

CASE REPORT

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Lateral periodontal cyst of the anterior maxilla: a rare case report

Manoj Adhikari^{1*} , Kanistika Jha² , Aashish Shah¹, Shova Kunwar¹, Bishwo Ram Amatya¹ and Junu Bhattarai¹

Abstract

Background The lateral periodontal cyst (LPC) is a rare developmental odontogenic cyst, constituting approximately 0.4% of all odontogenic cysts and 0.7% of all jawbone cysts. It is an intraosseous, non-keratinized, non-inflammatory, developmental odontogenic cyst of epithelial origin, typically occurring adjacent to the root of a vital tooth. The lesion usually measures less than one centimeter in diameter. However, in the present case, the lesion was larger, measuring 3.5 × 2.4 × 2.4 cm. Radiographically, LPCs present as unilocular, round, oval or teardrop-shaped radiolucent lesions with a sclerotic border, located laterally to the tooth roots.

Case presentation A 42-year-old female presented to the Oral and Maxillofacial Surgery outpatient department with a chief complaint of swelling on the anterior maxilla for one year. The swelling was asymptomatic, with no associated pain or discomfort. Clinical examination revealed a non-tender, fluctuant swelling on the left side of the anterior maxilla, located between the lateral incisor and canine teeth. A computed tomographic scan confirmed the presence of a lytic lesion on the left side of the anterior maxilla. Surgical enucleation of the cyst was performed through an intraoral crevicular approach under general anesthesia. The excised specimen was sent for histopathological examination. Histopathological examination of the cyst wall revealed a non-keratinized, non-ciliated stratified squamous epithelial lining, ranging from 1 to 5 cell layers in thickness. Interspersed among the epithelial cells were foci of Periodic Acid-Schiff (PAS) positive, glycogen-rich clear cells. Additionally, focal nodular areas of epithelial thickening with a whorled, swirling architecture were observed in continuity with the epithelial lining. The underlying subepithelial connective tissue was fibrous and exhibited a zone of hyalinization. These features were consistent with a diagnosis of lateral periodontal cyst. The healing process was uneventful, and no complications were observed during the one-year follow-up.

Conclusion Although the lateral periodontal cyst is a rare developmental odontogenic cyst, it should be considered in the differential diagnosis of cystic lesions adjacent to the root of a vital tooth. Surgical enucleation remains the treatment of choice, with histopathological examination confirming the diagnosis. Recurrence of lateral periodontal cysts is infrequent.

Keywords Periodontal cyst, Odontogenic cyst, Maxilla, Alveolar process, Tooth root, Case report

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Background

Cysts in the oral and maxillofacial regions are commonly encountered in clinical practice [1, 2]. They are categorized into odontogenic and non-odontogenic cysts based on their tissue of origin [1, 2]. The lateral periodontal cyst (LPC) is a rare developmental odontogenic cyst, accounting for approximately 0.4% of all odontogenic cysts and 0.7% of all jawbone cysts [3–9]. According to the 2022 WHO classification, it is classified as a cyst of the Jaw [10].

The LPC is an intraosseous, non-keratinized, non-inflammatory, developmental odontogenic cyst of epithelial origin occurring adjacent to the root of a vital tooth [4, 7, 11]. It is usually asymptomatic unless secondarily infected and is often detected incidentally during routine radiographic examinations [6, 12]. The cyst most commonly presents in the alveolar process of the mandibular canine and premolar region, followed by the anterior maxilla [9, 12, 13]. Cortical expansion may be observed, and resorption of the cortical bone has been reported [6, 13–15]. While the cyst is predominantly diagnosed in individuals between the fifth and seventh decades of life, cases in younger patients have also been documented [3, 6, 16, 17]. The lesion typically measures less than one centimeter in diameter; however, larger lesions exceeding one centimeter have been reported [9, 14, 18]. The lesion measured $3.5 \times 2.4 \times 2.4$ cm in the present case. The cyst originates from remnants of odontogenic epithelium, with its precise tissue of origin remaining controversial [3, 4, 16, 18]. It is hypothesized to arise from the rests of the dental lamina, the rests of Malassez or reduced enamel epithelium [4, 18].

Radiographically, the LPC appears as an unilocular, round, oval or teardrop-shaped radiolucency with a sclerotic border, positioned lateral to the tooth roots [11, 14]. It may be associated with the loss of periodontal ligament space and lamina dura of adjacent teeth [15]. Root divergence may be present; however, root resorption never occurs [8, 10, 15]. Histopathological examination remains the gold standard for confirming the diagnosis [11, 16]. The treatment of choice is surgical enucleation, with a low risk of recurrence [4, 17].

