



PAPILLIFEROUS KERATOAMELOBLASTOMA – A SYSTEMATIC REVIEW

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ABSTRACT

Background: Papilliferous keratoameloblastoma is an extremely rare keratinizing papilliferous variant within the ameloblastoma spectrum, with limited data regarding its clinicopathological profile and biological behaviour.

Objective: This systematic review aimed to evaluate the demographic, clinical, radiographic, histopathological, treatment-related, and follow-up characteristics of reported cases of papilliferous keratoameloblastoma.

Methods: A systematic literature search was performed using PubMed/MEDLINE, Scopus, Web of Science, and Google Scholar from database inception to June 2026. Human case reports and case series with histopathologically confirmed papilliferous keratoameloblastoma or definite papilliferous keratinizing ameloblastomatous features were included. Data were synthesized narratively.

Results: Eight studies reporting eight individual cases were included. Patient age ranged from 18 to 65 years, with a mean age of 43.4 years, and male predominance was observed. All cases involved the right mandible, mainly the posterior mandibular region. Clinically, most cases presented as slowly enlarging mandibular swelling. Radiographic findings were non-specific and included unilocular or multilocular radiolucent/osteolytic lesions. Histopathologically, all cases showed ameloblastomatous epithelial differentiation with papilliferous or verrucopapillary projections and keratinization. Treatment varied from excision to mandibular resection. Recurrence was reported in one case.

Conclusion: Papilliferous keratoameloblastoma is a rare, diagnostically distinct, and potentially locally aggressive variant requiring histopathological confirmation, complete surgical removal, and long-term follow-up.

Keywords: Papilliferous keratoameloblastoma, Ameloblastoma, Keratoameloblastoma, Odontogenic tumour, Mandible, Histopathology.

INTRODUCTION

Odontogenic tumours constitute a diverse group of lesions arising from odontogenic epithelium, odontogenic ectomesenchyme or both.^[1] Among these, ameloblastoma is one of the most clinically significant epithelial odontogenic tumours because of its locally aggressive behaviour and high tendency for recurrence when inadequately treated. Although ameloblastoma is histologically benign, it may cause progressive jaw destruction through slow but infiltrative growth, cortical expansion,

tooth displacement, root resorption, facial deformity, cortical perforation, and occasionally soft tissue extension.^[2-4]

Epidemiologically, ameloblastoma is considered a rare jaw tumour, with a reported global incidence of approximately 0.92 cases per million person-years. It accounts for nearly 1% of all jaw tumours and is one of the most frequently encountered odontogenic tumours after odontoma.^[2,5] Ameloblastoma may occur over a wide age range, but it is most commonly seen in young to middle-aged adults, particularly between the second and fourth decades of life. A slight male predilection has been described in some epidemiological studies, although many reports show almost equal sex distribution. The mandible is affected far more commonly than the maxilla, with a strong predilection for the posterior mandibular body, angle, and ramus region. This anatomical preference is clinically important because mandibular lesions may remain unnoticed until they become large and destructive.^[6,7]

The clinicopathological behaviour of ameloblastoma is influenced by its growth pattern and histological subtype. Conventional ameloblastoma is generally more aggressive than unicystic and peripheral forms.^[8,9] Histologically, ameloblastoma exhibits several patterns, including follicular, plexiform, acanthomatous, granular cell, basal cell, desmoplastic and other less common variants. These patterns may occur individually or in combination within the same tumour. Among the uncommon patterns, keratinizing forms are diagnostically important because they may resemble other keratin-producing odontogenic cysts and tumours.^[10,11]

Keratin formation in ameloblastoma may be observed in acanthomatous ameloblastoma, keratoameloblastoma, and rare papilliferous keratinizing variants.^[12] Acanthomatous ameloblastoma shows squamous metaplasia within ameloblastomatous epithelial islands, sometimes with keratin pearl formation. Keratoameloblastoma is a rare variant characterized by extensive keratin production within odontogenic epithelial follicles, cystic spaces, or stromal areas.^[13,14] Papilliferous keratoameloblastoma represents an even rarer histopathological pattern in which ameloblastomatous epithelium shows prominent papilliferous or verrucopapillary epithelial projections associated with surface keratinization and keratin accumulation.^[15,16]

