Benign Odontogenic Tumours

28

Anjan Kumar Shah

28.1 Introduction

The region of Head and Neck has a wide range of pathological disorders due to the complex nature of the tissues in this region. The tooth-forming tissues can give rise to a wide array of tumours, both benign and malignant. They vary in size from tiny swelling to a large variety, causing cortical bone perforation with the displacement of the adjoining normal anatomic structures. Odontogenic tumours are slow growing and generally non-aggressive, with aggressive behaviour shown by certain tumours.

It is important for the clinician to have a thorough knowledge of the pathology, clinical as well as radiological presentation in order to manage these conditions.

According to W.H.O [1]
'Odontogenic tumours and tumour-like lesions constitute a group of heterogeneous diseases that range from hamartomatous or non-neoplastic tissue proliferations to benign neoplasms and finally malignant tumours with metastatic potential. They are derived from epithelial, ectomesenchymal and/or mesenchymal elements of the tooth-forming apparatus. Odontogenic tumours are rare, some being extremely rare, but can pose a significant diagnostic and therapeutic challenge.'

The WHO first classified benign odontogenic tumours in 1971 followed by in 1992, 2005 and the recent classification was in 2017. The origin-based sub-classification was first defined in 1992, which is still in use, i.e. the tumours are sub-classified into Epithelial origin, Mixed origin and Mesenchymal origin. The metastasising (malignant) ameloblastoma is included in epithelial origin tumour in 2017 WHO classification and desmoplastic ameloblastoma was excluded from the classification of benign odontogenic tumour. In 1992 classification, adenomatoid odontogenic tumour (AOT) was considered under mixed origin but in 2005 and 2017 classification, it is included in the epithelial origin tumour [2].

The dentinogenic ghost cell tumour (DGCT) comes under the spectrum of ghost cell lesions. Gorlin et al. in 1962 first described calcifying odontogenic cyst (COC) as the earliest ghost cell lesions. In 2005 WHO classification, COC is renamed as calcifying cystic odontogenic tumour (CCOT). Fejerskov and Krogh used the term calcifying ghost cell odontogenic tumour for DGCT in 1972. Dentigerous ghost cell tumour term was given by Praetorious et al. in 1981, which is still retained in 2005 and 2017 WHO classification. In between, Shear in 1983 used the term dentinoameloblastoma, whereas Ellis and Shmookler proposed epithelial odontogenic ghost cell tumour. Hong et al. in 1991 suggests the term epithelial odontogenic ghost cell tumour [3, 4].

28.2 WHO (2017) Classification of Odontogenic Tumours [5]

28.2.1 Benign Odontogenic Tumours

28.2.1.1 Epithelial Origin

- Ameloblastoma, conventional
  Ameloblastoma, unicystic type
  Ameloblastoma, extraosseous/peripheral type
  Metastasising (Malignant) ameloblastoma
- Squamous odontogenic tumour
- Calcifying epithelial odontogenic tumour
- Adenomatoid odontogenic tumour

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28.2.1.2 Mixed (Epithelial-Mesenchymal) Origin

- Ameloblastic Fibroma
- Primordial odontogenic tumour
- Odontoma, complex type
- Odontoma compound type
- Dentinogenic ghost cell tumour

28.2.1.3 Mesenchymal Origin

- Odontogenic Fibroma
- Odontogenic myxoma/myxofibroma
- Cementoblastoma

28.2.2 Malignant Odontogenic Tumours

- Ameloblastic carcinoma
- Primary intra-osseous carcinoma, NOS
- Sclerosing odontogenic carcinoma
- Clear cell odontogenic carcinoma
- Ghost cell odontogenic carcinoma
- Odontogenic carcinosarcoma
- Odontogenic sarcomas

28.3 Ameloblastoma

28.3.1 Introduction

It is the most common odontogenic tumour arising from odontogenic epithelium.

Ameloblastoma originates from sources, which include [6]

- Epithelial cell rests of Malessez, which are the residual epithelium of the tooth-forming apparatus.
- Epithelium of the enamel origin and of odontogenic cysts
- Surface epithelium of the basal cells
- Heterotopic epithelium from the extra-oral sites such as pituitary gland

Edited 2017 WHO classification of odontogenic tumours has simplified the classification of Ameloblastomas. It has removed the various pathologically descriptive classification terminologies such as follicular, plexiform, basaloid, granular or desmoplastic as these terminologies do not have any relevance to clinical behaviour. Conventional-type Ameloblastomas are characterised by multilocular, expanding behaviour. The histological margin is characterised by the infiltrating margin, requires wide excision with a margin of 0.5–1 cm of normal bone, or one anatomical layer if grown out of the confines of the bone. That is if bony cortex invaded, then take muscle or subcutaneous tissue to maintain periosteum as anatomical barrier.