This report presents a rare case of a large LPC in the anterior maxilla, positioned between the roots of the left lateral incisor and canine teeth. The cyst was successfully managed through surgical enucleation, and no complications were observed during a one-year follow-up period.

Case presentation

A 42-year-old female presented to the Oral and Maxillofacial Surgery outpatient department (OPD) with a chief complaint of a painless swelling on the left anterior maxilla, persisting for one year. The patient reported no significant medical or family history and no genetic



Fig. 1 Preoperative clinical image demonstrating a cystic lesion on the left anterior maxilla, present between the left lateral incisor and canine teeth



Fig. 2 Clinical image depicting the aspiration of clear fluid from the lesion

abnormalities were noted. Clinical examination revealed a well-circumscribed, fluctuant, non-tender swelling between the left lateral incisor and canine teeth, with healthy overlying mucosa. The adjacent teeth were vital and no signs of infection or lymphadenopathy were observed (Fig. 1). Aspiration of the lesion yielded clear fluid, indicative of a cystic nature and effectively excluding abscess or vascular anomalies (Fig. 2).

Panoramic radiography revealed a radiolucent lesion with a sclerotic border between the left lateral incisor and canine teeth, accompanied by divergence of the adjacent tooth roots (Fig. 3). A computed tomographic scan demonstrated a lytic, oblong, expansile lesion measuring

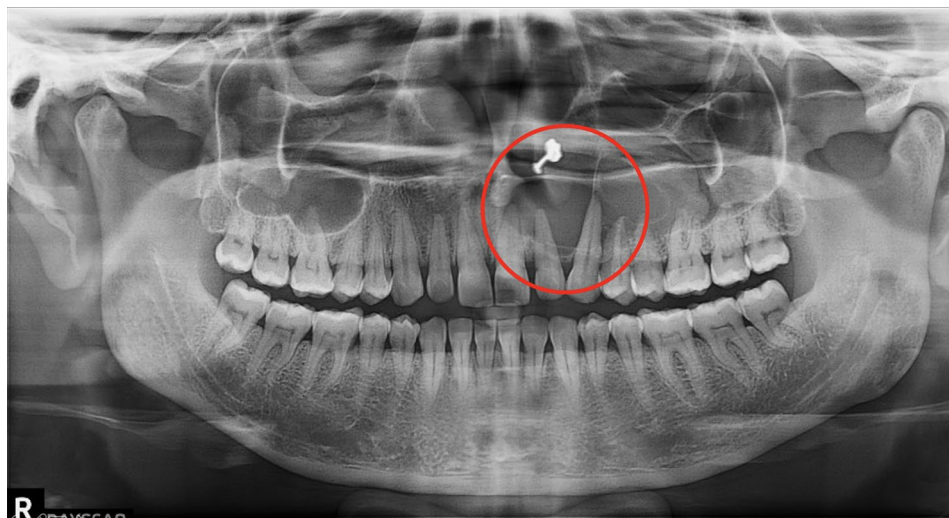


Fig. 3 Preoperative orthopantomogram revealing a radiolucent lesion in the left anterior maxilla, positioned between the left lateral incisor and canine teeth

