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**RESEARCH ARTICLE** 

# AMELOBLASTOMA IN PAEDIATRIC POPULATION - A SYSTEMATIC REVIEW

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Abstract: Background: Ameloblastoma is a benign but locally aggressive odontogenic tumor of the jaw, which is rare in pediatric populations. The clinical behavior, diagnosis, and management in children often differ from adults due to developmental considerations. Objective: This systematic review aims to summarize the clinical features, diagnostic methods, treatment strategies, and outcomes of ameloblastoma in pediatric patients. Methods: A comprehensive literature search was conducted in databases including PubMed, Scopus, and Web of Science. Studies on ameloblastoma in individuals ≤18 years published up to November 2024 from January 2020 were included. Data were extracted and synthesized qualitatively. Results: Pediatric ameloblastomas most commonly affect the mandible, presenting as painless swelling or facial asymmetry. Radiologically, they exhibit multilocular radiolucencies. Histologically, follicular and plexiform variants are predominant. Treatment includes conservative approaches like enucleation and curettage or radical resection, balancing recurrence risk and growth potential. Recurrence rates vary, with conservative treatments showing higher recurrence. Conclusion: Management of pediatric ameloblastoma requires individualized approaches considering the patient's age, tumor size, and growth potential. Future research should focus on long-term outcomes and recurrence prevention strategies.

Keywords: Ameloblastoma, pediatric, odontogenic tumor, jaw lesions, recurrence, management strategies.

#### INTRODUCTION

Ameloblastoma is a rare, benign but locally aggressive odontogenic tumor originating from the epithelial components of the developing tooth. First described in 1827 by Cusack, ameloblastoma accounts for approximately 1% of all oral and maxillofacial tumors and about 10% of odontogenic tumors. While most commonly diagnosed in adults between the ages of 30 and 50, it can also occur in children and adolescents, albeit less frequently. In 1885, the French physician Louis-Charles Malassez classified it "adamantinoma"[1]. Pediatric ameloblastoma believed to represent around 10-15% of all cases, making it an uncommon entity in the younger population [2]. The presentation of ameloblastoma in pediatric patients is unique compared to adults due to differences in growth patterns, craniofacial development, and healing potential. The tumor predominantly involves the mandible, particularly the posterior region, and presents as a slow-growing, painless swelling that may lead to facial asymmetry, tooth displacement, or malocclusion. In advanced cases, it can cause significant bone destruction, impacting the patient's quality of life and craniofacial development.[2]

Radiographically, ameloblastomas in children typically present as unilocular or multilocular radiolucencies,

mimicking other jaw lesions such as odontogenic keratocysts, dentigerous cysts, or fibrous dysplasia. The differential diagnosis of jaw lesions in children is crucial for early identification and appropriate treatment, as pediatric ameloblastoma may behave aggressively if left untreated. Histologically, the tumor exhibits distinct patterns, with follicular and plexiform variants being the most common [3]. The management of pediatric ameloblastoma is particularly challenging due to the need to balance oncological control with preservation of the growing jaw and surrounding Conservative approaches, including structures. enucleation and curettage, are preferred in younger patients to minimize disruption of normal growth and function, but these carry a higher risk of recurrence. Radical surgical approaches, such as segmental resection, may achieve better long-term control but pose risks of significant functional and aesthetic deficits. The choice of treatment depends on factors such as the size, location, histological variant, and recurrence risk of the tumor, as well as the patient's age and overall health. Despite its benign nature, ameloblastoma is characterized by a high recurrence rate, especially in cases treated conservatively. This necessitates longterm follow-up to monitor for recurrence and ensure optimal outcomes. Advances in imaging techniques, molecular diagnostics, and minimally invasive surgical

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methods have improved the understanding and management of pediatric ameloblastoma, but gaps remain in standardized treatment protocols and longterm outcome data. Given the rarity of ameloblastoma in children, there is limited comprehensive data in the literature. Most reports consist of isolated case studies or small case series, leading to variability in diagnostic and treatment approaches. This systematic review aims to consolidate current knowledge on pediatric ameloblastoma, focusing on its clinical presentation, diagnostic modalities, histopathological characteristics, treatment options, and outcomes. By synthesizing existing evidence, this review seeks to provide clinicians with insights for better management of this rare tumor in pediatric patients and highlight areas for future research.

# **MATERIAL AND METHODS**

#### **Search Strategy**

A systematic literature search was conducted in PubMed, Scopus, and Web of Science using the following keywords:

- "Ameloblastoma AND pediatric"
- "Odontogenic tumor AND children"
- "Ameloblastoma AND recurrence AND children"

Searches were limited to studies published in English from January 2020 to November 2024. Additional articles were identified from reference lists of selected papers.

