Oral Elastofibromatous Lesions: A Review and Case Series

Mark R. Darling · Maciej Kotalowski ·
David G. MacPherson · Linda Jackson-Boeters ·
George P. Wysocki

Received: 24 March 2011 / Accepted: 9 June 2011 / Published online: 18 June 2011
© Springer Science+Business Media, LLC 2011

Abstract Elastofibromas of the oral cavity are rare, with only 5 cases reported. In this paper, we present a series of five new cases of oral elastofibromatous lesions, occurring in 4 males and 1 female, with ages ranging from 33 to 76 years. The clinical differential diagnosis includes fibroepithelial polyp or fibroma, among other connective tissue tumours. Elastofibromas probably develop as reactive lesions, for which surgical treatment is definitive.

Keywords Elastofibroma · Oral cavity · Connective tissue lesions

Introduction

Elastofibroma is a benign, usually solitary lesion, most often representing a reactive process associated with frictional irritation or trauma. It was first described by Jarvi and Saxen [1], occurring in the subscapular region (elastofibroma dorsi) as a painless non-fixed mass [1–4]. Mucosal lesions are rare, but have occasionally been reported [5–9].

Elastosis in the oral mucosa is a well-known phenomenon often associated with some form of mucosal irritation or trauma [10, 11]. However, elastofibromas presenting as a solitary mass resembling a neoplasm, or as a plaque-like lesion, are not well documented in the oral mucosa, and only 5 cases have been reported to our knowledge [5, 6, 8, 9]. We present a series of five new cases to add to the

current literature on oral elastofibromas and elastofibromatous lesions.

Materials and Methods

Cases of lesions that clinically resembled fibromas (traumatic fibromas, irritation fibromas, or fibroepithelial polyps), but histologically revealed a proliferation of tissue more closely resembling elastic fibers under light microscopy, between 1999 and 2010, were retrieved from the archives of the division of Oral Pathology at the University of Western Ontario. Five cases of formalin-fixed, paraffin-embedded tissue were obtained, and the clinical information and histologic features were documented. All cases were stained with hematoxylin and eosin (H&E) and with a Masson's Elastic Trichrome (MET; Verhoeff-van Gieson and elastic trichrome) stain, to determine the presence of elastic fibers. In addition, 22 negative controls (diagnosed as either focal fibrous hyperplasia, fibroma, or fibroepithelial polyp) and 4 positive controls (actinic cheilitis showing solar elastosis) were also stained with MET. Moreover, positive control tissue (large elastic artery) was used with each elastic trichrome stain in order to verify that the stain was working. Although elastic fibre digestion can be achieved using pancreatic or bacterial elastases [12], these were not readily available nor routinely done in our laboratory, and we considered the positive controls sufficient to verify the reliability of the stain.

Results

Control cases (focal fibrous hyperplasia, fibroma, or fibroepithelial polyp) were negative or had minimal

M. R. Darling (✉) · M. Kotalowski · D. G. MacPherson ·
L. Jackson-Boeters · G. P. Wysocki
Division of Oral Pathology, Department of Pathology,
University of Western Ontario, London, ON N6A 5C1, Canada
e-mail: mark.darling@schulich.uwo.ca

amounts of staining for elastic fibers. All lesions in cases 1–5 were relatively similar in histologic appearance and are illustrated by representative photomicrographs (Figs. 1, 2, 3, 4, 5). The five cases of elastofibroma are presented as the following series:

Case #1

A 33-year-old male presented with an asymptomatic, 2×2 mm, non-ulcerated lesion on the left side of his palate. The lesion was completely excised and a clinical diagnosis of fibroma was submitted. Histopathologic examination of the tissue using a standard H&E stain revealed a mass of fibrous tissue with an overlying orthokeratinized squamous epithelium that displayed an irregular acanthosis (Fig. 1a). No inflammation or malignancy was noted. Subsequent staining with MET revealed that the mass of fibrous tissue was almost exclusively made up of elastic fibers (Fig. 1b).

