Unicystic Ameloblastoma Presenting in Gardner's Syndrome

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Sir, a 15-year old female patient was recently referred to the Department of Oral and Maxillofacial Surgery, Sunderland Royal Hospital, for investigation of an unerupted 31 (using the Universal Numbering System; 47 using FDI notation). The patient had recently been diagnosed with Gardner's syndrome. A DPT radiograph revealed a well-defined, unilocular radiolucency centred on the crown of an unerupted 31. Tooth 31 was displaced towards the lower border of the mandible (Fig. 1a). A posterior-anterior (PA) jaws radiograph showed minimal bucco-lingual expansion (Fig. 1b). The clinical and radiological features were consistent with a dentigerous cyst. The cyst was enucleated and the 31 surgically removed under general anaesthetic. Histopathological examination demonstrated a unilocular fibrous walled cyst lined by cuboidal and columnar epithelium. In places, the basal epithelial cells were palisaded and showed reversed nuclear polarity, similar to ameloblasts (Fig. 2). The epithelial cells showed strong expression of CD56 by immunohistochemistry, suggesting pre-ameloblastic differentiation [1]. The features were consistent with sampling of an ameloblastoma. The young age of the patient, the distinct clinical presentation (mimicking a dentigerous cyst)

and the surgical removal of a simple cyst were consistent with unicystic ameloblastoma. We were interested to read a case report by Patel and Rees [2] that documented an association between unicystic ameloblastoma and Gardner's syndrome. At the time, the authors conceded that the occurrence of the two conditions may have been coincidental. Gardner's syndrome has a well-recognised association with dental anomalies, such as odontomes and unerupted teeth, which account for an increased incidence of dentigerous cysts in these patients [3]. The histopathological differences between dentigerous cyst and unicystic ameloblastoma can be subtle and supervening infection and inflammation can also hamper accurate diagnosis. It is therefore conceivable that unicystic ameloblastoma is under-diagnosed in Gardner's syndrome. Furthermore, unicystic ameloblastoma is usually adequately treated by enucleation, similar to a dentigerous cyst; consequently, cases are unlikely to be revisited by the clinicians or the pathologist. This raises the possibility that there are other, as yet undocumented, cases of unicystic ameloblastoma in Gardner's syndrome. We would be interested to know whether other readers have encountered this association.

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Fig. 1 **a** DPT showing the unerupted 31 and associated unilocular radiolucent lesion. **b** PA jaws shows minimal bucco-lingual expansion around the 31

Fig. 2 Haematoxylin and eosin section through the surgical specimen ($\times 100$ original magnification)

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