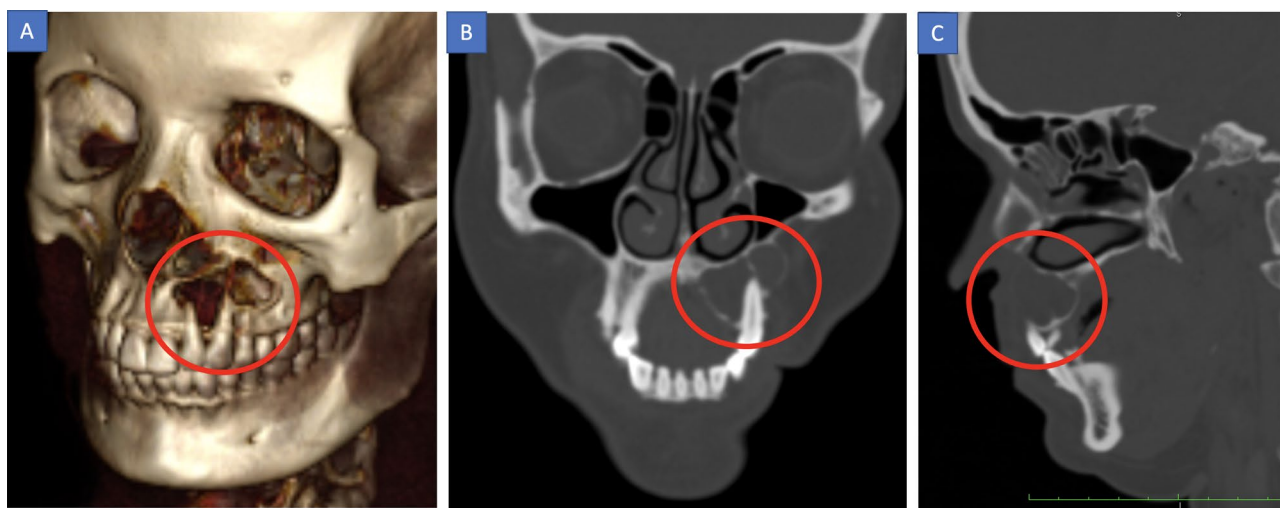


Fig. 4 Preoperative CT images of the face: (A) Volume Rendering Technique (VRT) images; (B) Coronal section; (C) Sagittal section. These images illustrate a lytic, oblong, expansile lesion within the left maxilla, extending from the left central incisor to the first molar. The lesion protrudes superiorly into the left maxillary sinus and causes obliteration of the alveolar bone associated with the lateral incisor, canine, and first premolar teeth. Additionally, divergence of the roots of the lateral incisor and canine teeth is evident

3.5 × 2.4 × 2.4 cm, with fluid density of average attenuation 14 HU, in the left maxilla. The lesion extended from the left central incisor to the first molar, protruded 12 mm into the left maxillary sinus superiorly and caused obliteration of the alveolus associated with the lateral incisor, canine, and first premolar teeth inferiorly. Divergence of the roots of the lateral incisor and canine teeth was also noted (Fig. 4).

The differential diagnosis for the observed cystic lesion included gingival cyst of the adult, odontogenic keratocyst, lateral radicular cyst, residual cyst, glandular odontogenic cyst, and radiolucent odontogenic tumors. A gingival cyst of the adult is typically located within the gingival tissue without involvement of the underlying

bone, a feature that was not observed in this case. Odontogenic keratocysts are characterized by the aspiration of thick, viscous, whitish keratinous material; however, aspiration in the present case yielded clear fluid. Lateral radicular cysts are associated with non-vital adjacent teeth, whereas the teeth adjacent to the lesion, in this case, were vital. Residual cysts are present in areas where a tooth is missing, yet no missing teeth were noted in this case. Based on these clinical and radiographic findings, the aforementioned lesions were excluded. The differential diagnosis was, therefore, narrowed to a lateral periodontal cyst, glandular odontogenic cyst or a radiolucent odontogenic tumor. Definitive diagnosis requires histopathological examination.

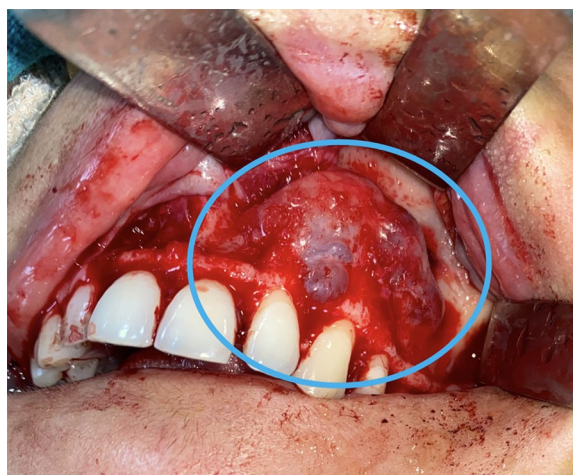


Fig. 5 Intraoperative photograph showing the cystic lesion located in the left anterior maxilla



Fig. 6 Intraoperative image presenting the surgical site and the bone following enucleation of the cystic lesion

Following a comprehensive pre-anesthetic evaluation, the patient was deemed fit for surgery under general anesthesia. Potential surgical risks, including bleeding, oroantral communication, infection and wound dehiscence, were thoroughly discussed with the patient and informed consent was obtained.

