Solitary myofibroma of the adult mandible: a case report

D. J. Brierley, BDS (Hons), MFDS RCS (Ed),[a] S. A. Khurram, BDS, MSc, PhD, MFDS RCS (Ed),[b] and P. M. Speight, BDS, PhD, FDSRCPS, FDSRCS (Eng), FDSRCS (Ed), FRCPath,[c] Sheffield, UK
THE SCHOOL OF CLINICAL DENTISTRY

A 43-year-old woman presented with a swelling in the anterior mandible appearing radiographically as a well-defined radiolucency causing mobility of the anterior teeth. A clinical diagnosis of a radicular cyst led to removal of the lesion and the associated mobile teeth. Postoperative histopathology led to a diagnosis of intraosseous solitary myofibroma of the mandible. Solitary lesions of myofibroma are exceedingly rare in adult jaws, with only 3 previously documented cases.
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Solitary myofibroma is a rare benign soft tissue tumor that most commonly presents in adults, with a predilection for the head and neck region.[1] Myofibromas of the oral tissues (either solitary or multiple), are well recognized, with numerous published cases.[2,3] However, solitary myofibroma is viewed as a separate entity, distinct from the multiple lesions of myofibromatosis.[4] Intraosseous solitary myofibromas have also been reported in adults and children,[5-7] but are exceedingly rare in adult jaws, with only 3 mandibular cases having been reported (Table I).[8-10] The remaining intraosseous lesions have been reported in children.[2,3,5]

CASE REPORT

A 43-year-old female patient was referred with a 2-month history of a swelling involving the anterior mandible. The patient had the lower left lateral incisor extracted 2 months before initial presentation with no reduction in the swelling. The tooth was mobile and was extracted by the patient's dentist who assumed periodontal disease. No further clinical or radiographic investigations had been carried out. At the time of the referral, the lower left central incisor and canine had also become mobile. Clinical examination revealed an ulcerated area adjacent to the site of the previously extracted lateral incisor in addition to a firm erythematous swelling approximately 1 cm in diameter extending between the mandibular left central incisor and canine tooth.

[a]SHO Oral Medicine and Pathology, Unit of Oral and Maxillofacial Pathology, The School of Clinical Dentistry, Sheffield, UK.
[b]Academic Clinical Lecturer and Honorary Specialist Registrar, Unit of Oral and Maxillofacial Pathology, The School of Clinical Dentistry, Sheffield, UK.
[c]Professor in Oral and Maxillofacial Pathology, Honorary Consultant Histopathologist and Dean of Clinical Dentistry, Unit of Oral and Maxillofacial Pathology, The School of Clinical Dentistry, Sheffield, UK.
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A panoramic radiograph showed a well-demarcated, unilocular radiolucency in the anterior mandible, appearing to extend beyond the cortical plates with perforation of the superior alveolar bone (Fig. 1). A clinical diagnosis of a radicular or other odontogenic cyst was made. Treatment involved extraction of the lower left central incisor and canine tooth with curettage of the lesion.

Initial histologic examination by the regional histopathology unit suggested a differential diagnosis of a fibro-osseous lesion or nodular fasciitis. The case was referred for further specialist maxillofacial pathology input to arrive at a definitive diagnosis.

Histologic examination showed a well-demarcated tumor mass composed of sheets, islands, and fascicles of spindled cells (Fig. 2), arranged in a slight "organoid" pattern, with prominent storiform areas. Overall, the lesion had a slight biphasic appearance containing lighter staining; plump spindle cells with elongated nuclei; and hyperchromatic, mildly pleomorphic, polygonal cells (Fig. 3). Tumor was seen to infiltrate muscle at the margins. Centrally, large dilated vessels with a branching, "stag-horn" morphology were noted, as well as areas with a hyalinized stroma. Reactive bone with a slight psammomatoid pattern was also noticed at the deep aspect. Immunohistochemistry revealed the lesion to be negative for cytokeratins, S100, and desmin but positive for smooth muscle actin (Figs. 4 and 5). CD34 and CD31 showed prominent blood vessels, although the lesional tissue was negative (Fig. 6).

The lesion was diagnosed as an intraosseous solitary myofibroma.

DISCUSSION

Mandibular myofibromas often cause no symptoms, tending to be asymptomatic at the time of diagnosis, with an intraoral or extraoral swelling forming the initial presentation.[5] Radiologically, it is usually a unilocular radiolucent lesion, with a well-defined border and slow growth rate.[5] It also has a tendency to thin and expand the cortical plates, imitating the biological behavior of odontogenic lesions.[5]

Histologically, the lesion is characterized as a well-circumscribed mass with a biphasic pattern of smaller polygonal cells with pale open nuclei, admixed with

Table I. Summary of previously documented cases of solitary myofibroma affecting the adult jaws

Study	*Gender*	*Age*	*Site*	*Treatment*
Oliver et al.[8]	Female	34	Ramus of mandible	Surgical excision with bone graft
Sedghizadeh et al.[9]	Male	20	Body of mandible and ramus	Surgical excision and reconstruction using bone graft, costochondral graft, and titanium mesh tray
Ramadorai et al.[10]	Female	32	Body of mandible (premolar and molar region)	Surgical excision and reconstruction with stainless steel plate
Current case	Female	43	Anterior mandible	Curettage of lesion and extraction of associated mobile teeth

