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Case Report

Recurrent dentinogenic ghost cell tumour of mandible: A case report and literature review

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ABSTRACT

Dentinogenic ghost cell tumours (DGCTs) are uncommon neoplasms classified as solid variants of calcifying odontogenic cyst and are defined as a locally invasive neoplasm, characterized by ameloblastoma-like islands of aberrant keratinization of odontogenic epithelium in the form of ghost cells in association with dysplastic dentine. We report an interesting case of recurrent DGCT of the mandible in a 52-year-old male which was previously misdiagnosed as adenoid cystic carcinoma (ACC) and adenomatoid odontogenic tumour (AOT).

1. Introduction

Dentinogenic ghost cell tumour (DGCT) is a rare odontogenic tumour with a locally invasive behaviour, posing significant diagnostic and therapeutic challenges, which was initially described as a solid variant of calcifying odontogenic cyst. DGCT usually affects elderly people and represents 1.9–2.1 percent of all odontogenic tumours [1]. DGCT is characterized by a peculiar histology composed of ameloblastoma-like islands of epithelial cells in a fibrous connective tissue stroma with variable amount of keratin or dentinoid material. Aberrant keratinization may be found in the form of ghost cells in association with varying amounts of dysplastic dentine [2]. We report an interesting case of recurrent DGCT of the mandible in a 52-year-old male which was previously misdiagnosed as adenoid cystic carcinoma (ACC) and adenomatoid odontogenic tumour (AOT).

2. Case report

A 52-year-old male reported to our department with chief complaint of slow growing painful swelling in the reconstructed left mandibular posterior region. On extra-oral examination a slow growing hard, lobulated swelling involving the left submental and submandibular region which was present for the past 1 year was revealed (Fig. 1A). Intra-oral examination revealed a diffuse swelling extending from the mandibular left second premolar region up to the left retromolar trigone area was seen (Fig. 1B). Orthopantomogram showed radiolucent lesion in left body region of mandible extending from parasymphyseal region till left angle of mandible and radio-opaque reconstruction plate (Fig. 1C). Computerized tomography scan revealed collection of homogenously enhancing soft tissue in the left submandibular region measuring 2 cm × 1.5 cm representing granulation tissue (Fig. 1D).

The patient gave past history of similar type of hard, painless swelling in the left mandibular posterior region 5 years ago (Fig. 2A). At that time the patient had an ill-defined hard mass in the left mandibular body involving the left submandibular and submental space without any surface ulceration. Intra-oral

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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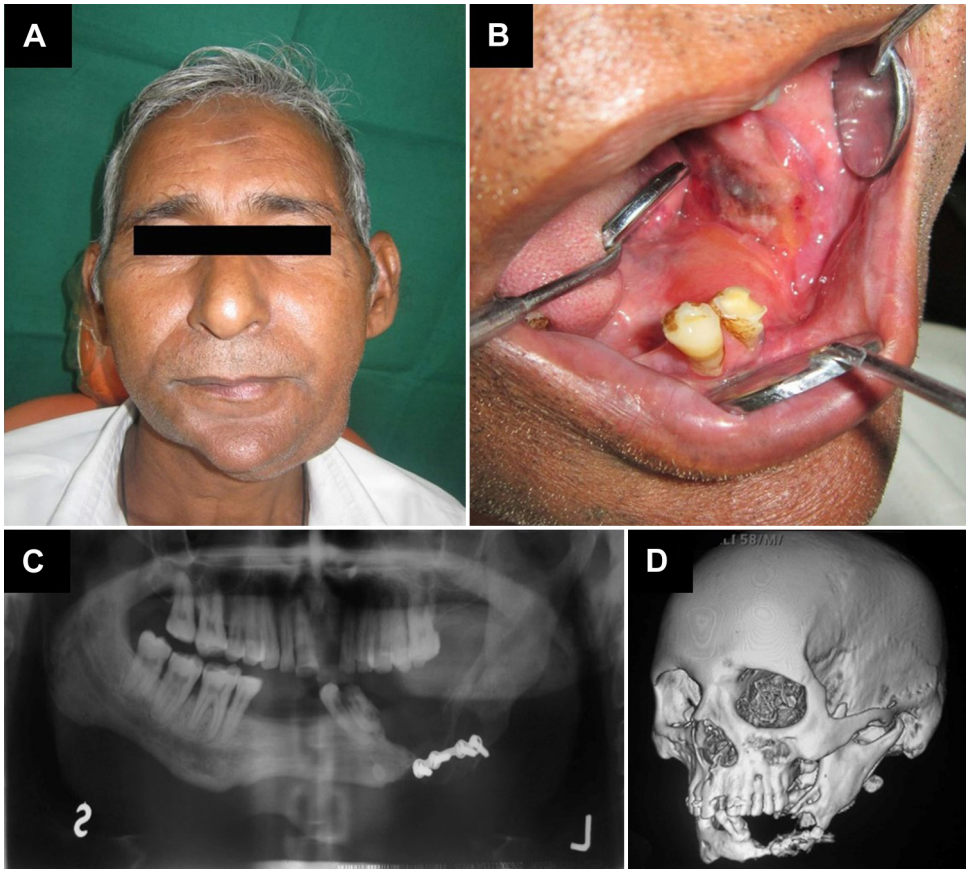

