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# Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: [www.elsevier.com/locate/jomsmmp](http://www.elsevier.com/locate/jomsmmp)

## Case Report

# Sclerosing polycystic adenosis with intraluminal crystalloids of the buccal mucosa: A case report and review of the literature



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## ARTICLE INFO

### Article history:

Received 4 September 2014

Received in revised form

17 November 2014

Accepted 25 November 2014

Available online 19 December 2014

### Keywords:

Sclerosing polycystic adenosis (SPA)

Intraluminal crystalloids

Gross cystic disease fluid protein 15

(GCDFP-15)

Buccal mucosa

## ABSTRACT

Sclerosing polycystic adenosis (SPA) is a recently described, rare, neoplastic or reactive sclerosing inflammatory process of the salivary glands. It is similar to sclerosing adenosis or adenosis tumor of the mammary gland, but pathogenesis is still unclear. The commonest sites of involvement are parotid and submandibular gland, while minor salivary glands are very rare. Here we report an extremely rare case of SPA with intraluminal crystalloids in the buccal mucosa.

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## 1. Introduction

Intraluminal crystalloids have been described in the prostate [1], salivary gland [2–4], ovary [5], and breast tumors [6,7], but have not yet been reported in sclerosing polycystic adenosis (SPA) of the minor salivary gland. SPA is a rare lesion of the salivary glands, first described by Smith et al. [8] in 1996, and showing histomorphological aspects similar to fibrocystic disease of the mammary gland. The pathogenesis of SPA remains ambiguous as to whether it is reactive or neoplastic in nature [9]. Most of the 55 reported cases have occurred in the parotid gland [8,10–15]. On the other hand, nine cases affected the minor salivary gland [10,16–19], with only two that arose in the buccal mucosa [10,18]. Observation

studies of SPA in the minor salivary glands are listed in Table 1. We presented a very rare case of SPA with intraluminal crystalloids in the buccal mucosa. To our knowledge, this is the first reported case of intraluminal crystalloids identified in benign cystic ducts of SPA.

## 2. Case report

A 59-year-old Japanese man presented with a single, firm, non-tender nodule 1.5 cm in diameter in the left buccal mucosa. The surface of the lesion was smooth, non-ulcerated, and partly bullous in appearance (Fig. 1A). Although the patient had been aware of the lesion for about 10 years, he had ignored it. However, he visited the Machida Municipal Hospital with the intention of having the lesion excised, because recently he had begun to accidentally bite it. The patient's medical history was unremarkable. The clinical differential diagnosis included a benign salivary gland tumor and a mucous extravasation cyst. The lesion was resected under local anesthesia. The surgical specimen was a 15-mm × 10-mm × 7-mm mass, and grossly, the unencapsulated lesion was focal pale cream-white in color with various-sized cysts (Fig. 1B).

Microscopically, the lesion consisted of unencapsulated, circumscribed sclerotic masses with many variable-sized cystic ducts

\* Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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**Table 1**  
Review of documented cases of sclerosing polycystic adenosis (SPA) of the minor salivary glands.

Gender/ age	Site	Histopathological finding									EBV	Ki-67	GCDFP-15	Treatment/ recurrence	Reference
		Encapsulated	Ductal epithelium			Ductal contents	Acinus	Stroma							
			Atypia	Hyperplasia	Apocrine-like/ sebaceous-like			Eosinophilic cloudy fluid/ crystalloid	Eosinophilic cytoplasmic granules	Hyalinized fibrosis (periductal/ diffusely)					
M/75	Buccal mucosa	–	+	NA	NA	+/?	NA	NA	NA	NA	NA	NA	NA	Excision/–	Gnepp et al. [18]
F/48	Maxillary mucobuccal fold	NA	– (3)	NA	+/+	+/?	+	+/-	NA	NA	NA (3)	NA (3)	NA (3)	Excision (3)/–	Noonan et al. [19]
M/80	Floor of mouth	NA		+	–/–	?/?	–	+/+	+	NA					
M/70	Hard palate	–		–	–/–	?/?	+	+/+	+	NA					
M/35	Buccal mucosa	–	–	+	+/-	NA	– (lack of acini)	+/+	+	NA	NA	NA	NA	Excision/–	Meer and Altini [10]
F/82	Floor of mouth	?	+	+	+/-	NA	–	?	+	NA	NA	+, (<1%)	+	Excision/–	Gurgel et al. [16]
M/39	Retromolar area	NA	–	+	+/-	NA	–	+/-	+	NA	+	+	NA	Excision	Swelam [17]
M/53	Posterior palatal mucosa	(3)	(3)	(cribriform)	(3,?)		(3)	(3,?)	(mild)		(3) *, ¶ (higher)	(3) (generally higher)	(3)	Excision (3)/–	
F/44	Retromolar area														
F/59	Buccal mucosa	–	+	+	+/+	+/ ‡, #	– (lack of acini)	+/+	+	+	– §, †, #	+	+	Excision/–	Present case

M, male; F, female; L, left; R, right; +, positive; –, negative; EBV, Epstein–Barr virus; \*, EBV-LMP-1 by immunohistochemistry; †, EBER-in situ hybridization (EBER-ISH); ¶, EBV-gene by RT-PCR; §, CD68 positive; ‡, diastase resistant periodic acid-Schiff (d-PAS) positive; #, Masson-Trichrome stain positive; NA, not available; ?, unknown; ( ), (total number).

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