

A 9-year-old boy was referred from a local hospital to the Department of Paedodontics, Government Dental College, Trivandrum with pain and swelling in relation

to the right side of the lower jaw. The patient had a history of surgery having been performed on the swelling at a local hospital. Patient records revealed that curettage had been performed on the lesion, following which a surgical drain and iodoform gauze pack were placed and changed at regular intervals. Histopathologic examination of the specimen reported an appearance consistent with a dentigerous cyst or unicystic ameloblastoma. An OPG (Figure 1) was taken which showed a large unilocular radiolucency with well-defined borders which extended from the distal aspect of the right deciduous second molar, involved the lower border of the mandible on the same side and the ramus up to the neck of the condyle. The crown of the second permanent molar appeared to be within the radiolucency, while the roots of the first permanent molar showed resorption. There was a radio-opaque mass in relation to the crown of the second permanent molar which seemed like a gauze pack. Clinically, slight expansion of the cortical bone was evident. Enucleation of the cystic mass (Figure 2) was performed, followed by a thorough curettage. The first and second permanent molars had to be extracted. Histopathologic examination of the specimen gave a diagnosis of plexiform type of ameloblastoma (Figure 3). An iodoform gauze pack was given which was changed every third day. The patient was

reviewed every 3 months in the first year, every 6 months in the second and third year and annually in the following 2 years. Both clinical and radiographic examination showed evidence of healing (Figure 4).

## Discussion

Ameloblastomas are most often found between the third and fifth decades, with an average age of 27 years. However, some investigators believe that, as the tumour grows slowly, it probably starts to develop between early childhood and young adulthood.<sup>2</sup> The incidence of ameloblastoma in children seems to be on the increase. Some reports have given the incidence as high as 22% in individuals younger than 16 years of age.<sup>5,6</sup> Some investigators have also concluded that the tumour in children might be a different type of lesion from that observed in adults.<sup>5</sup>

Many investigators believe in radical treatment of ameloblastomas, with resection of at least 1 cm of bone beyond the tumour margins. This is because tumour cells may often remain in cancellous bone after conservative treatment, leading to recurrence of the tumour,<sup>7,8</sup> which in turn will result in removal of more bone and teeth. However, there are many reports which have warned of the difficulties in controlling the tumour after

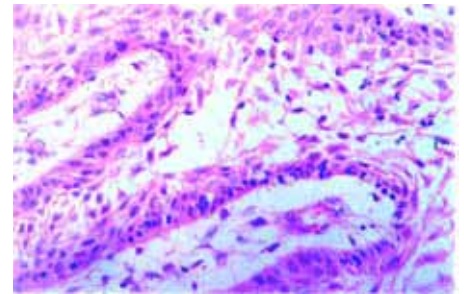
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**Figure 1.** Pre-operative radiograph – unilocular radiolucency (4.5 cm x 3 cm) extending from distal root of right mandibular deciduous second molar, extending close to lower border of mandible and up the ramus of mandible on same side. The radiolucency contains crown of the developing second molar and an ill-defined radio-opaque mass.



**Figure 2.** Excised mass.



**Figure 3.** Histopathology of excised mass (Haematoxylin and Eosin x 100).



**Figure 4.** Post-operative radiograph at 5 years showing uniform arrangement of trabeculae, normal radio-density and absence of recurrence.

such treatment.<sup>9</sup>

Even though there is general agreement that conservative methods of treatment may lead to a higher recurrence

rate, such radical procedures might not be acceptable in children as the growth of jaws is not yet completed. Radical procedures, such as segmental resection, will often lead

to deformity and dysfunction of the jaws in growing children, which in turn will hamper not only the physical growth of the child, but also his/her mental well-being. At the very least, conservative treatment will gain time until the growth of the jaws is complete.<sup>10</sup> In such cases, regular follow-up is a must. The presence of cancellous bone in children with an increased rate of bone turnover, a reactive periosteum and the presence of numerous unerupted teeth will further complicate treatment. The presence of cancellous bone might favour more rapid growth of the lesion, with extensive destruction, thus making surgery more demanding and difficult.<sup>9</sup>

The majority of ameloblastomas in children have been found to be unicystic. As only a small percentage of unicystic ameloblastomas extend outside the capsule, they have a very low rate of recurrence.<sup>11</sup> They can therefore be managed by more conservative forms of treatment, such as enucleation.<sup>12,13,14</sup> Another reason favouring a conservative treatment in children is the histopathologic feature of the ameloblastoma. The plexiform type is found more frequently in children,<sup>15</sup> which is found to behave less aggressively than the follicular type.<sup>16</sup> Also, plexiform ameloblastomas are found to remain in the primitive stage of tumour differentiation, whereas follicular and

acanthomatous types are thought to undergo squamous differentiation.<sup>5,17</sup>

## Conclusion

There is growing evidence that extensive resection of the mandible in ameloblastomas in children may not be required. If the pre-requisite of constant detailed follow-up can be satisfied, and the histopathologic examination of the lesion confirms it to be of the plexiform variety, conservative treatment should be the preferred treatment, as this type of ameloblastoma is shown to respond well to conservative modes of management. There is also the added advantage that the growth of the jaws is not affected, thus ensuring not only the physical well-being of the growing child but also his/her psychological well-being.

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