

Case Report

Low-grade malignant fibrosarcoma of the dental follicle of an unerupted third molar without clinical evidence of any follicular lesion

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SUMMARY. Background: Sarcomas are rarely seen in the head and neck region. They make up less than 1% of all malignant head and neck tumours. Not only the location, size and systemic manifestation, but also the histological differentiation plays an essential role in establishing the best treatment. If resectable, surgical removal of the tumour with clear margins is the preferred method. Adjuvant chemo- and/or radiotherapy might be considered in the case of high-grade lesions or narrow surgical margins. Patient: This paper reports a 23-year-old female patient who underwent routine surgical removal of unerupted wisdom teeth without clinical evidence of any follicular lesion and was diagnosed with a low-grade malignant fibrosarcoma of the dental follicle of the lower left unerupted third molar. Conclusion: This case report raises the question as to whether histological examination of all removed dental follicles should be carried out on a routine basis. © 2007 European Association for Cranio-Maxillofacial Surgery

Keywords: fibrosarcoma, head and neck cancer, dental follicle

INTRODUCTION

Sarcomas are seen rarely in the head and neck region. They make up less than 1% of all malignant head and neck tumours (Weber et al., 1986). Not only the location, size and systemic manifestation, but also the histological differentiation plays an essential role in establishing the best treatment (Chen et al., 2005). If resectable, surgical removal of the tumour with clear margins is the preferred method (Mendenhall et al., 2005). Adjuvant chemo- and/or radiotherapy might be considered in the case of high grade lesions or narrow surgical margins (Colville et al., 2005).

CASE REPORT

The case of a 23-year-old female patient is described who underwent a minor oral surgical procedure in this unit under general anaesthesia in order to have her four unerupted third molars removed because of imminent dental crowding. Fig. 1 shows the pre-operative orthopantomogram which reveals no signs of bone or soft tissue pathology other than the four impacted third molars.

As the dental follicle of the lower left third molar was increased in size and slightly indurated (but otherwise normal looking), it was sent for histological

examination. Histomorphologically, mesenchymal neoplasia was seen. It was composed of spindle-shaped cells arranged in interlacing fascicles (Fig. 2). The pleomorphic tumour cells expressed only a few mitoses (Fig. 3). No necrosis was seen. Immunohistochemically, the tumour cells showed no reactivity with antibodies against smooth muscle actin (Fig. 4), actin, S100-protein, QBEND-10 (anti-CD34), vimentin, pancytokeratin, PGM-1 (anti-CD68), or desmin. The diagnosis of a low-grade malignant fibrosarcoma was made, and was verified by a soft tissue tumour panel. Staging procedures consisted of a head and neck CT-scan, chest X-ray, full body bone scintigraphy and ultrasound of the abdomen and did not reveal any evidence of metastatic disease.

When presenting the case at the interdisciplinary sarcoma conference of this hospital a further resection with a 1 cm circular safety margin including a rim resection of the mandible and removal of the lower left second molar was recommended and performed later under general anaesthesia (Fig. 5). Both the lingual nerve and the inferior alveolar nerve were spared. In order to protect the mandible from a pathological fracture, temporary immobilization of the lower jaw was carried out by means of orthodontic splinting.

Forty months after the last surgical procedure the patient is in a good general health and there are no clinical or radiological signs of recurrence.

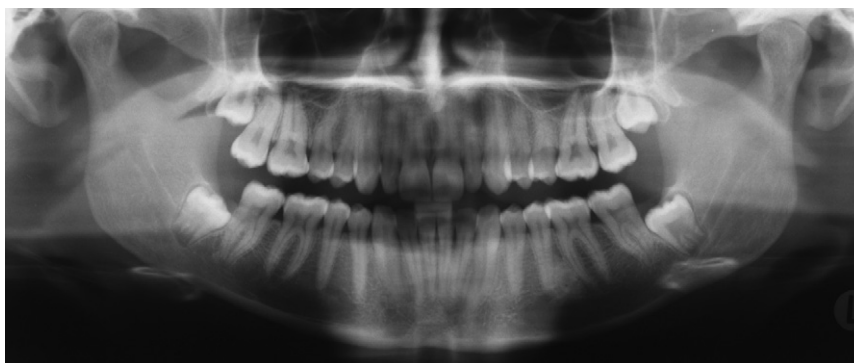


Fig. 1 – 23-year-old female. Preoperative orthopantomogram.