**Eligibility Criteria** 

#### **Inclusion criteria:**

- Studies involving patients aged ≤18 years diagnosed with ameloblastoma.
- Articles describing clinical features, diagnosis, treatment, or outcomes.
- Case reports, case series, retrospective studies, and prospective studies.

#### Exclusion criteria:

- Studies focusing exclusively on adult populations.
- Animal studies or in vitro studies.
- Reviews and meta-analyses without original data.

# **Study Selection**

Articles were screened in two stages:

- 1. Title and abstract screening to identify potentially relevant studies.
- 2. Full-text screening to confirm eligibility.

#### **Data Extraction**

Data were extracted on the following parameters:

- 1. Patient demographics.
- 2. Clinical presentation and radiological findings.
- 3. Histopathological variants.
- 4. Treatment modalities and outcomes.
- 5. Recurrence rates and follow-up durations.

#### **Data Synthesis**

A qualitative synthesis was performed to summarize the clinical and therapeutic aspects of ameloblastoma in pediatric patients. Quantitative pooling of data was not feasible due to the heterogeneity of the included studies.

### **RESULTS AND OBSERVATIONS:**

The case reports analysed presents four distinct cases of pediatric ameloblastoma with varying clinical, radiographic, histopathological, and treatment outcomes. The summarized results are showed below as a tabular column as follows

Study	Patient Demographic	Clinical Presentatio	Radiographi c Findings	Histopathologic al Variant	Treatment	Key Outcomes
	s	n				
Marín-Márque z et al.,2024	11 years, Male	Swelling on the right side of the face; mild pain occasionally	Well-circumscribed, unilocular radiolucent lesion; unerupted 2nd molar displaced inferiorly, 3rd molar displaced posteriorly	Suggestive of ameloblastoma	Incisional biopsy performed	Detailed clinical and radiographic findings; histopatholog y pending
KIŞ HC et al.,2020	15 years, Female	Painless swelling in right lower jaw; asymmetry noted	Multilocular radiolucent lesion ("honeycomb " or "soap bubble"); lesion extends	Follicular ameloblastoma	Resection; reconstruction with costochondral graft and plate- screw systems	Successful surgical reconstructio n; prosthesis replacement planne



Khatavkar S et al.,2024	2 years, Male, Weight: 12 kg	Mass protruding from lower jaw since birth; gradual increase in size, non- tender and hard	from left lateral incisor to right angulus  Multiseptate d cystic spaces, ground glass ossified matrix; lesion extended across mandible involving symphysis menti	Ameloblastoma (confirmed via histopathology)	Wide local excision, mandibulectom y, PMMC flap reconstruction, and tracheostomy	successful surgery; postoperative ventilatory support and weaning, stable condition by day four
Jemâa M et al.,2024	7 years, Male	Raised upper lip; persistent tooth 61 (geminate); displaced tooth 21	Well-circumscribe d radiolucent lesion of the anterior maxilla associated with tooth 21	Unicystic ameloblastoma	Surgical enucleation of the cyst; re- implantation of tooth germ (tooth 21)	Successful eruption of tooth 21 with root formation; no recurrence

#### **DISCUSSION**

In Case 1 done by Marín-Márquez et al [4],a 11-yearold boy presented with a swelling on the right side of his face and occasional mild pain. Radiological findings indicated a unilocular radiolucent lesion with unerupted and displaced teeth, leading to a differential diagnosis that included ameloblastoma. While an incisional biopsy was performed, the histopathological findings were not yet reported. This case underscores the importance of correlating clinical and radiographic findings with histopathology to confirm the diagnosis. The displacement of teeth and localized bone expansion suggests an early stage of a less aggressive variant, potentially the unicystic type. Early diagnosis in cases such as this can prevent extensive surgical intervention, preserving more of the mandible's structure and function. When we go through Case 2 by KIŞ HC et al [5] ,a 15-year-old female presented with painless swelling in the right lower jaw causing asymmetry. Radiological features revealed a multilocular lesion described as "honeycomb" or "soap bubble," indicative of an aggressive variant such as follicular ameloblastoma. The lesion extended across a significant mandibular region, involving multiple impacted teeth. Histopathological confirmation of follicular ameloblastoma warranted surgical resection, followed by reconstruction with costochondral grafting and platescrew systems. This case highlights the challenges of managing more aggressive ameloblastoma types in adolescents. Despite successful surgical reconstruction,