Papilliferous keratoameloblastoma is an exceptionally rare variant described within the spectrum of conventional ameloblastoma. Microscopically, it is characterized by cystic spaces or epithelial proliferations lined by odontogenic epithelium showing peripheral palisading, reverse nuclear polarity, stellate reticulum-like cells, squamous metaplasia, keratin pearl formation, desquamated keratin, necrotic debris, and papillary projections extending into cystic lumina. The coexistence of classic ameloblastomatous differentiation with papilliferous epithelial projections and keratinization forms the diagnostic basis of this lesion.^[17,18]

The exact epidemiological profile of papilliferous keratoameloblastoma remains uncertain because only a very small number of cases have been documented in the English-language literature. Available reviews suggest that it occurs mainly in adults and older individuals, with male predominance and a striking mandibular predilection, particularly for the right posterior mandible, mandibular body, angle, and ramus. Clinically, most patients present with a slowly enlarging mandibular swelling, which may be painless or associated with pain, tooth mobility, mucosal alteration, cortical expansion, or extraosseous growth. Radiographically, the lesion usually appears as a unilocular or multilocular radiolucency and may show buccolingual expansion, cortical thinning, cortical perforation, root resorption, or extension into the ramus.^[18,19]

However, the clinical and radiographic features of papilliferous keratoameloblastoma are not pathognomonic. It may closely mimic conventional ameloblastoma, odontogenic keratocyst, odontogenic myxoma, calcifying odontogenic cyst, central giant cell lesion, and other odontogenic tumours. Therefore, definitive diagnosis depends on careful histopathological examination. Recognition of the papilliferous keratinizing pattern is particularly important in incisional biopsies, where limited tissue sampling may lead to misdiagnosis as acanthomatous ameloblastoma, keratoameloblastoma, odontogenic keratocyst, or a nonspecific keratinizing odontogenic lesion.^[19,20]

The terminology and classification of papilliferous keratoameloblastoma remain controversial. Earlier reports regarded it as a rare keratinizing form of ameloblastoma, while one recurrent case with less

differentiated areas led to the suggestion of the term “papillary ameloblastic carcinoma.” However, subsequent cases have not consistently shown unequivocal cytological malignancy, abnormal mitoses, metastasis, or other features required for a malignant diagnosis. Hence, papilliferous keratoameloblastoma is currently best regarded as a rare histopathological subtype of conventional ameloblastoma unless definite malignant features are present.^[15,21]

At present, papilliferous keratoameloblastoma is not recognized as an independent odontogenic tumour entity in the World Health Organization classification. Nevertheless, its reproducible microscopic pattern has raised the possibility that it may deserve recognition as a distinct histopathological subtype.^[19] Because the literature is limited, scattered, and largely composed of isolated case reports, a systematic review is necessary to consolidate the available evidence. Therefore, the present review was undertaken to evaluate the demographic profile, site predilection, clinical and radiographic features, histopathological criteria, differential diagnosis, treatment modalities, recurrence potential, follow-up outcomes, and current classification status of papilliferous keratoameloblastoma.

MATERIALS AND METHODS

Review Design

This systematic review was conducted and reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines.^[22]

Review Question

What are the demographic, clinical, radiographic, histopathological, treatment-related and follow-up characteristics of papilliferous keratoameloblastoma reported in the literature?

Review Framework

A modified PICO framework^[23] was used to define the scope of the review.

Population: Patients diagnosed with papilliferous keratoameloblastoma of the jaws.

Intervention/Exposure: Histopathologically confirmed papilliferous keratinizing ameloblastomatous lesion.

Comparator: Not applicable, as the available evidence consisted mainly of case reports and case series.

Outcomes: Demographic profile, clinical presentation, radiographic findings, histopathological features, treatment modality, recurrence, and follow-up outcome.

Eligibility Criteria

Inclusion Criteria

Studies were included if they fulfilled the following criteria:

- Human case reports or case series.
- Lesions involving the jaws or maxillofacial skeleton.
- Histopathological diagnosis of papilliferous keratoameloblastoma or ameloblastoma showing definite papilliferous keratinizing features.
- Articles providing individual patient data.
- Articles published in the English language.
- Studies reporting at least one relevant clinical, radiographic, histopathological, treatment-related, or follow-up outcome.

Exclusion Criteria

Studies were excluded if they met any of the following criteria:

- Conventional ameloblastoma without papilliferous keratinizing features.
- Keratoameloblastoma without definite papilliferous epithelial projections.
- Odontogenic keratocyst or other odontogenic cysts without ameloblastomatous epithelial proliferation.
- Review articles without extractable individual case data.
- Duplicate publications.

- Animal studies, in vitro studies, conference abstracts, editorials, letters, and expert opinions.
- Non-English articles where translation was unavailable.