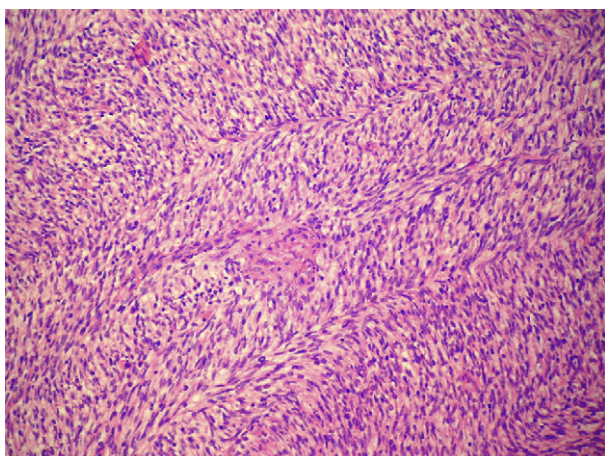


Fig. 2 – Low-grade fibrosarcoma from the dental follicle of the lower left third molar. Parallel arrangement of spindle cells in a fascicular pattern (H&E, $\times 180$).

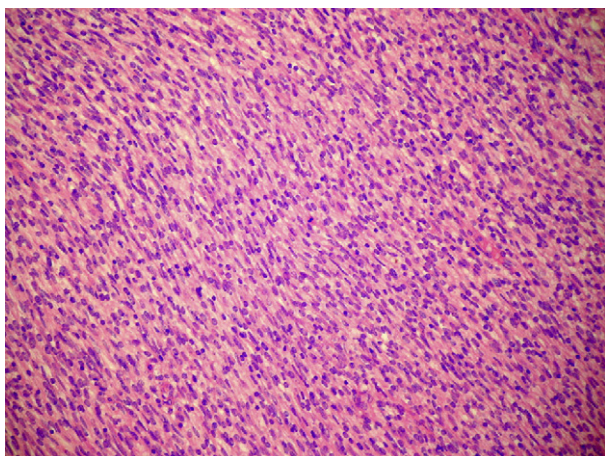


Fig. 3 – The spindle cells of this low-grade fibrosarcoma have slightly pleomorphic nuclei and only very few mitoses (H&E, $\times 420$).

DISCUSSION

Fibrosarcoma accounts for approximately 15% of all soft tissue sarcomas which represent only 1% of all malignant tumours of the head and neck region

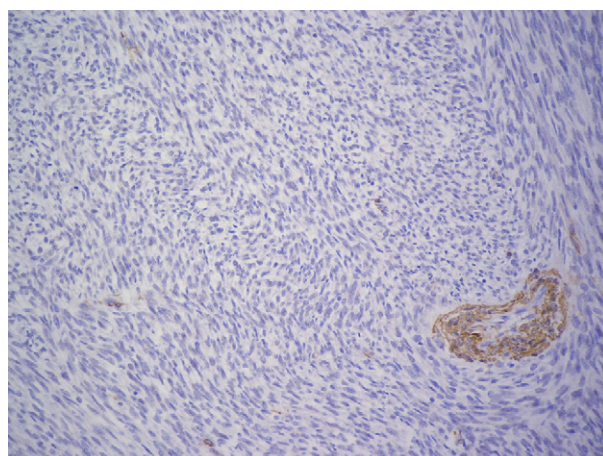


Fig. 4 – Immunohistochemistry: no reactivity of the tumour cells with the antibody against smooth muscle actin. Note the positively stained bundles of muscle cells of blood vessels. Anti-sm-actin (Avidin–biotin–complex stain, $\times 180$).

(Weber et al., 1986). Frankenthaler et al. (1990) found 118 fibrosarcomas of the head and neck region with the neck being the most common site (25%), followed by face (20%), scalp (16%), and maxillary sinus (12%). Only 12% of the fibrosarcomas were located intraorally, approximately half of them in the lower jaw.

The clinical behaviour of the fibrosarcoma is characterized by a high local recurrence rate and a low incidence of locoregional lymph node and/or distant haematogenous metastases. However, haematogenous metastasis may involve the lungs, mediastinum, abdominal cavity and bone (Conley et al., 1967; Swain et al., 1974; Weber et al., 1986). Local recurrence poses a serious and complex problem with occurrence of infiltration, local destruction, airway compression, oesophageal compression and extension into the mediastinum (Conley et al., 1967; Swain et al., 1974; Odell, 1996).

Before therapy, the local extent of the neoplasm and the presence or absence of local and distant metastases must be determined. Contrast-enhanced head and neck C.T. has proved to be a valuable tool for delineating the size of the tumour and the infiltration of neighbouring tissue. Metastatic surveys should include chest radiography, scintigraphic bone