The case was operated under general anesthesia. The patient was positioned supine. Nasotracheal intubation was done. Parts preparation was done and a throat pack was placed. A crevicular incision was made extending from the right central incisor to the left first molar, with releasing incisions placed at both ends. A full-thickness mucoperiosteal flap was elevated, providing access to the underlying cystic lesion (Fig. 5). The overlying thin cortical bone was carefully removed, and the cyst was enucleated in its entirety (Fig. 6). The residual bony cavity was initially separated from the maxillary antrum by a thin bony partition, which was inadvertently perforated during the procedure. This resulted in communication

between the surgical site and the antral cavity. The excised specimen was fixed in 10% buffered formalin and submitted for histopathological examination. Hemostasis was achieved, and the surgical field was irrigated thoroughly with sterile normal saline. The flap was repositioned and primarily closed using 3–0 90/10 polyglycolide-co-L-lactide, braided, coated, absorbable sutures (Novosyn B. Braun Surgical, S.A. Rubi, Spain). The throat pack was subsequently removed and the patient was extubated without complications.

Postoperative management included a five-day course of Amoxicillin and potassium clavulanate 625 mg every 8 h, Ornidazole 500 mg every 12 h and Diclofenac 75 mg every 8 h. Additionally, intraoral irrigation with 0.12% chlorhexidine was advised twice daily for one week.

Histopathological examination of the cyst wall revealed a non-keratinized, non-ciliated stratified squamous epithelial lining, ranging from 1 to 5 cell layers in thickness. Interspersed among the epithelial cells were foci of Periodic Acid-Schiff (PAS) positive, glycogen-rich clear cells. Additionally, focal nodular areas of epithelial thickening with a whorled, swirling architecture were observed in continuity with the epithelial lining. The underlying subepithelial connective tissue was fibrous and exhibited a zone of hyalinization. Notably, there was an absence of apical snouting, hobnail cells, inflammatory cells, mucinous goblet cells, dysplasia or evidence of malignancy (Figs. 7 and 8).

Based on the above clinical, radiological and histopathological findings, the diagnosis of the Lateral periodontal cyst was confirmed.

The postoperative course was uneventful, with no complications observed during the one-year follow-up period. (Figures 9 and 10)

Discussion and conclusions

LPC is considered a rare entity in the realm of odontogenic cysts [3, 4]. The first report of LPC was by Standish and Shafer in 1958, who documented five cases located in the mandibular canine-premolar region [19].

The LPC is considered the intraosseous counterpart to the extraosseous gingival cyst of adults [4, 14]. Some studies suggest no gender predilection [4], while others argue a higher prevalence in males [18]. LPCs are most commonly identified in individuals between the fifth and seventh decades of life; however, cases in younger individuals have also been documented [3, 6, 14–16]. In the present case, the patient was a 42-year-old female, which is consistent with the literature.

Typically, LPCs occur in the alveolar process of the mandibular canine-premolar region, followed by the maxillary anterior region [5, 9, 12]. Approximately 70% of these cysts are found in the mandible, while 30% are in the maxilla [8, 9, 16]. In this case, the cyst was found