Search Strategy

A comprehensive electronic literature search was performed using PubMed/MEDLINE, Scopus, Web of Science, and Google Scholar. The search was conducted from database inception to June 2026. The following keywords and Boolean combinations were used:

“papilliferous keratoameloblastoma”
“papilliferous ameloblastoma”
“keratoameloblastoma”
“keratinizing ameloblastoma”
“keratinising ameloblastoma”
“ameloblastoma” AND “papilliferous”
“ameloblastoma” AND “keratinization”
“ameloblastoma” AND “keratinisation”
“odontogenic tumour” AND “papilliferous”
“odontogenic tumor” AND “keratoameloblastoma”.

In addition to the electronic search, the reference lists of all eligible articles and relevant review papers were manually screened to identify additional studies that may not have been retrieved through database searching.

Study Selection Process

All records identified through the search strategy were screened initially by title and abstract. Duplicate records were removed before full-text assessment. Articles that appeared to meet the eligibility criteria, or where eligibility was unclear from the title and abstract, were retrieved for full-text review. Full-text articles were then assessed in detail according to the predefined inclusion and exclusion criteria.

Studies were included only when the lesion demonstrated both ameloblastomatous differentiation and a definite papilliferous keratinizing component. Articles that reported keratinizing ameloblastoma without papillary epithelial projections were excluded unless the histopathological description supported the diagnosis of papilliferous keratoameloblastoma. Any uncertainty regarding eligibility was resolved by careful reassessment of the clinical and histopathological details.

Data Extraction

Data extraction was performed using a standardized data extraction form. The following variables were recorded from each eligible study:

- Author and year of publication.
- Country or geographic origin of the reported case.
- Patient age and sex.
- Site and side of the lesion.
- Duration of symptoms.
- Clinical presentation.
- Radiographic appearance.
- Histopathological findings.
- Immunohistochemical or molecular findings, if available.
- Treatment modality.
- Follow-up duration.
- Recurrence or complications.
- Authors' interpretation of the lesion's biological behaviour.

Outcome Measures

The primary outcome of the review was the histopathological diagnostic profile of papilliferous keratoameloblastoma.

Secondary outcomes included demographic distribution, site predilection, clinical presentation, radiographic appearance, treatment modality, recurrence, and follow-up status.

Quality Assessment

Since the included studies were expected to be case reports or small case series, methodological quality was assessed using domains adapted from the Joanna Briggs Institute critical appraisal checklist for case reports. Each study was evaluated for the following parameters:

- Clear description of patient demographic characteristics.
- Adequate clinical history.
- Clear clinical examination findings.
- Adequate radiographic description.
- Clear histopathological diagnostic criteria.
- Description of treatment modality.
- Adequate post-treatment follow-up.
- Reporting of recurrence or complications.
- Clarity of final diagnosis.
- Contribution to the clinicopathological understanding of papilliferous keratoameloblastoma.

Based on the completeness and clarity of reporting, each study was qualitatively categorized as having low, moderate or high risk of reporting bias.

Data Synthesis

A qualitative narrative synthesis was performed due to the rarity of the lesion, small number of reported cases, heterogeneity of reporting, and absence of comparative clinical studies. The extracted data were grouped and analyzed under the following headings: demographic profile, site distribution, clinical presentation, radiographic features, histopathological findings, differential diagnosis, treatment modalities, follow-up outcomes, recurrence and diagnostic significance.

RESULTS

Study Selection

The electronic search identified 21 records, and 3 additional records were obtained through manual reference screening, giving a total of 24 records. After removal of 6 duplicates, 18 records were screened by title and abstract. Six records were excluded as they were unrelated to the review question, lacked individual case data, or did not show definite papilliferous keratinizing ameloblastomatous features. Full texts of 12 articles were assessed, of which 4 were excluded for not fulfilling the eligibility criteria. Finally, 8 studies were included in the qualitative synthesis. (Figure 1)

General Characteristics and Clinical Presentation

The 8 included studies reported 8 individual cases of papilliferous keratoameloblastoma between 1994 and 2025. The patients' age ranged from 18 to 65 years, with a mean age of 43.4 years. Male predominance was observed, with 7 males and 1 female. All cases involved the mandible, and all lesions were located on the right side. The posterior mandible, including the premolar-molar region, mandibular body, angle, ramus, and condyle, was the most commonly affected region. (Table 1)