Fig. 1 **a** Case 1: elastofibroma showing a raised nodular growth. (H&E stain; original magnification $\times 100$; inset $\times 25$). **b** Case 1: elastic fiber proliferation shown with MET stain (original magnification $\times 100$; inset $\times 25$)

Fig. 2 Case 2: proliferation of elastic fibers, resembling elastosis in many areas (H&E stain; original magnification $\times 400$)

Fig. 3 **a** Case 3: This elastofibroma was described clinically as a “leukoplakia”, and shows an apparent raised area microscopically (H&E stain; original magnification $\times 100$; inset $\times 25$). **b** Case 3: Elastic fiber proliferation shown with MET stain (original magnification $\times 100$; inset $\times 25$)

Case #2

A 43-year-old male presented with an asymptomatic, $3 \times 2 \times 1$ mm, yellow growth on the midline of his palate. The lesion was excised with a working clinical

Fig. 4 Case 3: Globular and occasional “serrated edge” configuration of elastic fibers (H&E stain; original magnification $\times 400$)

Fig. 5 Case 4: Elastic fibers dominated, showing a mostly globular arrangement (MET stain; original magnification $\times 400$)

diagnosis of condyloma. Histopathologic examination of the tissue by H&E stain revealed a mass of homogenous, slightly granular tissue that displayed features of elastosis (Fig. 2), with an overlying orthokeratinized squamous epithelium displaying mild reactive changes. No malignancy was noted. A subsequent elastic trichrome stain revealed that the mass was composed almost exclusively of elastic fibers.

Case #3

A 50-year-old male presented with a “leukoplakia” on the floor of his mouth. He had a positive tobacco history. A $7 \times 3 \times 2$ mm lesion was excised with a clinical diagnosis of hyperkeratosis. Histopathologic examination revealed deep fibroadipose tissue and a nodular homogeneous and granular zone within the submucosa, suggesting proliferation of elastic fibers, with overlying acanthotic parakeratinized stratified squamous epithelium (Fig. 3a). Microscopically, the lesion appeared raised, despite the

clinical description. There was no evidence of malignancy. An MET stain confirmed the elastic fiber proliferation (Fig. 3b).

Case #4

A 76-year-old female presented with a lesion of the midline of the lower lip. She had a history of squamous cell carcinoma at this site that had been treated with radiation, and a recurrent lesion was suspected. The lesion was excised and submitted as a $7 \times 4 \times 2$ mm tan and white ellipse of mucosa. Microscopic examination revealed a moderately thick layer of keratinized squamous epithelium exhibiting minimal dysplastic changes. The underlying connective tissue exhibited a nodular proliferation of elastic fibers, with homogenous and granular presentation. The deeper connective tissue consisted of fibrous scar tissue with mild focal chronic inflammation. No evidence of malignancy was present. A MET stain confirmed the presence of elastic fibers (Fig. 5).

Case #5

A 75-year-old male presented with a $5 \times 4 \times 4$ mm white nodule on the right side of his tongue. Biopsy was performed with the clinical diagnosis of fibroma. An H&E stain revealed a nodular, homogenous lesion and a MET stain confirmed that the mass was composed predominantly of elastic fibers.

A summary of clinical findings from previously reported cases and the current series is presented in Table 1. A fairly wide age range is represented (33–98 years), with no gender predilection. Most lesions arose either in the floor of mouth or the palate. In our series, only one case had a history of trauma in the form of prior surgery (case 4), and while it was suspected in 3 other cases (cases 1, 2, and 5), a supportive history was not given. In 3 of the previously reported cases, trauma was the proposed etiology.