Fig. 1. Clinicopathological features of recurrent tumour. (A) Extra-oral lobulated swelling involving the left submental and submandibular region; (B) intra-oral swelling extending from the mandibular left second premolar region up to the left retromolar trigone area; (C) orthopantomogram showed radiolucent lesion in left body region of mandible extending from parasymphyseal region till left angle of mandible and radio-opaque reconstruction plate; (D) CT scan revealed collection of homogenously enhancing soft tissue in the left submandibular region measuring 2 cm × 1.5 cm.

examination had reported a hard, non-tender swelling extending from left mandibular first premolar to left retromolar trigone with bicortical expansion (Fig. 2B). Previous radiographs revealed a well-circumscribed radiolucent lesion extending from apical area of the left mandibular canine to the mesial of the left mandibular first molar (Fig. 2C). On incisional biopsy, Haematoxylin & Eosin stained sections showed lesional tissue composed of nests of uniform basaloid cells. Fibrous septa divided the lesional tissue, giving it a lobular pattern. Therefore it was reported as adenoid cystic carcinoma (ACC) (Fig. 2D). Hence the tumour was excised via left segmental mandibulectomy and the defect was reconstructed with iliac osteocutaneous flap. Grossly resected specimen was a solid mass, brownish black in colour, measuring 3.5 cm × 3 cm × 2 cm (Fig. 2E). To our surprise it showed features diagnostic of adenomatoid odontogenic tumour (AOT) instead of ACC (Fig. 2F). Histopathological examination revealed densely arranged cuboidal to spindle-shaped odontogenic epithelial cells in variable ductal, rosettes and convoluted pattern. Scattered small globular to irregular calcifications were seen in the proliferative tissue. On re-evaluating his primary tumour histopathological sections, some areas of ghost cells and dentinoid-like material were present in close proximity to odontogenic islands having peripheral ameloblast-like cells, suggestive of dentinogenic ghost cell tumour which perhaps went unnoticed previously.

As mentioned earlier the patient reported back with recurrence in the same region for which an incisional biopsy was performed under local anaesthesia. H&E section showed solid tumour composed of proliferating odontogenic islands with peripheral palisading columnar cells resembling ameloblast-like cells, dentinoid tissue and numerous dentinoid tissue, and of some dystrophic calcification (Fig. 3A–C). The ghost cells were seen associated with proliferating odontogenic epithelial islands and also as clusters in dense connective tissue. The dentinoid areas were seen in close proximity to the odontogenic epithelial islands and ghost cells, as well as in isolated areas of the dense connective tissue stroma. The connective tissue stroma was composed of moderately dense collagen fibres. Histopathological diagnosis of DGCT was given. Subsequently, segmental mandibulectomy of previously reconstructed mandible with iliac graft was performed. Macroscopically, it was 7 cm × 5 cm × 2 cm, irregular in shape, brown in colour and firm in consistency (Fig. 3D). Histopathological features of excised tissue were consistent with that of incisional diagnosis of intraosseous DGCT. No dysplastic features were noted. Postoperative healing was uneventful. The patient is well without any recurrence after 3-year postoperative follow-up.

3. Discussion

Calcifying odontogenic tumours contain cystic, neoplastic elements in different proportions which are termed as dentinogenic ghost cell tumour, odontogenic ghost cell tumour, dentinoameloblastoma, calcifying ghost cell odontogenic tumour or epithelial odontogenic ghost cell tumour [2–4]. Calcifying odontogenic cyst (COC) is a well-known entity to clinicians and pathologists. COC was first described as a separate entity by Gorlin [5] in 1962 and by Gold [6] in 1963, as a benign odontogenic cyst. Two organizing principles of classification of COCs have been put forward: monistic and dualistic [7]. The monistic concept, best

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