The most common presentation was a slowly enlarging mandibular swelling. Norval et al. (1994)^[24] reported pain and firm swelling in the right mandibular premolar-molar region, while Collini et al. (2002)^[25] described a right facial mass with difficulty in mandibular movement. Mohanty et al. (2013)^[26], Konda et al. (2016)^[27], Rathore et al. (2017)^[16], Kuberappa et al. (2020)^[15] and Jacob et al. (2023)^[28] reported mandibular swelling with variable features such as pain, tooth mobility, facial asymmetry, missing teeth and cortical expansion. Urs et al. (2025)^[29] described a recurrent exophytic, cauliflower-like growth in the right posterior mandible. Initial biopsy diagnosis differed from the final

diagnosis in the cases reported by Jacob et al. (2023)^[28] and Urs et al. (2025)^[29] highlighting the diagnostic difficulty of this lesion.

Radiographic Findings

Radiographically, the lesions were not pathognomonic and appeared as radiolucent or osteolytic mandibular lesions. Multilocular radiolucency was reported by Mohanty et al. (2013)^[26], Kuberappa et al. (2020)^[15], Jacob et al. (2023)^[28], and Urs et al. (2025)^[29]. Konda et al. (2016)^[27] and Rathore et al. (2017)^[16] described predominantly unilocular radiolucent lesions. Collini et al. (2002)^[25] reported an irregular osteolytic lesion involving the mandibular condyle and ramus with adjacent soft tissue infiltration. Common radiographic findings included cortical expansion, cortical thinning, cortical perforation, root resorption and extension into the ramus or soft tissues. (Table 2)

Histopathological Findings

Histopathological examination was the key diagnostic method in all cases. The common diagnostic pattern was ameloblastomatous odontogenic epithelium with papilliferous or verrucopapillary projections and keratinization. Norval et al. (1994)^[24] described cystic ameloblastic follicles with orthokeratin, parakeratin, desquamated squames, necrotic material, and papilliferous epithelial lining. Collini et al. (2002)^[25] reported papillary and micropapillary epithelial projections with necrotic debris and recurrent less differentiated areas. Mohanty et al. (2013)^[26], Konda et al. (2016)^[27], Rathore et al. (2017)^[16] and Kuberappa et al. (2020)^[15] described features such as peripheral palisading, reverse polarity, stellate reticulum-like cells, squamous metaplasia, keratin pearls, keratin flakes and papillary projections into cystic spaces. Jacob et al. (2023)^[28] emphasized macro-microscopic correlation with papillary folds, thick keratinized surfaces, and vascular connective tissue cores, while Urs et al. (2025)^[29] reported papilliferous odontogenic epithelial proliferation with surface keratinization. (Table 2)

Treatment, Follow-up and Risk of Bias

Treatment varied from excision to wide local excision and mandibular resection. Norval et al. (1994)^[24] performed segmental resection with iliac crest bone graft, while Collini et al. (2002)^[25] reported hemimandibulectomy with modified neck dissection. Konda et al. (2016)^[27] performed excision in toto with extraction of 44, and Rathore et al. (2017)^[16] treated the lesion by wide local excision. Urs et al. (2025)^[29] reported resection of the right posterior mandible with titanium plate stabilization. Follow-up was inconsistently reported. Collini et al. (2002)^[25] reported recurrence after 39 months and further relapse, whereas Rathore et al. (2017)^[16] reported no recurrence after 2 years. Konda et al. (2016)^[27] reported uneventful follow-up for 1 year before the patient was lost to follow-up. (Table 2)

The reporting quality of the included studies varied. Most studies provided adequate clinical, radiographic, and histopathological details, but follow-up was incomplete in several reports. Collini et al. (2002)^[25] and Rathore et al. (2017)^[16] provided stronger follow-up information, while Jacob et al. (2023)^[28] contributed detailed macro-microscopic correlation. Overall, the certainty of evidence was low because all included studies were case reports. (Table 3)

Summary of Findings

Overall, papilliferous keratoameloblastoma showed a strong predilection for the right posterior mandible and commonly presented as a slowly enlarging mandibular swelling. Radiographically, it appeared as a unilocular or multilocular radiolucent/osteolytic lesion with variable cortical expansion, perforation, root resorption, or soft tissue extension. Histopathologically, all cases demonstrated ameloblastomatous epithelial differentiation with papilliferous projections and keratinization. Recurrence was reported by Collini et al. (2002)^[25], while Rathore et al. (2017)^[16] reported no recurrence after 2 years. These findings suggest that papilliferous keratoameloblastoma is a rare, diagnostically distinct and potentially locally aggressive histopathological variant requiring adequate surgical management and long-term follow-up.

