

Squamous Cell Carcinoma Arising in a Residual Cyst: A Case Report

Mehtap Muglali, DDS, PhD; Ayse Pinar Sumer, DDS, PhD



Abstract

Aim: The purpose of this report is to present a case of squamous cell carcinoma (SCC) arising from a mandibular residual cyst.

Background: Although rare, SCC may arise in the epithelial lining of odontogenic cysts. The diagnosis of the development of carcinoma from the cyst lining can only be established by histopathologic examination.

Report: A case of SCC arising from a mandibular residual cyst in a 55-year-old man is presented along with a discussion of the critical elements needed for accurate diagnosis and treatment.

Summary: The development of SCC from residual cysts is rare but should always be considered in the differential diagnosis. This case report clearly demonstrates the importance of clinician awareness of the malignant potential of apparently innocuous cystic lesions. It also underscores the importance of a careful histological examination and the necessity of obtaining biopsy materials from various areas to prevent a misdiagnosis of large-sized cysts.

Keywords: Residual cyst, squamous cell carcinoma, SCC, odontogenic cyst

Citation: Muglali M, Sumer AP. Squamous Cell Carcinoma Arising in a Residual Cyst: A Case Report. J Contemp Dent Pract 2008 September; (9)6:115-121.

FREE full text provided by P&G Professional Oral Health.

Introduction

Although rare, the epithelial lining of an odontogenic cyst may undergo malignant transformation. The incidence of carcinomas arising in odontogenic cysts was reported to be approximately 1-2/1000.¹ The pathogenesis is unknown, but long-standing inflammation and continuous intracystic pressure were suggested as possible causative factors.²

Differential diagnosis of odontogenic cyst and malignant tumor arising in the cyst may be difficult due to the nonspecific clinical and radiological presentation.² The definitive diagnosis must be made by histological examination.^{3,4}

Among the odontogenic cysts, malignant transformation of the keratocyst and dentigerous cyst is high.^{2,5} Although squamous cell carcinoma (SCC) arising in various developmental and inflammatory odontogenic cysts has been well established, to the best of the authors' knowledge, there has been only four reports in the English literature on the development of SCC from residual cysts.^{4,6-8} This report presents an additional case of SCC arising from a mandibular residual cyst.

Case Report

Diagnosis

A 55-year-old man presented complaining of a swelling in the right mandibular molar region. The dental history revealed he had his right mandibular second premolar extracted two months earlier. A painless swelling in the extraction area was noted by the patient four weeks after the extraction. He reported a slight paraesthesia in the right lip.

Extraoral examination revealed a slight swelling on the right mandibular region. Buccal expansion of the alveolar ridge posterior to the right mandibular first premolar was observed on intraoral examination. The mucosa covering the alveolar ridge, floor of the mouth, and the buccal vestibule was intact with no ulceration. The panoramic radiography showed a well-defined radiolucent lesion extending from the right canine to the angle of the mandible measuring 6.5 x 3 cm (Figure 1). There was no cervical lymphadenopathy.

Because of the large size of the lesion, marsupialisation was performed and the specimen

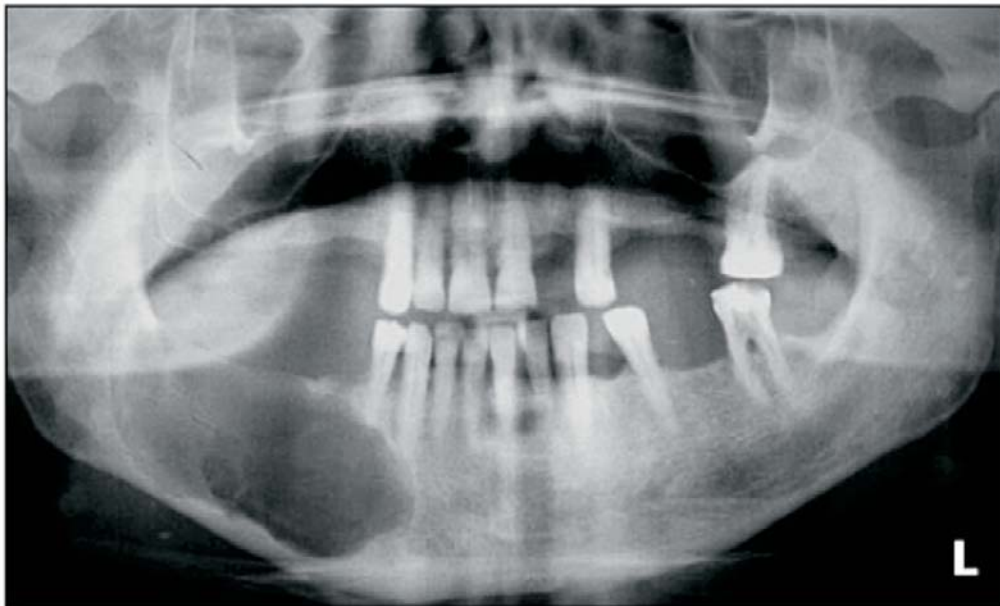


Figure 1. Panoramic radiograph demonstrating a unilocular radiolucency extending from the right premolar teeth to the angle of the mandible.