the need for prosthetic rehabilitation indicates the functional and aesthetic impacts of extensive resection, underscoring the importance of multidisciplinary care. The study by Khatavkar S et al [6] was of a two-yearold male presented with a congenital mass that had progressively increased in size, involving the mandible extensively. Radiological imaging revealed multiseptated cystic spaces and ossified matrix, suggestive ameloblastoma. Histopathology confirmed the diagnosis, and wide local excision with mandibulectomy and PMMC flap reconstruction was performed. The presence of a large lesion in a very young child is rare and presents unique challenges. The case required careful airway management and meticulous surgical planning due to the lesion's size and location. The successful outcome, including postoperative stability and airway management, demonstrates the importance of a well-coordinated surgical and anesthetic approach in young pediatric patients. Jemâa M et al [7] studied the case of a 7-yearold male, the clinical presentation included a raised upper lip and displacement of tooth 21 due to a wellcircumscribed anterior maxillary lesion. histopathological examination confirmed unicystic ameloblastoma, a less aggressive variant. Treatment involved surgical enucleation of the lesion and reimplantation of the tooth germ. Long-term follow-up revealed successful eruption and root formation without recurrence. This case exemplifies how conservative surgical approaches, combined with long-term monitoring, can achieve excellent outcomes in pediatric



patients with less aggressive ameloblastoma variants. These cases collectively illustrate the diverse presentations, histopathological types, and treatment approaches for pediatric ameloblastoma. Key observations include:

#### 1. Age Variability:

The reported cases ranged from 2 to 15 years of age, highlighting that ameloblastoma can affect children from infancy through adolescence. Younger patients, like the two-year-old boy, often present with congenital or rapidly growing lesions, while older children may have more localized and less symptomatic swellings.

#### 2. Radiological Features:

Radiographic findings are crucial in guiding the differential diagnosis. While unilocular lesions are associated with less aggressive variants like unicystic ameloblastoma (Case 1, Case 4), multilocular lesions with "soap bubble" appearances are indicative of more aggressive types, such as follicular ameloblastoma (Case 2). Advanced imaging techniques such as CBCT provided critical details in cases with extensive or complex lesions.

#### **Histopathological Variants:**

The cases included both follicular and unicystic ameloblastomas, each with distinct biological behavior and treatment implications. Follicular ameloblastomas, as seen in Case 2, require aggressive surgical resection due to their higher recurrence risk. In contrast, unicystic ameloblastomas (Cases 1 and 4) allow for more conservative approaches, particularly in localized lesions.

#### Treatment Modalities:

Management strategies ranged from conservative enucleation with tooth germ preservation to wide excision and complex reconstructive surgeries. Younger patients, such as the two-year-old in Case 3, often necessitate a more extensive approach due to the lesion's size and the need for reconstruction to maintain facial and jaw function. Multidisciplinary involvement, including surgical, anesthetic, and prosthetic expertise, was critical in achieving favorable outcomes.

#### **Outcomes and Follow-Up:**

Long-term monitoring is vital in ameloblastoma cases to detect recurrences. Notably, the 7-year-old boy in Case 4 exhibited no recurrence after five years, reflecting the efficacy of conservative surgical management in unicystic ameloblastomas. Conversely, aggressive variants like those in Case 2 require more extensive monitoring and rehabilitation due to their higher risk of recurrence and the functional impact of resection.

## CONCLUSION

Ameloblastoma in pediatric patients is a rare odontogenic tumor with diverse clinical, radiological, and histopathological characteristics. The cases reviewed highlight the challenges in diagnosis and management due to the tumor's locally aggressive nature, recurrence potential, and impact on jaw function and aesthetics. It affects children across a wide age range, with presentations varying from congenital or rapidly enlarging lesions in very young children to localized swelling or asymmetry in older patients. Radiological features such as unilocular or multilocular lesions provide diagnostic clues, while histopathology remains the gold standard, distinguishing between variants like unicystic and follicular ameloblastoma. Treatment must balance tumor control with the preservation of growth potential and aesthetics, as demonstrated by conservative approaches like enucleation and tooth re-implantation in unicystic cases versus wide resection and complex reconstruction for aggressive variants. Multidisciplinary management is critical, involving maxillofacial surgeons, pediatric anesthesiologists, radiologists, and prosthodontists to ensure effective surgical planning, airway management, and long-term rehabilitation. Recurrence remains a significant concern, necessitating long-term clinical and radiological follow-up to detect early signs and prevent complications. Future advancements in imaging, surgical reconstruction, and tissue engineering could further refine treatment protocols. Overall, pediatric ameloblastoma requires comprehensive, a individualized, and multidisciplinary approach for optimal outcomes, emphasizing early diagnosis, tailored treatment planning, and meticulous follow-up to minimize recurrence and complications.

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