DISCUSSION

Papilliferous keratoameloblastoma is an extremely rare keratinizing variant within the ameloblastoma spectrum. The present systematic review included eight reported cases by Norval et al. (1994)^[24], Collini et al. (2002)^[25], Mohanty et al. (2013)^[26], Konda et al. (2016)^[27], Rathore et al. (2017)^[16], Kuberappa et al. (2020)^[15], Jacob et al. (2023)^[28], and Urs et al. (2025)^[29]. The limited number of cases confirms the rarity of this lesion and explains the uncertainty regarding its epidemiology, biological behaviour, recurrence risk, and classification status. This observation is consistent with Sachdev et al. (2023)^[17], who emphasized that the literature on papilliferous keratoameloblastoma is mainly limited to isolated case reports. Goh et al. (2026)^[19] also suggested that this lesion may represent a distinct histopathological subtype of conventional ameloblastoma rather than an independent clinicopathological entity.

In the present review, the age of patients ranged from 18 to 65 years, with a mean age of 43.4 years. The youngest case was reported by Rathore et al. (2017)^[16], while the oldest was reported by Kuberappa et al. (2020)^[15]. A marked male predominance was observed, with seven males and one female. However, because of the small number of cases, this finding should be interpreted cautiously. Conventional ameloblastoma has also been reported across a wide age range, commonly affecting young to middle-aged adults. Reichart et al. (1995)^[30], Masthan et al. (2015)^[31], Effiom et al. (2018)^[32], and Hendra et al. (2020)^[33] described ameloblastoma as a tumour with broad age distribution and variable sex predilection. Therefore, the demographic pattern of papilliferous keratoameloblastoma appears to overlap with conventional ameloblastoma, although available evidence is insufficient to establish a definitive epidemiological profile.

A striking finding in this review was the consistent mandibular involvement. All included cases occurred on the right side of the mandible, most commonly involving the posterior mandibular body, premolar-molar region, angle, ramus, or condylar region. This pattern correlates with conventional ameloblastoma, which shows a strong predilection for the posterior mandible, as reported by Reichart et al. (1995)^[30], Philipsen and Reichart (1998)^[34], Effiom et al. (2018)^[32], and Hendra et al. (2020)^[33]. However, the exclusive right-sided occurrence in the included cases may reflect the small number of reported cases rather than a true biological predilection.

Clinically, most patients presented with a slowly enlarging mandibular swelling. Norval et al. (1994)^[24] reported pain and firm swelling, while Collini et al. (2002)^[25] described a facial mass with difficulty in mandibular movement. Mohanty et al. (2013)^[26], Konda et al. (2016)^[27], Rathore et al. (2017)^[16], Kuberappa et al. (2020)^[15], and Jacob et al. (2023)^[28] reported mandibular swelling with variable features such as pain, tooth mobility, facial asymmetry, missing teeth and cortical expansion. Urs et al. (2025)^[29] described a recurrent exophytic, cauliflower-like growth. These findings resemble conventional ameloblastoma, which frequently presents as a slow-growing jaw swelling with variable pain, tooth displacement, root resorption, and facial deformity, as noted by Masthan et al. (2015)^[31] and Effiom et al. (2018)^[32].

Radiographically, papilliferous keratoameloblastoma did not show pathognomonic features. Mohanty et al. (2013)^[26], Kuberappa et al. (2020)^[15], Jacob et al. (2023)^[28], and Urs et al. (2025)^[29] reported multilocular radiolucencies, whereas Konda et al. (2016)^[27] and Rathore et al. (2017)^[16] described predominantly unilocular radiolucent lesions. Norval et al. (1994)^[24] observed a circumscribed lobulated radiolucency with buccal cortical plate penetration, while Collini et al. (2002)^[25] reported an irregular osteolytic lesion with soft tissue infiltration. These findings overlap with conventional ameloblastoma, which may show unilocular or multilocular radiolucency, cortical expansion, root resorption and perforation. Hence, radiographic evaluation alone is insufficient and histopathological confirmation is essential.