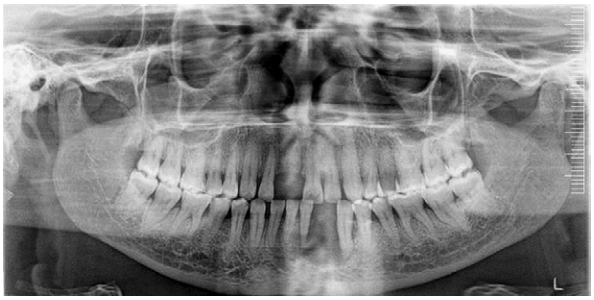

Fig. 1. Panoramic radiograph showing unilocular radiolucency in the anterior mandible.

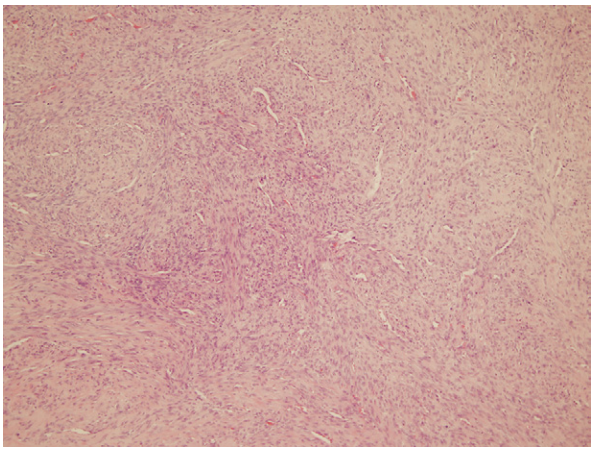

Fig. 2. Histologic appearance of lesion with hematoxylin and eosin staining. The lesion has a prominent vascular pattern and is biphasic with zones of polygonal cells and plumper spindle cells arranged in fascicles and whorls. (Magnification x4.)

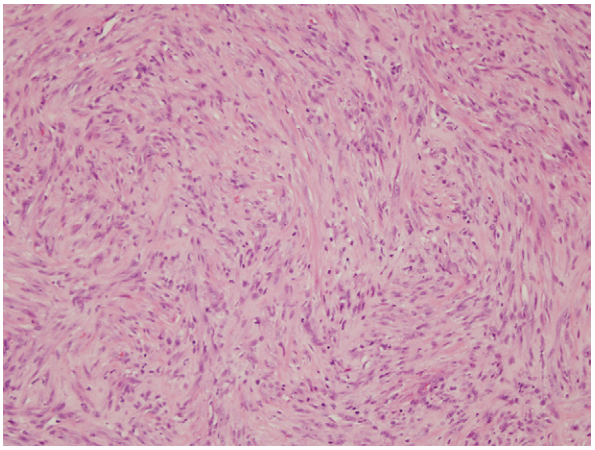

Fig. 3. Higher power view of lesion with hematoxylin and eosin staining showing areas composed of darkly staining polygonal cells and spindle cells with elongated, cigar-shaped nuclei. (Magnification x10.)

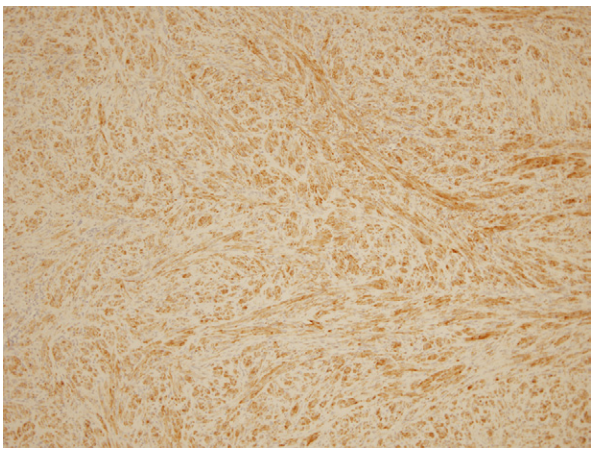

Fig. 4. Immunohistochemistry showing diffuse positivity for smooth muscle actin. (Magnification x10.)

eosinophilic spindle-shaped cells that usually form fascicles and a storiform pattern.[1,11] The biphasic nature of the lesion is caused by the transverse and longitudinal sectioning of the spindle cells, which may also result in a characteristic organoid pattern. Infiltration of adjacent muscle is frequently seen[12] and was also recorded in a previous report of a solitary lesion in the adult mandible.[8]

The differential diagnosis of myofibroma can be wide and should include a range of spindle-cell lesions. In particular, other myofibroblastic lesions, such as nodular fasciitis should be considered, as well as fibrous histiocytoma, neurofibroma, and smooth muscle tumors. Nodular fasciitis was considered by the referring pathologist but the lesion lacked both a prominent myxoid stroma and scattered chronic inflammatory cells. The tumor also displayed a hemangiopericytomalike vascular pattern, which is not seen in nodular

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