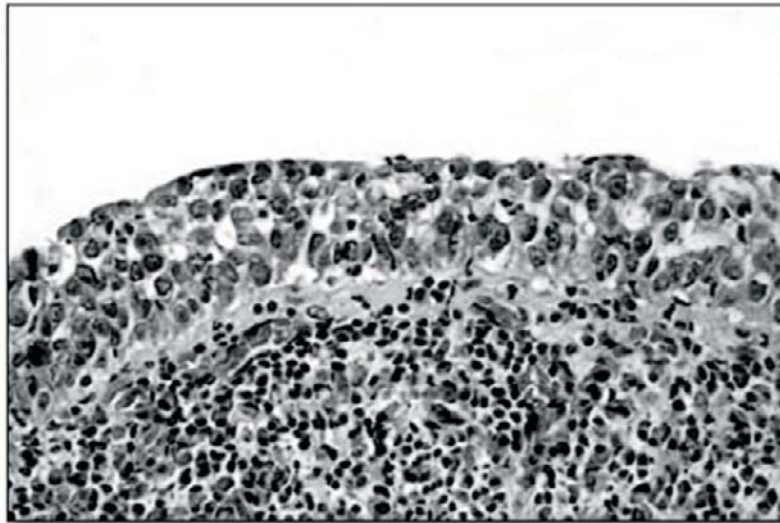


Figure 2. High power appearance of the cyst epithelium showed nuclear atypia and irregularity in maturation and organization (HE x400).

was submitted for microscopic examination. Histopathologic examination showed a full thickness of the epithelium was composed of large squamous cells. Loss of the normal cell polarity and maturation was noted. Abnormal mitotic figures could be seen over the basal layer of the epithelium. There was no invasion of the basal layer of the epithelium (Figure 2). In the light of these histological features a diagnosis of residual cyst with dysplastic features in the lining epithelium was established.

Treatment

The decision was made to totally enucleate the lesion followed by close follow-up examinations since the lesion was confined only to the epithelium without any connective tissue invasion. Enucleation of the lesion was performed under general anaesthesia using an intraoral approach.

During the enucleation it was noted the wall of the lesion was adherent to the surrounding bone. Both the lingual and buccal cortex of the mandible was

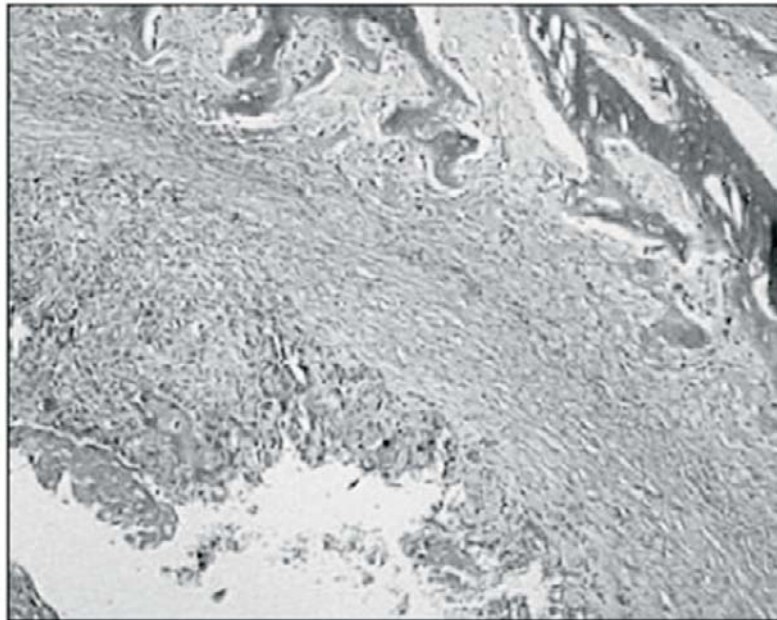


Figure 3. Histopathology of the resection specimen revealed SCC (HE x100).

thin owing to the expansion of the lesion and the neurovascular bundle was pushed inferiorly. The thin mandible was fractured in the corpus region during the enucleation process and had to be stabilized by intermaxillary fixation. The specimen was then sent for histopathological examination. This time the lesion was reported to be SCC (Figure 3).