Histopathology remains the diagnostic cornerstone. Vickers and Gorlin (1970)^[35] described important histological criteria of ameloblastoma, including peripheral palisading, reverse nuclear polarity, subnuclear vacuolization and stellate reticulum-like cells. These features were observed in several included cases. Mohanty et al. (2013)^[26], Konda et al. (2016)^[27] and Kuberappa et al. (2020)^[15] reported basal palisading, reverse polarity, stellate reticulum-like cells, squamous metaplasia, and

keratin formation. The defining feature of papilliferous keratoameloblastoma, however, was the presence of papillary or verrucopapillary epithelial projections with keratinization. Norval et al. (1994)^[24], Collini et al. (2002)^[25], Mohanty et al. (2013)^[26], Konda et al. (2016)^[27], Rathore et al. (2017)^[16], Kuberappa et al. (2020)^[15], Jacob et al. (2023)^[28], and Urs et al. (2025)^[29] all demonstrated this essential papilliferous keratinizing pattern.

This histological pattern is important for distinguishing papilliferous keratoameloblastoma from acanthomatous ameloblastoma and keratoameloblastoma. Acanthomatous ameloblastoma may show squamous metaplasia and keratin pearls, but usually lacks prominent papillary epithelial projections into cystic spaces. Keratoameloblastoma shows extensive keratin production, but papilliferous architecture is not consistently present. Whitt et al. (2007)^[36], Bedi et al. (2015)^[20], and Palaskar et al. (2015)^[37] described keratoameloblastoma as a rare and heterogeneous keratinizing ameloblastomatous lesion. The present review supported the view that papilliferous keratoameloblastoma represents a more specific papilliferous keratinizing pattern within this spectrum.

The biological behaviour of papilliferous keratoameloblastoma remains uncertain. Collini et al. (2002)^[25] reported the most aggressive case, with recurrence after 39 months and further local relapse, leading the authors to suggest the term “papillary ameloblastic carcinoma.” However, other reports did not consistently demonstrate cytological malignancy, abnormal mitoses, metastasis, or unequivocal malignant behaviour. Rathore et al. (2017)^[16] reported no recurrence after two years, while Konda et al. (2016)^[27] reported uneventful one-year follow-up before the patient was lost to follow-up. Therefore, papilliferous keratoameloblastoma should not be uniformly classified as carcinoma unless definite malignant features are present.

The current WHO classification does not recognize papilliferous keratoameloblastoma as a separate odontogenic tumour entity. Vered and Wright (2022)^[38] and Soluk-Tekkesin and Wright (2022)^[39] summarized the fifth edition of the WHO classification, in which PKA is not listed independently. Nevertheless, its repeated microscopic pattern suggests that it may deserve recognition as a distinct histopathological subtype.

Treatment varied among the included cases, ranging from excision to segmental resection and hemimandibulectomy. Broader ameloblastoma literature supports adequate surgical removal, especially for solid, multicystic, extensive, or recurrent lesions. Carlson and Marx (2006)^[40], Lau and Samman (2006)^[41], Antonoglou and Sándor (2015)^[42], Hendra et al. (2019)^[43], Qiao et al. (2021)^[44], Hendra et al. (2023)^[45], and Gasparro et al. (2024)^[46] reported lower recurrence with more radical or aggressive approaches compared with conservative treatment. Therefore, papilliferous keratoameloblastoma should be managed with complete surgical removal and long-term follow-up, particularly when cortical perforation, soft tissue extension, or recurrence is present.

Molecular evidence specific to papilliferous keratoameloblastoma is lacking. However, Kurppa et al. (2014)^[47], Brown et al. (2014)^[48], Sweeney et al. (2014)^[49], and Mamat Yusof et al. (2022)^[50] demonstrated the importance of BRAF V600E and MAPK pathway alterations in conventional ameloblastoma. Future cases should include molecular profiling to determine whether papilliferous keratoameloblastoma shares the same molecular background as conventional ameloblastoma or represents a distinct subgroup.

Overall, papilliferous keratoameloblastoma appears to be a rare, diagnostically distinct, keratinizing papilliferous subtype of conventional ameloblastoma. Its clinical and radiographic features overlap with conventional ameloblastoma, but histopathological recognition of papilliferous epithelial projections with keratinization is essential. The recurrence reported by Collini et al. (2002)^[25] supported adequate surgical management and long-term surveillance. Further well-documented cases with complete clinical, radiographic, histopathological, molecular, treatment, and follow-up data are required to clarify its biological behaviour and classification status.