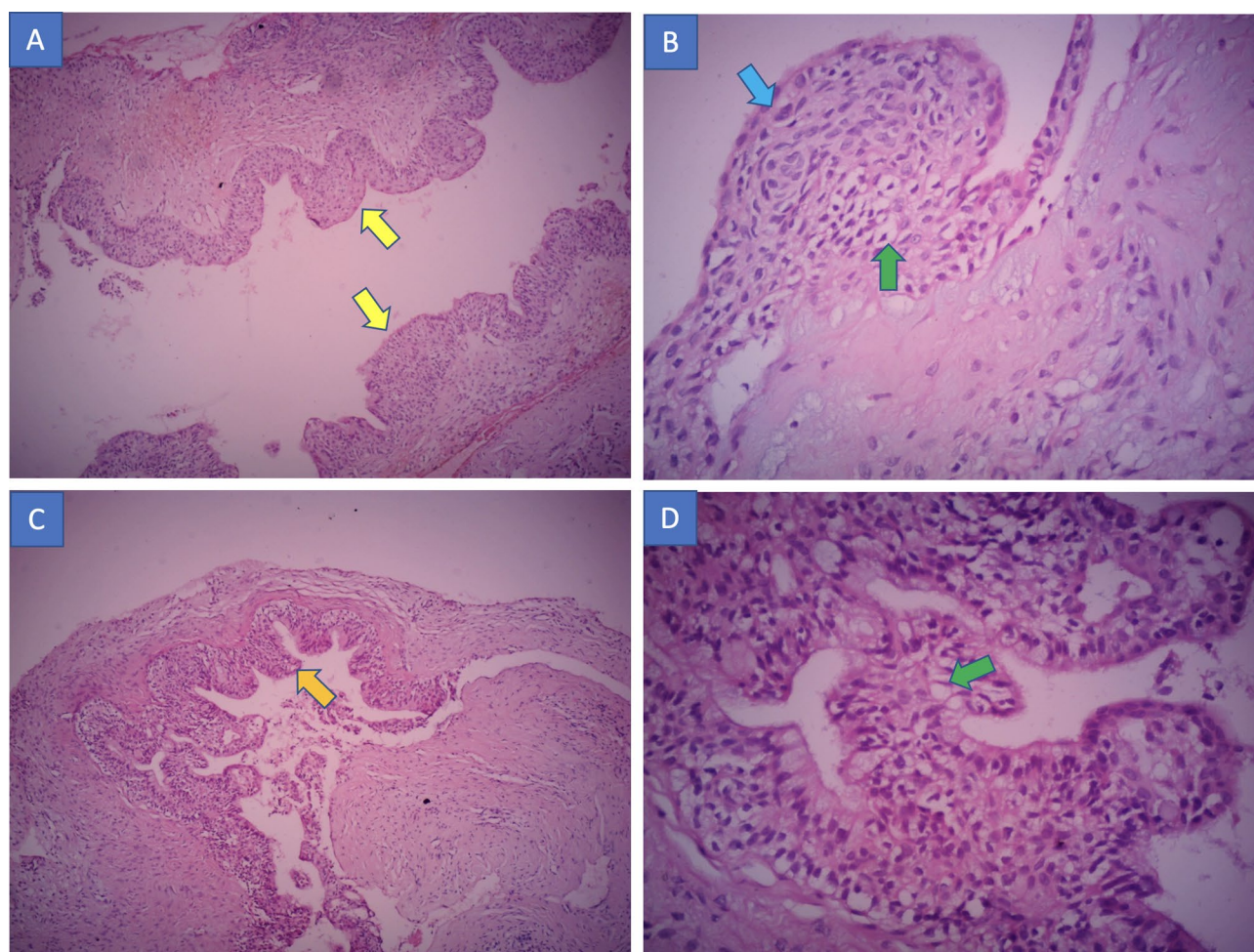


Fig. 7 Microscopic examination: (A) 10X magnification reveals the cyst wall lined by stratified squamous epithelium which is 3–5 cells thick in most areas (yellow arrow). (B) 40X magnification shows foci of glycogen-rich clear cells interspersed among the lining epithelial cells (green arrow), along with focal nodular areas of epithelial thickening exhibiting a swirling architectural pattern (blue arrow). (C) 10X magnification highlights epithelial thickenings within the lining epithelium (orange arrow). (D) 100X magnification reveals foci of glycogen-rich clear cells scattered among the lining epithelial cells (green arrow)

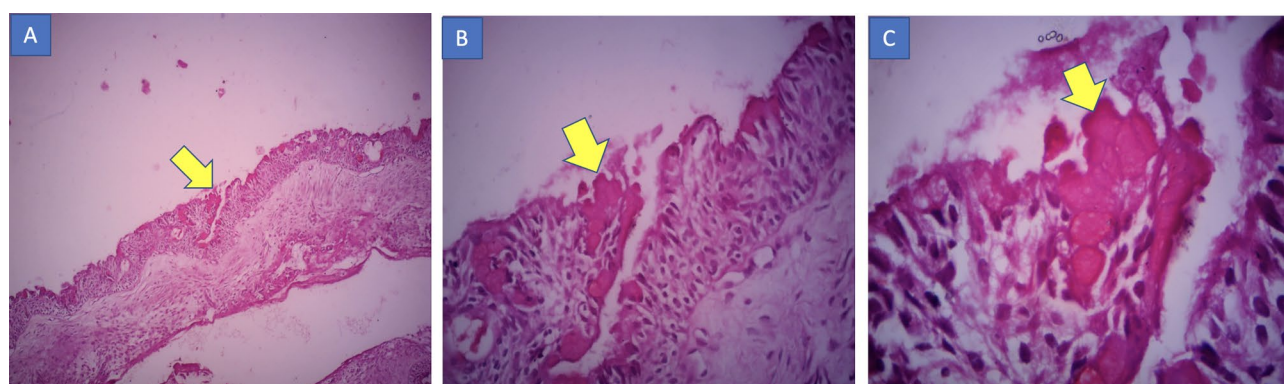


Fig. 8 (A) 10x magnification, (B) 40x magnification and (C) 100x magnification images demonstrating periodic acid–Schiff (PAS)-positive, glycogen-rich clear cells interspersed among the lining epithelial cells



