# case report

## Ameloblastic fibroma

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**Background.** Ameloblastic fibroma (AF) is a rare odontogenic tumour. It consists of odontogenic ectomesenchyme resembling the dental papilla and epithelium resembling dental lamina and enamel organ without dental hard tissues.

**Case report.** A case report of a large ameloblastic fibroma involving the body of mandible from the lower left second incisor (32) to the lower left second molar (37) is presented. To our knowledge this is the only case of ameloblastic fibroma reported from Slovenia.

**Conclusions.** An aggressive surgical treatment is suggested because of the possibility of recurrence and the possibility of malignant transformation of an AF to an ameloblastic fibrosarcoma.

Key words: fibroma; odontoma; mandibula neoplasms

#### Introduction

Despite Ameloblastic fibroma (AF) is a rare odontogenic tumour, it occurs predominantly in children and therefore remain an important diagnostic consideration. It usually arises from the mandibular dentition although it can arise in maxilla. 2

AF consists of odontogenic ectomesenchyme resembling the dental papilla and epithelium resembling dental lamina and

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enamel organ without dental hard tissues.<sup>3</sup> Knowledge of the malignant potential in the mesenchymal spindle cells of AF should assist in determining the management of these benign tumours, and may prevent malignant transformation to ameloblastic fibrosarcoma.<sup>4</sup>

At the Clinical Department of Maxillofacial and Oral Surgery in Ljubljana this is the only case of this tumour and to our knowledge the only one in Slovenia. Table 1 presents the frequency of odontogenic tumours from June 1995 to June 2005.

### Case report

A 23-years old Caucasian asymptomatic woman visited her dentist in March 2004. On dental panoramic tomogram (DPT) a radiolucent lesion formed from three separate com-

Tumour	Number of cases	Relative frequency among odontogenic tumours (%)
Ameloblastoma	20	35.1
Odontogenic myxoma	3	5.3
Ameloblastic fibroma	1	1.8
Adenomatoid odontogenic tumour	2	3.5
Ameloblastic Fibrodentinoma	1	1.8
Cementoma	1	1.8
Odontogenic Fibroma (peripheral)	4	7.0
Calcifying Odontogenic Cyst	4	7.0
Complex Odontoma	21	36.8
Total	57	100

**Table 1.** Odontogenic tumours seen at the Clinical Department of Maxillofacial and Oral Surgery in Ljubljana, Slovenia from June 1995 to June 2005.

partments from the region 32 to 36 was found (Figure 1A). In February 2005 another DPT showed the lesion had increased. The roots of the teeth 35 and 36 were resorbed. The lesion was presumed to be a cyst so she was not sent to a maxillofacial surgeon before May 2005.

The patient's family anamnesis was positive for neoplasms, diabetes and coronary heart disease. Her brother has Down's syndrome.

A large hard swelling under intact soft tissues was palpated in the lower left vestibulum, all teeth were vital.

A pathohistologic examination showed strands of odontogenic epithelium. In the centre the cells were focally similar to the embryonic stellate reticulum. The epithelial islands were surrounded by a rich mesenchymal component reminiscent of the dental papilla cells. Rare mitoses were present and the nuclear polymorphism was minimally expressed.

The teeth 31, 32, 33 and 37 were endodontically filled before surgery (Figure 1B), and then the extirpation of the tumour and the surrounding bone with nerve preservation was done. The teeth 33, 35 and 36 were extracted and the teeth 31, 32 and 37 were apiectomized (Figure 1C).

The postoperative course was uneventful with good bone regeneration (Figure 1D). A

long-term follow-up and an implant-prosthodontic treatment is planned.