CONCLUSION

Papilliferous keratoameloblastoma is an extremely rare and diagnostically distinct keratinizing papilliferous variant within the ameloblastoma spectrum. The available literature is limited to a small

number of case reports, but the included studies consistently showed a predilection for the mandible, particularly the posterior mandibular region, and most cases presented clinically as a slowly enlarging mandibular swelling. Radiographically, the lesion showed non-specific unilocular or multilocular radiolucent/osteolytic features that overlapped with conventional ameloblastoma and other odontogenic lesions. Therefore, histopathological examination remains essential for diagnosis, with the characteristic features being ameloblastomatous epithelial differentiation, papilliferous or verrucopapillary projections, and keratinization. Although most reported cases lacked long-term follow-up, the recurrence reported in one case suggests that the lesion may show locally aggressive behaviour. Complete surgical removal with long-term clinical and radiographic surveillance is therefore recommended. Further well-documented cases with complete histopathological, immunohistochemical, molecular, treatment, and follow-up data are required to clarify its biological behaviour, recurrence potential, and exact classification status.

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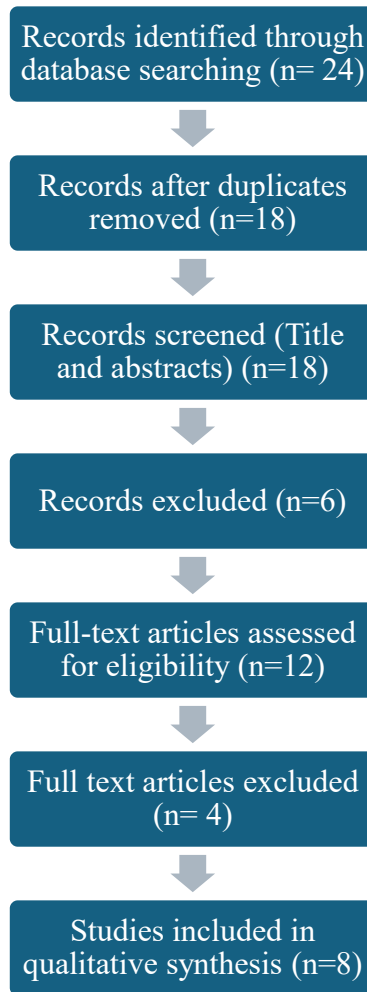
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FIGURES

**Figure 1: PRISMA Flowchart****TABLES****Table 1: Study Characteristics, Demographic Profile and Clinical Presentation of Included Cases**

Author and Year	Study Type	Country / Region	No. of Cases	Age / Sex	Site	Clinical Presentation	Duration / History
Norval et al. (1994) ^[24]	Case report	South Africa	1	26 / Female	Right mandibular premolar-molar region	Pain and firm swelling in the right mandibular premolar region	Not clearly reported
Collini et al. (2002) ^[25]	Case report with 6-year follow-up and review	Italy	1	62 / Male	Right mandible involving condyle, ramus and adjacent soft tissues	Right facial mass with difficulty in mandibular movement	3 months
Mohanty et al. (2013) ^[26]	Case report	India	1	46 / Male	Right mandible, 44 to angle region	Localized right lower jaw swelling with intraoral swelling and bicortical expansion	1 year
Konda et al. (2016) ^[27]	Case report	India	1	44 / Male	Right posterior mandible	Swelling in lower right posterior region with intermittent pain and tooth mobility	6 months

Rathore et al. (2017) ^[16]	Case report	India	1	18 / Male	Right mandibular premolar-molar / angle region	Diffuse swelling on right side of mandible without pain or paresthesia	3 months
Kuberappa et al. (2020) ^[15]	Case report with review	India	1	65 / Male	Right mandibular body region	Swelling and pain in right lower jaw with facial asymmetry	4 months
Jacob et al. (2023) ^[28]	Case report	India	1	42 / Male	Right mandible, 35 to 46 region	Expansile swelling of right mandible with multiple missing teeth	History of trauma 3 months earlier
Urs et al. (2025) ^[29]	Case report with review	India	1	44 / Male	Right posterior mandible extending to ramus	Recurrent exophytic / cauliflower-like growth	Enlarged again over 3 months

Table 2: Radiographic, Histopathological, Treatment and Follow-up Characteristics of Included Cases