Fig. 9 One-year follow-up intraoral clinical picture revealing healthy mucosa and no sign of recurrence

on the left side of the anterior maxilla, located between the lateral incisor and canine teeth. The reported case of LPCs of the maxilla are summarized in Table 1.

LPCs are generally asymptomatic unless secondarily infected and may present as a gingival swelling during their growth [5, 9, 11–13]. Cortical expansion and erosion of the cortical plate may also occur [8, 10, 14]. In the current case, the patient was asymptomatic and labial cortical expansion and erosion were observed, which aligns with the findings in the literature.

Adjacent teeth to the LPCs remain vital unless affected by caries or periodontitis [5, 6]. In this case, the adjacent teeth were vital and remained so after one year of treatment, consistent with previous reports.

The differential diagnosis of LPCs includes the gingival cyst of an adult, odontogenic keratocyst, lateral radicular cyst, residual cyst, glandular odontogenic cyst, and radiolucent odontogenic tumors [9, 11, 17]. A definitive

Table 1 Lateral periodontal cysts of the maxilla

S.N.	Study	Number of lesion	Age of the patient(years)	Region of teeth*
1	Cohen et al. 1984	7	54(21–82)	15–16, 14–15, 21–22, 3 in 23–24, 24–25
2	Rasmusson et al. 1991	4	55(26–77)	12–13, 13–14, 2 in 14–15
3	Carter et al. 1996	3	41, U, 14	13–14, 16, 22–23
4	Kerezoudis et al. 2000	1	62	13–14
5	Ortega et al. 2007	1	31	U
6	Formoso Senande et al. 2008	8	37 (18–71)	All 8 in 22–23
7	Nikitakis et al. 2010	1	70	22–23
8	Kumuda Arvind Rao et al. 2012	1	13	24–25
9	Govil S, et al. 2013	2	14	11–12, 21–22
10	Salaria, et al. 2017	1	45	21–22
11	Meseli, et al. 2019	1	32	12–13
12	Ilias Karveleas et al. 2020	1	59	13–14
13	Davi-da Silva Barbi-rato et al. 2020	1	43	14–15
14	Shi-Qiang Wen, et al. 2023	1	74	23–24

Abbreviation: U: Unknown

*The tooth numbering is in the FDI (Federation Dentaire Internationale) system

diagnosis can be confirmed through detailed histopathological examinations [5, 9, 15].

Radiographically, LPCs typically appear as unilocular, round, oval or teardrop-shaped radiolucent lesions with a sclerotic border, located laterally to the tooth roots within the alveolus [11, 12, 14]. They are present between the cervical region and the apex of the tooth roots and can

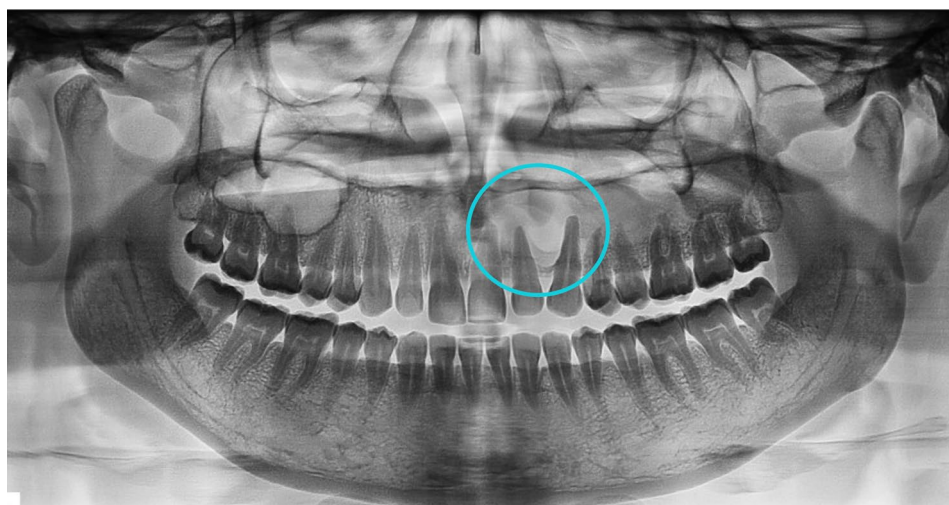


Fig. 10 One-year follow-up orthopantomogram revealing healing radiolucent area in the left anterior maxilla, positioned between the left lateral incisor and canine teeth