28.3.2 Definition

'It is a true neoplasm of enamel organ-type tissue, which does not undergo differentiation to the point of enamel formation" proposed by WHO. Robinson [7] defined it as a "Non-functional, unicentric, intermittent in growth, anatomically benign and clinically persistent type of tumour'

28.3.3 Incidence

It represents 19.3–41.5 % of all odontogenic tumours [8, 9]. Posterior Mandible is the commonest site of occurrence in almost 80% of ameloblastomas [10]. It occurs over a broad range; cases ranging from adults older than 90 to the children younger than 10 years. Most frequently, they occur in the second and fourth decade of life. Some authors found no gender predilection [11] while many studies showed female predilection [12] between the first and the third decade of life [13].

28.3.4 Clinical Features

1. Swelling of maxilla or mandible
2. Facial disfigurement
3. Tooth displacement and mobility
4. Paraesthesia
5. Ulceration or Nasal obstruction

28.3.5 Radiological Features

- Ameloblastomas commonly originate within the bone, thus often they are detected on routine dental x-rays or on orthopantomogram (OPG).
- Root resorption of the involved teeth
- The solid/multilocular-type ameloblastomas, which is most common, classically shows "soap bubble appearance" [14]
- Computed tomography (CT) is very helpful in surgical planning by giving exact cortical destruction and soft tissue extension, it typically demonstrates well-defined radiolucent uni/multilocular lesions [15]
- Magnetic resonance imaging (MRI) is very helpful in maxillary lesions for defining the extension of ameloblastomas into paranasal sinuses, orbit and skull base [16]
- PET–CT generally indicated in patients with metastatic ameloblastomas.

28.4 Solid/Multicystic Ameloblastoma

In 2005 WHO classification, the Ameloblastoma, solid/multicystic type, was mentioned, which is replaced by Ameloblastoma alone in 2017 classification again. Historically, the Ameloblastoma is classified into Unicystic and Multilocular or Solid.

The solid/multicystic or intra-osseous ameloblastoma is locally invasive, slow-growing and odontogenic tumour of epithelial origin with a high rate of recurrence if not removed or treated properly. It has no tendency to metastasise. It invades the bone marrow spaces.

It has no gender predilection and occurs equally in both sexes. Most commonly diagnosed between 30 and 60 years of age. The posterior mandible region affected in more than eighty percent of the tumour cases. It may present as variably sized swelling of jaws. Pain and paraesthesia are rare. It usually appears multilocular (soap bubble appearance) in radiographs.

It occasionally is associated with an impacted tooth and causes expansion of the bony cortex, with the possibility of resorption of roots of the involved teeth [17, 18]. The most common histological patterns found are plexiform and follicular types. The others are desmoplastic, acanthomatous, granular and basal [2]. To confirm the diagnosis, the combination of imaging (plain as well as computed tomography) and biopsy can be performed.

The tumour infiltrates through the medullary spaces and might erode the cortical bone. After resorbing the cortical bone, it may extend into the adjacent tissues. The maxillary tumours of the posterior region tend to obliterate the maxillary sinus and may extend to infiltrate the skull base.

28.4.1 The Treatment Goals

- Complete removal of tumour
- Aesthetic reconstruction with minimal disfigurement
- Good prognosis
- Long-term follow-up

The treatment depends on the best judgement of the surgeons and individual status of the patient. The surgical planning should be based on the lesions present in the mandible or maxilla. The maxilla has got higher percentage of cancellous bone, which facilitates the spread of tumour in comparison to the mandible having thick and dense cortical plates, which limit the spread of neoplasm.

The treatment of the conventional/multicystic variant is classified into radical and conservative. The conservative methods include

- Curettage
- Enucleation
- Peripheral ostectomy with surgical excision
- Surgical excision with other adjuvant therapy such as cryotherapy or use of carnoy's solution
- Liquid nitrogen therapy [19]
- Marsupialisation

Radical treatment includes resection of bone. At least 1 cm of the surrounding healthy tissues should be removed along with the tumour in cases of cortical bone perforation because if tumour cells are left behind, they may give rise to locoregional recurrence even several years after resection [20]. There is a 1.26-fold increase in the chance of the recurrence rate with the increase in the size of every 10 mm of tumour [21].