Discussion

Elastofibromas of the skin (elastofibroma dorsi) are tumour-like lesions composed of elastic and fibrous connective tissue, and are frequently associated with local trauma [5]. Most lesions appear to occur in middle aged to older patients, and more commonly in females. Patients usually have a history of intense repetitive manual labour [12]. Subsequent to the initial report by Jarvi and Saxen [1], numerous additional sites have been reported, including the small bowel, tracheobronchial tree, colon, omentum, deltoid region, foot, stomach and rectal submucosa, amongst others [4, 5, 7, 12–14]. Most commonly, it

Table 1 Summary of data for cases of reported oral elastofibroma

References	Age	Gender	Location	Aetiology	Treatment	Follow-up
Potter et al. [5]	56	F	Floor of mouth	Trauma?	Surgical excision	2 years, NER
Manchandu et al. [6]	71	M	Floor of mouth	Prior surgery, radiation therapy?	Surgical excision	14 months, NER
Tosios et al. [8]	76	F	Floor of mouth	?	NS	NS
Tosios et al. [8]	98	F	Alveolar mucosa	Prior surgery?	NS	NS
Nonaka et al. [9]	55	M	Soft Palate	Trauma: Oro-tracheal intubation?	Surgical excision	8 months, NER
Current report	33	M	Palate	Trauma?	Surgical excision	1 year, NER
Current report	43	M	Palate	Trauma?	Surgical excision	9 years, NER
Current report	50	M	Floor of mouth	Trauma?	Surgical excision	3 years, NER
Current report	76	F	Lower lip	Prior surgery, radiation therapy?	Surgical excision	10 years, NER
Current report	75	M	Tongue	Trauma?	Surgical excision	6 months, NER

NS not stated, NER no evidence of recurrence

presents as a slow growing firm deep-seated mass which is usually painless [12].

Elastofibromas are ill-defined, oblong or spherical masses of varying size, not larger than 10 cm. Microscopically, they consist of swollen eosinophilic collagen and elastic fibers in about equal proportions, with small amounts of interstitial myxoid material and some fibroblasts [5, 12]. The elastic fibers often have a degenerated beaded appearance, or are fragmented into a flower-like or “petaloid” arrangement [12], which was seen focally in one of our cases. Elastic stains show branched and non-branched fibers that have a central dense core and an irregular margin. In the cases described here, the elastic fibers showed mostly a globular, and only occasional “serrated edge,” configuration (Fig. 4) [12]. In all our cases, elastic fibers dominated, as confirmed by the elastic trichrome stains (Fig. 5), rather than showing an equal distribution of collagen and elastic fibers; in this respect, these cases resembled an elastosis rather than the typical histopathologic features usually seen in elastofibroma dorsi. This was consistent with most of the previously described cases in the oral cavity [5, 6, 8, 9].

The differential diagnosis in the oral cavity would include fibrous proliferations, such as localized fibrous hyperplasia, irritation or traumatic fibromas, and fibroepithelial polyps, all of which would show similar histologic features. However, elastic stains would show no to minimal elastic fibers in the aforementioned lesions. Since focal fibrous hyperplasia and irritation or traumatic fibromas are relatively common intraoral lesions, it is possible that many oral elastofibromatous lesions have been misdiagnosed as such. However, this would carry no clinical significance as the outcome and prognosis are similar.

The cause of the abnormal elastic fibers has been debated, with proposals ranging from elastic degeneration of collagen fibers, degeneration of pre-existing elastic fibers, or disturbed elastic fiber production. According to Weiss, evidence suggests that elastofibromas are

degenerative pseudotumors that result from the excessive formation of collagen and abnormal elastic fibers [5, 12]. One study suggested a genetic predisposition, which was seen in about one-third of patients in their series [15].

Although elastic fibers form a significant proportion of the oral submucosal tissues, disorders of elastic tissue in the oral cavity have rarely been described, with some exceptions. In fact, elastosis is routinely seen in several lesions in the oral cavity that are associated with some form of irritation: solar elastosis associated with actinic cheilitis and actinic keratosis [11]; trauma-induced elastosis associated with repetitive movement of a denture flange against vestibular tissues during mastication or jaw-clenching [10]; elastosis in tumours of the salivary glands [16–18]; and elastosis associated with intra-oral epithelial dysplasia and in carcinoma-in situ (unpublished observations). In the oral cavity, altered elastic fiber production is most likely associated with local trauma. In this report, only 1 case (case 4) gave a supportive history, although it could not be definitively excluded in the other 4 cases.