lead to the loss of periodontal ligament space and lamina dura of adjacent teeth [9, 15]. However, the periodontal ligament space is generally not enlarged and there is no communication between the cyst and the oral cavity [12]. In the present case, a unilocular radiolucent lesion with a sclerotic margin was noted between the lateral incisor and canine teeth, with loss of periodontal ligament space and lamina dura, in agreement with the literature.

Unlike other odontogenic cysts, which can cause the resorption of adjacent teeth, the LPC does not typically cause tooth resorption but may lead to divergence of adjacent roots [8, 10, 15]. In the present case, the adjacent roots of the maxillary lateral incisor and canine teeth were laterally splayed without resorption.

LPCs are typically less than one cm in diameter, although larger lesions have been documented [3, 9, 17, 18]. The lesion in this case measured $3.5 \times 2.4 \times 2.4$ cm, suggesting that untreated cysts have the potential to grow to considerable sizes.

The treatment of choice for LPC is enucleation [3, 7, 17]. After enucleation, the resultant bone defect may be allowed to heal spontaneously or be managed with guided bone regeneration techniques [3, 7, 17]. In this case, the cyst was treated with surgical enucleation, and the defect was left to heal spontaneously.

Histologically, LPCs exhibit characteristic features, including a lining of non-keratinized squamous or cuboidal epithelial cells, 1–5 cells thick, resembling reduced enamel epithelium. The epithelial cells are small, with pyknotic nuclei and are occasionally separated by intercellular fluid. Focal epithelial thickenings in fusiform or spindle-shaped patterns, along with clear cells containing glycogen, are often observed. The subepithelial connective tissue shows a zone of hyalinization [4, 8, 10, 12, 15]. These histopathological features were also present in the current case, which is consistent with the literature.

The LPC is believed to arise from remnants of odontogenic epithelium, though the exact tissue of origin remains debated [3, 14, 15]. It is generally considered to originate from either the dental lamina remnants, the rests of Malassez, or the reduced enamel epithelium [3, 4, 14]. Microscopic findings, such as non-keratinized epithelium, support the hypothesis of its origin from the reduced enamel epithelium [5, 12]. The presence of glycogen-rich clear cells suggests an origin from the remnants of the dental lamina, while the cyst's location adjacent to the tooth root indicates its origin from the cell rests of Malassez [5, 10, 12].

Histologically, the lining epithelium of LPC is similar to that of botryoid odontogenic cysts, although botryoid odontogenic cysts present as multicystic lesions both macroscopically and microscopically [5, 8, 9]. Radiographically, LPCs typically appear as unilocular, oval, round or teardrop-shaped radiolucencies, whereas

botryoid odontogenic cysts are characterized by multilocular radiolucencies [6, 16].

LPC recurrence is rare, with reported rates ranging from 3 to 4% [4, 8, 10, 12, 15]. However, recurrence can occur several years after initial treatment [8, 10, 15].

In conclusion, despite being a rare developmental odontogenic cyst, the LPC should be included in the differential diagnosis of cystic lesions adjacent to the root of a vital tooth. Surgical enucleation remains the treatment of choice, with histopathological examination confirming the diagnosis. Recurrence of LPC is infrequent.

Abbreviations

LPC	Lateral Periodontal Cyst
WHO	World Health Organization
OPD	Outpatient Department
HU	Hounsfield Unit
CT	Computed Tomography
VRT	Volume Rendering Technique
PAS	Periodic Acid-Schiff
FDI	Federal Dentaire Internationale

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Author contributions

Manoj Adhikari was the chief surgeon, oversaw patient follow-up and wrote the manuscript. Kanistika Jha contributed to the diagnosis, treatment planning and manuscript preparation. Shova Kunwar and Junu Bhattarai did a detailed histopathological examination of the case. Aashish Shah and Bishwo Ram Amatya provided general anesthesia during the surgery and participated in postoperative follow-up.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Competing interests

The authors declare no competing interests.

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