In mandible, resection can be carried out on the basis of the extension of the lesion in the form of alveolectomy, marginal mandibulectomy, segmental resection, hemimandibulectomy or hemimandibulectomy with disarticulation depending on the extension of the tumour. Based on the extension of tumour, maxillectomy (partial, total or subtotal) has been performed in radical treatment of maxillary lesions [22].

Enucleation and curettage are inadequate because the tumour invades the adjoining cancellous bone. However, some conditions in which it is carried out are given below:

28.4.1.1 Indications of Enucleation and Curettage

1. In medically compromised patient or conditions in which patient is unfit for general anaesthesia.
2. Very young or old patient who is not willing for segmental resection.

If enucleation and curettage has to be done, it is preferable to be carried out in association with chemical cauterisation with the help of modified carnoy' solution and peripheral ostectomy.

The surgical treatment of solid/multicystic variant is controversial. A high rate of recurrence is reported if it is not adequately excised or resected. According to some authors, the initial treatment should be conservative as the tumour has low metastatic potential and the radical treatment should be done in cases of recurrence. While others believe that when-

ever possible complete removal of the tumour with preservation of lower border of the mandible will be the treatment of choice [23].

Conservative treatment has a recurrence rate ranging from 33 to 90%, as compared to the rate of recurrence by radical treatment, i.e. 7–25% in the literature. However, the patients experience serious functional and aesthetic impairments with radical treatment [24, 25]. Hasegawa et al. [26] also reported the recurrence rate of 43.5% following conservative management.

Enucleation alone showed the highest rate of recurrence amongst all the modalities of conservative management [27]. According to Esquillo ME [28], small multicystic and solid lesions can be treated by marsupialisation with good results. This will maintain good facial aesthetics of the patient, lessening the treatment cost but patients have to keep on long-term follow-up.

About 40% of intra-osseous Ameloblastomas did not recur after conservative management, which led to the conclusion that the small intra-osseous ameloblastomas can be treated with conservative management initially, leading to fewer post-operative complications and if the lesion recurs, the radical treatment can be carried out in the second-stage surgery aggressively [29].

Almeida AC et al. [30] in the systematic review and meta-analysis concluded that the bone resection should be the treatment of choice for primary multicystic ameloblastoma, also the chances of recurrence were 3.15 fold more when the conservative treatment was performed in comparison to radical treatment. Hendra FN et al. also suggest that the rate of recurrence was less when radical treatment was opted for the treatment of intra-osseous ameloblastoma in their systematic review and meta-analysis [31].

Antonoglou GN and Sandor GK [32] in the systematic review and meta-analysis concluded that no strong recommendations have been made regarding the treatment options regarding the intra-osseous ameloblastoma. However, radical treatment in the form of resection is the treatment of choice for solid/multicystic ameloblastoma.

Pogrel MA and Montes DM [33] concluded that Enucleation alone should not be the choice of treatment for multicystic or solid lesions. In the case of maxilla, partial maxillectomy and in mandibular lesions, segmental resection with 1 cm margin will be preferred to avoid recurrence.

Sampson DE and Pogrel MA [34] suggested the management algorithm for management of mandibular ameloblastoma, which is, in the case of mandibular ameloblastoma, if lesion is less than 1 cm, in plain radiograph, then curettage and cryotherapy is the treatment of choice and patient is to be kept on long-term clinical and radiological follow-up. If the lesion is more than 1 cm, CT scan is to be done. If findings are positive, then segmental resection with involved soft tissue is carried out with suitable reconstruction and patient is to be kept on long-term clinical and radiological follow-up.

Sammartino et al. advocated the following treatment plan in Mandibular tumour management [24]. In the cases of small lesions, box resection has to be done and the patient has to keep for long-term follow-up to 10 years. For large lesions if cortical perforation is there on CT examination. Then segmental or marginal resection has been carried out along with excision of overlying soft tissues, if cortical perforation is absent then curettage is the choice of treatment with 0.5–1 cm of clinically uninvolved surrounding bone. In both cases, 10-year followup of patients is mandatory. If no recurrence occurs, orthopantomogram has to be done in every 2–3 years on the other hand if recurrence occurs, then the first or second small recurrence can be treated with marginal resection while for the third recurrence, the segmental resection is the choice of treatment.