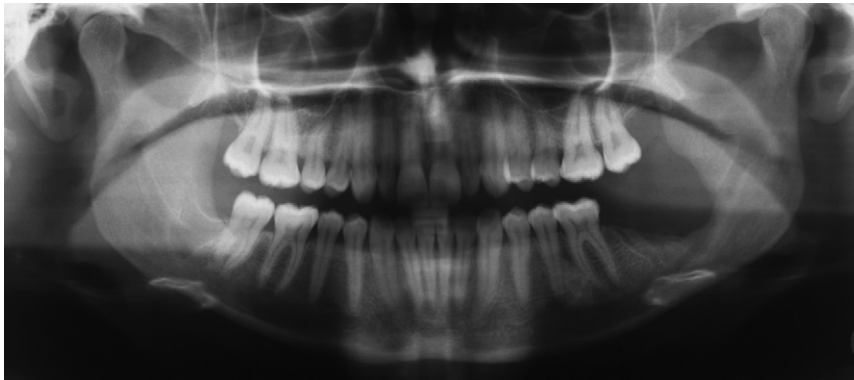


Fig. 5 – Postoperative orthopantomogram: note the defect caused by resection with 1 cm circular safety margin including removal of the lower left second molar.

scanning and abdominal ultrasound and/or computed tomography.

Wide local excision remains the treatment of choice (Conley et al., 1967; Sessions and Ogura, 1974; Frankenthaler et al., 1990; Wanebo et al., 1992; Odell, 1996; Cecchetto et al., 2001). Radiotherapy is mandatory when adequate safety margins cannot be obtained and a re-operation is not possible (Suit et al., 1985; Slater et al., 1986; Greager et al., 1994; Miser et al., 1997). Adjuvant chemotherapy for sarcomas has been applied in tumours of the trunk and extremities, as well as in the head and neck. Although some reports have raised the possibility of some benefit in certain types of sarcomas, the benefit of adjuvant chemotherapy with regard to prolonged survival remains controversial as other studies have not reported prolonged survival (Das Gupta et al., 1982; Rosenberg et al., 1983; Antman, 1997; Scurr and Judson, 2005).

To our knowledge, this is the first description of a fibrosarcoma of the dental follicle of an unerupted third molar. Data concerning other pathologic changes in pericoronal tissues associated with unerupted third molars is limited because most surgeons discard pericoronal tissues after third molar removal and do not submit the soft tissue for histological examination. Rakprasitkul (2001) histologically examined the dental follicles of 104 unerupted third molar teeth which showed no evidence of any follicular lesion. Of the 61 (59%) pathologic changes found, 53 were diagnosed as dentigerous cysts, five as chronic, non-specific inflammatory tissue, two as odontogenic keratocysts and one as an ameloblastoma. In two other studies, pathological alterations of the dental follicle were found respectively in 42% and 34% of 96 and 100 unerupted third molar teeth removed without clinical sign of any follicular lesions (Glosser and Campbell, 1999; Adelsperger et al., 2000). The dentigerous cyst was the only pathological change found in any specimen by Glosser and Campbell (1999), whereas Adelsperger et al. (2000) found in 34% of the specimens examined, squamous metaplasia indistinguishable from the histological changes found in a dentigerous cyst.

However, a paper published by Kim and Ellis (1993) puts the high incidence of pathologic changes of dental follicles found in the literature into perspective. These two oral pathologists reviewed 847 dental follicles which were removed between 1970 and 1988 and which were examined initially by general and non-specialist pathologists. Over 71% of these specimens were associated with impacted third molar teeth. Interestingly, only 53.4% of the specimens were correctly identified by the contributing medical pathologists. In an additional 16.9% of specimens, only a histological description or a differential diagnosis was offered. No histological diagnosis was made in 9.8% of the specimens. Twenty percent of the specimens were given inappropriate diagnoses. The most common incorrect diagnosis was odontogenic cyst (17%), followed in descending order by odontogenic myxoma, odontogenic fibroma, ameloblastic fibroma, odontoma and ameloblastoma. While diagnostic errors such as the misinterpretation of dental follicular tissue as a dentigerous cyst are generally regarded to be of minor importance with regard to surgical management and overall prognosis, other errors, including the misinterpretation of the tissues from dental follicles, papillae or dentigerous cysts as neoplasms, are more serious and of greater consequence. Kim and Ellis (1993) identified 96 cases (11.3% of the total examined) in which the initial misdiagnosis comprised various odontogenic tumours. These included ameloblastoma, myxoma and calcifying epithelial odontogenic tumour. The investigators suggest that the misdiagnoses are presumably attributable to non-specialist pathologists' lack of familiarity both with the histomorphologic variations of normality and abnormality in tissues from the oral cavity and jaws and with pathologic processes that develop from them.

CONCLUSION

The case presented here and the high incidence of pathological changes in the dental follicles of

unerupted third molars without clinical evidence of any follicular lesion found in the literature, automatically raises the question as to whether routine histological examination of all removed dental follicles should be carried out. It is the authors' opinion that further prospective studies by experienced oral pathologists are needed to establish the true incidence of pathological changes of dental follicles of unerupted third molars. Until then, histological examination of all removed dental follicles cannot be recommended unless there are any macroscopic signs of follicular disease.

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