As a result, a hemimandibulectomy was performed together with a neck dissection of 32 lymph nodes along the right posterior cervical lymph chain. The surgical site was reconstructed with a tibial bone graft. Histological examination of the resected mandible revealed the tumor invaded the surrounding bone and one of the cervical lymph nodes. The patient is under a close post-surgical follow-up regimen.

Discussion

Neoplastic transformation in the epithelial lining of an odontogenic cyst is a rare but a well-described phenomenon. The neoplasms associated with epithelial lining of the cyst include ameloblastoma, SCC, and mucoepidermoid carcinoma.⁹⁻¹³

Malignant squamous epithelium within an odontogenic cyst may represent (a) an invasion of the cyst from an adjacent primary carcinoma of the jaw, (b) a cystic change in a primary carcinoma, or (c) a malignant change within the cyst wall.¹¹⁻¹⁴ The histopathologic criterion employed to document malignant transformation of the cyst lining is the identification of a transition from the normal lining epithelium to dysplasia and to carcinoma.^{2,11,15} As in the case presented in this report, this sequence was followed. The most probable reason for the development of carcinoma seems to be due to malignant transformation in the residual cyst wall.

In a study concerning the malignant transformation of odontogenic cysts keratinization of cystic epithelium and chronic inflammation lesions were found to be the main risk factors.¹⁶ In the present case there was no keratinization of cystic epithelium, only dense chronic inflammation. Therefore, a malignant lesion was not considered and enucleation was done following marsupialisation.

In general, odontogenic cysts grow by bone expansion and the expansion is mostly to the buccal/labial vestibule. Intraosseous tumors, on the other hand, expand on both the buccal and lingual sides of the jaws.¹⁷ Therefore, the existence of a buccolingual expansion should remind clinicians of the possibility of a tumor; most probably an ameloblastoma or an intraosseous carcinoma. The probability of a malignant lesion was never of concern in the present case because of the absence of lingual expansion.

Reported clinical signs of malignant lesions generally include the presence of cervical lymphadenopathy. There was no palpable lymphadenopathy present in the present case, although a metastasis in a nodule was determined following neck dissection. Lack of a clinically palpable lymphadenopathy was misleading. When cysts reach a large size, paresthesia of the mental nerve may occur. However, the existence of paraesthesia should serve as a reminder of the possibility of an intraosseous carcinoma. In the present case, although paraesthesia of the lip together with buccal expansion should have raised the suspicion of carcinoma, the benign radiological presentation also served to mislead clinicians.

Keratocysts appear as well-defined radiolucent areas, either more or less rounded with a scalloped margin or multiloculated. Keratocyst may be confused radiographically with an ameloblastoma or with dentigerous cysts.¹⁸ Ameloblastomas have a honeycomb pattern and a single, well-defined cavity indistinguishable from a radicular or, rarely, a dentigerous cyst.¹⁹ Multilocular areas in the present case were evaluated as perforations in the buccal and lingual cortex arising from the enlarging dimensions of the residual cyst.

Enucleation is the preferred treatment of odontogenic cysts. However, when the lesion is large, marsupialisation can be performed due to the risk of fracture during the removal of the lesion by enucleation. In the present case marsupialisation was the initial treatment planned due to the large size of the lesion. However, enucleation was carried out later because the lesion had dysplastic features. As anticipated,

the mandible was fractured during the procedure despite careful manipulation. However, this case shows marsupialisation may lead to false negative results. Enucleation should be considered regardless of the risk of fracture. If marsupialisation is selected as a treatment choice, then a biopsy should be taken from different regions of the lesion. To decide on the mode of therapy based on only one biopsy from such a large lesion was a wrong approach. An initial surgical approach through a buccal window would provide specimens with a lower probability of compromising tissue by the inflammatory process caused by potential exposure to the oral cavity. The patient indicated his face swelled after the extraction of the mobile teeth. That would support the possibility a malignant change could result from a

communication with the oral cavity and concurrent exposure to the pathogenic mechanisms that affect the oral mucosa.

Summary

The development of SCC from residual cysts is rare, however, it should always be considered in the differential diagnosis.

This case report clearly demonstrates the importance of clinician awareness of the malignant potential of apparently innocuous cystic lesions. It also underscores the importance of a careful histological examination and the necessity of obtaining biopsy materials from various areas to prevent a misdiagnosis of large-sized cysts.