Author and Year	Radiographic Findings	Key Histopathological Findings	Treatment Performed	Follow-up / Outcome
Norval et al. (1994) ^[24]	Circumscribed lobulated radiolucent lesion; CT showed penetration of buccal cortical plate	Cystic ameloblastic follicles containing orthokeratin, parakeratin, desquamated squames, necrotic material and dystrophic calcification; some areas showed papilliferous epithelial lining	Segmental resection with vascularized iliac crest bone graft	Follow-up not clearly reported; recurrence not reported
Collini et al. (2002) ^[25]	Large irregular osteolytic lesion involving the right mandibular condyle and ramus with adjacent soft tissue infiltration	Cysts lined by papillary and micropapillary projections with necrotic debris; cribriform, solid and tubular epithelial patterns; recurrent lesion showed granular cell and less differentiated areas	Right hemimandibulectomy with modified neck dissection; later treatment for recurrent lesion	Six-year follow-up; local recurrence after 39 months and further local relapse reported
Mohanty et al. (2013) ^[26]	Multilocular radiolucency with bicortical expansion, cortical perforation and root resorption	Cystic spaces filled with necrotic debris and lined by papillary keratin-lined infoldings; stellate reticulum-like cells; basal tall columnar ameloblast-like cells	Referred for surgical management	Follow-up and final outcome not reported

		with palisading and reverse polarity		
Konda et al. (2016) ^[27]	Predominantly well-defined unilocular radiolucency with buccal cortical expansion and thinning of lingual cortex	Cystic lining with squamous metaplasia; plexiform odontogenic epithelium; peripheral palisading and reverse polarity; keratin pearl formation; numerous papillary extrusions	Excision in toto with extraction of 44	Uneventful follow-up for 1 year; patient later lost to follow-up
Rathore et al. (2017) ^[16]	Well-defined radiolucent unilocular osteolytic lesion; CBCT showed expansion, cortical perforation, bony septae and thinning of inferior mandibular border	Tall columnar lining cells with stellate reticulum-like cells; verrucopapillary projections into cystic lumen; keratin plugging, parallel lamellae and keratin flakes; acantholytic cells and focal necrosis	Wide local excision	No recurrence reported after 2 years
Kuberappa et al. (2020) ^[15]	Multilocular radiolucency extending from 33 to 46 region with root resorption and buccal cortical expansion	Multiple cystic spaces lined by keratinized stratified squamous epithelium; papillary projections into cystic lumen; odontogenic islands with reverse polarity and subnuclear vacuolization; squamous metaplasia and keratin pearl formation	Complete excision / excision biopsy with final histopathological diagnosis	Follow-up not clearly reported; recurrence not clearly reported
Jacob et al. (2023) ^[28]	Well-defined multilocular radiolucency extending from 35 to 46 with root resorption involving 34, 35, 47 and 48	Large cystic cavity with necrotic debris; papillary folds with thick white keratinized covering; basal ameloblastomatous layer, stellate reticulum-like cells and vascular connective tissue core within papillae	Excisional biopsy after initial incisional biopsy	Follow-up and recurrence not reported
Urs et al. (2025) ^[29]	Well-defined multilocular radiolucency extending from right mandibular body to	Papilliferous proliferation of odontogenic epithelium with surface keratinization	Resection of right posterior mandible involving second molar and ramus up to coronoid, followed	Presented as recurrent exophytic growth

	anterior two-thirds of ramus	and keratin accumulation; final diagnosis made on unique histopathological features	by titanium plate stabilization	before definitive surgery; post-treatment recurrence not reported
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Table 3. Risk of Bias / Quality Assessment of Included Case Reports

Author and Year	Patient Details	Clinical Details	Radiographic Details	Histopathological Details	Treatment Details	Follow-up / Recurrence Reporting	Overall Risk of Bias
Norval et al. (1994) ^[24]	Adequate	Adequate	Adequate	Adequate	Adequate	Limited follow-up; recurrence not clearly reported	Moderate
Collini et al. (2002) ^[25]	Adequate	Adequate	Adequate	Adequate	Adequate	Adequate; recurrence and relapse reported	Low to Moderate
Mohanty et al. (2013) ^[26]	Adequate	Adequate	Adequate	Adequate	Limited	Follow-up and final outcome not reported	Moderate to High
Konda et al. (2016) ^[27]	Adequate	Adequate	Adequate	Adequate	Adequate	One-year follow-up reported; later lost to follow-up	Moderate
Rathore et al. (2017) ^[16]	Adequate	Adequate	Adequate	Adequate	Adequate	Adequate; no recurrence reported after 2 years	Low to Moderate
Kuberappa et al. (2020) ^[15]	Adequate	Adequate	Adequate	Adequate	Adequate	Follow-up and recurrence not clearly reported	Moderate
Jacob et al. (2023) ^[28]	Adequate	Adequate	Adequate	Detailed macro-microscopic correlation	Adequate	Follow-up and recurrence not reported	Moderate
Urs et al. (2025) ^[29]	Adequate	Adequate	Adequate	Adequate	Adequate	Post-treatment follow-up not reported	Moderate