In conclusion, we present several oral elastofibromatous lesions. Although rare, these lesions may appear clinically identical to other small exophytic oral mucosal lesions. They probably develop as a reaction to local irritation or trauma of some type. Surgical excision is the treatment of choice, and recurrence has not been reported. Clinical distinction from fibromas or fibroepithelial polyps would be of academic value, since there is no difference in clinical behaviour, but the general dental practitioner and specialist should be aware that these lesions can occur in the oral cavity.

References

1. Jarvi O, Saxen E. Elastofibroma dorse. Acta Pathol Microbiol Scand Suppl. 1961;51(Suppl 144):83–4.

2. Jarvi OH, Saxen AE, Hopsu-Havu VK, Wartiovaara JJ, Vaissalo VT. Elastofibroma—a degenerative pseudotumor. *Cancer*. 1969;23:42–63.
3. Jarvi OH, Lansimies PH. Subclinical elastofibromas in the scapular region in an autopsy series. *Acta Pathol Microbiol Scand A*. 1975;83:87–108.
4. Muratori F, Esposito M, Rosa F, et al. Elastofibroma dorsi: 8 case reports and a literature review. *J Orthop Traumatol*. 2008;9:33–7.
5. Potter TJ, Summerlin DJ, Rodgers SF. Elastofibroma: the initial report in the oral mucosa. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2004;97:64–7.
6. Manchandu R, Foote J, Alawi F. Elastofibroma presenting as an oral soft tissue mass. *J Oral Pathol Med*. 2008;37:125–6.
7. Markl B, Kerwel TG, Langer E, et al. Elastosis of the colon and the ileum as polyp causing lesions: a study of six cases and review of the literature. *Pathol Res Pract*. 2008;204:395–9.
8. Tosios KI, Economou I, Vasilopoulos NN, Koutlas IG. Elastofibromatous changes and hyperelastosis of the oral mucosa. *Head Neck Pathol*. 2010;4:31–6.
9. Nonaka CF, Rego DM, Miguel MC, de Souza LB, Pinto LP. Elastofibromatous change of the oral mucosa: case report and literature review. *J Cutan Pathol*. 2010;37:1067–71.
10. Sharma AK, Mirza FD. Palatal mucosa under dentures: a qualitative histologic and histochemical analysis. *J Prosthet Dent*. 1986;56:574–82.
11. Chi AC. Epithelial pathology. In: Neville BW, Damm DD, Allen CM, Bouquot JE, editors. *Oral and maxillofacial pathology*. St. Louis, Mo: Saunders/Elsevier; 2009. p. 404–6.
12. Weiss SW, Goldblum JR. Benign fibrous tissue tumours. In: Weiss SW, Goldblum JR, editors. *Enzinger and Weiss' soft tissue tumors*. 5th ed. Philadelphia: Mosby Elsevier; 2008. p. 207–12.
13. Hsu JK, Cavanagh HD, Green WR. An unusual case of elastofibroma oculi. *Cornea*. 1997;16:112–9.
14. McPherson FC, Norman LS, Truitt CA, Morgan MB. Elastofibroma of the foot: uncommon presentation: a case report and review of the literature. *Foot Ankle Int*. 2000;21:775–7.
15. Nagamine N, Nohara Y, Ito E. Elastofibroma in Okinawa. A clinicopathologic study of 170 cases. *Cancer*. 1982;50:1794–805.
16. Adkins KF, Daley TJ. Elastic tissues in adenoid cystic carcinomas. *Oral Surg Oral Med Oral Pathol*. 1974;38:562–9.
17. Azzopardi JG, Zayid I. Elastic tissue in tumours of salivary glands. *J Pathol*. 1972;107:149–56.
18. David R, Buchner A. Elastosis in benign and malignant salivary gland tumors. A histochemical and ultrastructural study. *Cancer*. 1980;45:2301–10.