References

1. Stoelinga PJW, Bronkhorst FB. The incidence, multiple presentation and recurrence of aggressive cysts of the jaws. *J Cranio Maxillofac Surg* 1988; 16:184-195.
2. Bradley N, Thomas DM, Antoniadis K, Anavi Y. Squamous cell carcinoma arising in an odontogenic cyst. *Int J Oral Maxillofac Surg* 1988; 17:260-263.
3. Makowski GJ, McGuff S, Van Sickels JE. Squamous cell carcinoma in a maxillary odontogenic keratocyst. *J Oral Maxillofac Surg* 2001; 59:76-80.
4. van der Wal KG, de Visscher JG, Eggink HF. Squamous cell carcinoma arising in a residual cyst. A case report. *Int J Oral Maxillofac Surg* 1993; 22:350-352.
5. Yoshida H, Onizawa K, Yusa H. Squamous cell carcinoma arising in association with an orthokeratinized odontogenic keratocyst: report of a case. *J Oral Maxillofac Surg* 1996; 54:647-651.
6. Martinelli C, Melhado RM, Callestini EA. Squamous-cell carcinoma in a residual mandibular cyst. *Oral Surg Oral Med Oral Pathol* 1977; 44:274-278.
7. Schwimmer AM, Aydin F, Morrison SN. Squamous cell carcinoma arising in residual odontogenic cyst. Report of a case and review of literature. *Oral Surg Oral Med Oral Pathol* 1991; 72:218-221.
8. Swinson BD, Jerjes W, Thomas GJ. Squamous cell carcinoma arising in a residual odontogenic cyst: Case report. *J Oral Maxillofac Surg* 2005; 63:1231-1233.
9. Copete MA, Cleveland DB, Orban RE JR, Chen SY. Squamous carcinoma arising from a dentigerous cyst: report of a case. *Compend Contin Educ Dent* 1996;17:202-204.
10. Eversole LR, Sabes WR, Rovin S. Aggressive growth and neoplastic potential of odontogenic cyst. *Cancer* 1975; 35:270-281.
11. Gardner AF. A survey of odontogenic cyst and their relationship to squamous cell carcinoma. *J Canad Dent Assoc* 1975; 41:161-167.
12. Johnson LM, Sapp JP, McIntire DN. Squamous cell carcinoma arising in a dentigerous cyst. *J Oral Maxillofac Surg* 1994; 52:987-990.
13. Manganaro AM, Cross SE, Startzell JM. Carcinoma arising in a dentigerous cyst with neck metastasis. *Head Neck* 1997; 19:436-439.
14. Browne RM, Gough NG. Malignant change in the epithelium lining odontogenic cyst. *Cancer* 1972; 29:1199-1207.
15. Berenholz L, Gottlieb RD, Cho YS, Lowry LD. Squamous cell carcinoma arising in a dentigerous cyst. *Ear Nose Throat Journal* 1988; 67:764-772.

16. Timosca GC, Cotutiu C, Gavrilita L. Malignant transformation of odontogenic cysts. Rev Stomatology Chir Maxillofacial 1995; 96:88-95.
17. Thomas G, Sreelatha KT, Balan A, Ambika K. Primary intraosseous carcinoma of the mandible- a case report and review of the literature. Eur J Surg Oncol 2000; 26:82-86.
18. Cawson RA, Odell EW, Poeter S. (2002) Cawson's Essentials of Oral Pathology and Oral Medicine. 7th edn. UK: Churchill Livingstone.
19. Regezi JA, Sciubba JJ, Jordan RCK.(2003) Oral pathology. Clinical Pathologic Correlations. 4th edn. USA: Saunders.

About the Authors

Mehtap Muglali, DDS, PhD



Dr. Muglali is an Associate Professor in the Department of Oral and Maxillofacial Surgery of the Faculty of Dentistry at Ondokuz Mayıs University in Samsun, Turkey. She received her dental degree in 1992 and her doctorate in 1998 from Ankara University in Ankara, Turkey.

e-mail: mehtapm@gmail.com

Ayşe Pinar Sumer, DDS, PhD



Dr. Sumer is an Assistant Professor in the Department of Oral Diagnosis and Radiology of the Faculty of Dentistry at Ondokuz Mayıs University in Samsun, Turkey. She is a graduate of the University of Hacettepe Faculty of Dentistry in Ankara, Turkey. She received her PhD degree from the Faculty of Dentistry at Selcuk University in Konya, Turkey in 1997.

e-mail: psumer1970@yahoo.com