#### Discussion

The relative frequency of AF among odontogenic tumours seen at the Clinical Department of Maxillofacial and Oral Surgery in Ljubljana (1.8%) is within the frequencies described in the literature,<sup>5</sup> although in some other countries it is more frequent.<sup>6</sup> The age of described patient is above the mean age at presentation being 14.8 years.<sup>3</sup> Our AF was incidentally found as 17% of the cases in a survey of 24 cases of AF from the Armed Forces Institute of Pathology.<sup>7</sup>

An aggressive surgical treatment is suggested by some authors because of the possibility of malignant transformation of an AF to an ameloblastic fibrosarcoma. 8,9 Muller *et al.*8 reported that since 1960 44% cases of ameloblastic fibrosarcoma (19/43) arose from AF. There is also the consideration that the majority, if not all, of AFs are true neoplasms with a potential to recur and/or of malignant transformation and that some, especially those occurring during childhood, could represent the primitive stage of a developing odontoma. 10

The recurrence rate of AF found by Trodahl *et al.*<sup>7</sup> was 43.5%; on the other hand

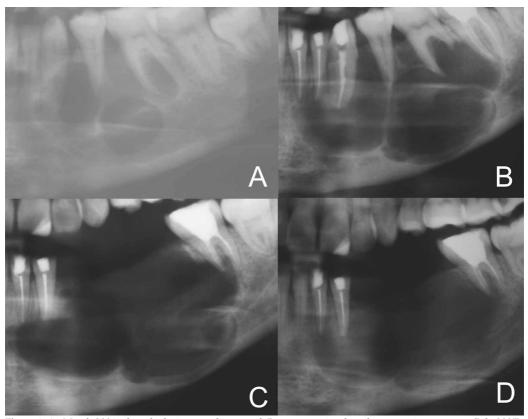


Figure 1. A - March 2004 when the lesion was discovered; B - preoperative dental panoramic tomogram (July 2005); C - postoperative dental panoramic tomogram; D - two months after the operation.

by Zallen *et al.*<sup>11</sup> it was 18.3% after reviewing the literature with 85 cases of AF. Lysell and Sund<sup>12</sup> proposed the incomplete primary removal as a reason of recurrence in their cases. This was supported by Mosby *et al.*<sup>13</sup> explaining that after the complete removal of a tumour clinically, this cannot be stated at a cellular level. They suggest the conservative removal of AF and modified block resection of any recurrence. No matter what the reason of recurrence is, all authors agree that a long-term follow-up is necessary.

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# Zmanjšana moč vratnih iztegovalnih mišic (sindrom padajoče glave) kot posledica obsevalnega zdravljenja

### Bhatia S, Miller RC, Lachance DL

**Izhodišča.** Sindrom padajoče glave je redka bolezen, pri kateri bolnik ne more držati glave vzravnano v anatomski legi. Nastane zaradi zmanjšanja moči vratnih iztegovalnih mišic.

**Prikaz primera.** Prikazujemo primer sindroma padajoče glave pri bolnici, ki je bila pred desetimi leti obsevana zaradi Hodgkinovega limfoma, predhodno pa se je zdravila tudi zaradi multiple skleroze. Razpravljamo o možnih vzrokih nastanka bolezni.

**Zaključki.** Najbolj verjeten vzrok nastanka bolezni je bila poškodba prednjega roga hrbtenjače ob obsevanju, kar je povzročilo poškodbo spodnjega motoričnega nevrona in mišično atrofijo.

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# Ameloblastični fibrom

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**Izhodišča.** Ameloblastični fibrom je redek odontogeni tumor. Sestavljen je iz odontogenega ektomezenhima podobnega dentalni papili ter epitelija podobnega dentalni lamini in sklenini, je brez trdih zobnih tkiv.

**Prikaz primera.** Opisujemo bolnico z velikim ameloblastičnim fibromom, ki se je razraščal v spodnji čeljustnici in je segal od spodnjega levega drugega sekalca (32) do spodnjega levega drugega kočnika (37). Po do sedaj zbranih podatkih je to prvi opisani primer ameloblastičnega fibroma v Sloveniji.

**Zaključki.** Zaradi možnosti ponovitve bolezni pa tudi spremembe ameloblastičnega fibroma v ameloblastični sarkom svetujemo radikalno kirurško zdravljenje in daljše pooperativno sledenje bolnikov.