28.4.2 Surgical Management of Ameloblastoma According to the Anatomic Locations

28.4.2.1 Mandibular Anterior Region (Canine to Canine)

According to Gardner, anterior mandible should be approached conservatively because no important major anatomic structures are present in the anterior mandible. However, if curettage is used as a choice of treatment, recurrence is anticipated and it is preferred in smaller lesions. Always an attempt should be made to preserve the inferior border of mandibular in the anterior region because the tumour infiltration is less, due to the thick cortical bone of the symphyseal region. In large lesions with cortical perforation, marginal mandibulectomy will be the choice of treatment followed by long-term follow-up (Fig. 28.1). After marginal mandibulectomy, the lower border of mandible is reinforced with a reconstruction plate, to avoid pathological fracture in future (Videos 28.1 and 28.2).

28.4.2.2 Posterior Mandible (Bicuspid to Condyle)

Marginal mandibulectomy should be the choice of surgical treatment for the posterior mandible and body region while maintaining the inferior and posterior border whenever possible for solid/multilocular variant. Maxillomandibular fixation might be required after marginal resection without

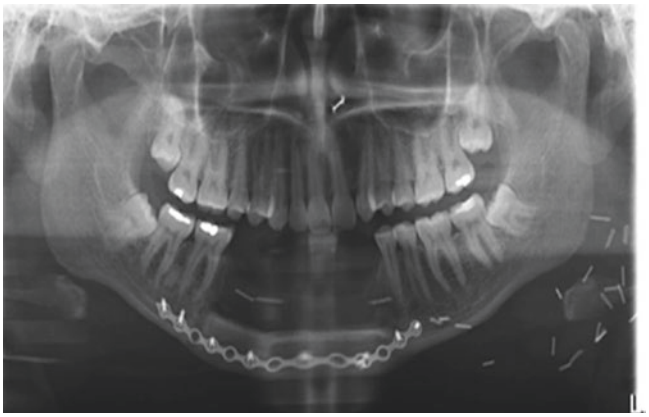

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Fig. 28.1 Orthopantomogram representing marginal mandibulectomy, lower border protected with reconstruction plate

continuity defects to avoid the chances of pathological fracture. A reconstruction plate may be contoured before resection, in order to maintain the normal anatomic relationship between proximal and distal segments by marking the points of screw fixation on the normal bone. In recent years, mandibular reconstruction using intra-oral microvascular anastomosis following segmental resection in cases of ameloblastomas has been carried out with great success. Most commonly, free fibular graft has been used for the same, followed by dental rehabilitation with the help of dental implants [35, 36].

28.4.2.3 Anterior Maxilla (Canine to Canine)

Goodcell JF reported 2% ameloblastomas in anterior maxilla [37]. However, Sehdev MK et al. show 9% of ameloblastomas occurs in the canine and incisor region [38]. Less-aggressive treatment has been advocated in anterior maxilla when compared to posterior, because a sufficient distance from the vital structures allows being less radical. Radical treatment in the form of partial or total maxillectomy may result in significant deformity.

28.4.2.4 Posterior Maxilla (Bicuspid to Pterygoid Plates)

Ameloblastoma occurs 47% in the molar region, 15% in the maxillary antrum and floor of the nose, while 9% in the premolar region.

Lack of cortical bone in maxilla makes it much more dangerous than mandibular ameloblastomas. The definitive treatment becomes difficult in the posterior maxillary region because tumours are not well confined by the thin maxillary cortical bone and easily spread beyond the maxillary bone boundaries. Early detection is also very difficult. Posterior maxillary tumours are rarely treated by conservative management. Extra-oral or intra-oral resection of the tumour is carried out, sometimes Le fort I down fracture is required to access the tumours of maxillary sinus or the tumour invades the posterolateral wall of the maxillary sinus.

Weber Fergusson incision and mandibulotomy can be used for accessing the tumors of maxilla, pterygoid and infra temporal fossa. When reconstruction of the defect after maxillectomy has been planned by temporalis muscle, Rai A et al. [39] advocated use of Borle's extension weber fergusson incision.

Various surgical options in management of maxilla/mandible tumours.

Benign Mandible Tumours

Type or surgery depends on number of factors

1. Type of pathology—Benign/Benign aggressive
2. Extent of involvement—e.g. Sufficient lower border of mandible present or not
3. Site of tumour—Anterior mandible/Posterior mandible

Based on this options starting from the most conservative to most radical are:

1. Marsupialisation
2. Enucleation or curettage
3. Peripheral ostectomy or En bloc resection
4. Segmental resection

Access incisions:

1. Intraoral transmucosal access
2. Combined Intraoral and extraoral submandibular access
3. Combined Intraoral and Visor incision

Repair:

1. Primary mucosal closure
2. Packing cavity and healing by secondary intention
3. Reconstruction plate with primary mucosal closure
4. Reconstruction plate with free bone graft
5. Vascularised Bone free flap

Benign Maxillary Tumours

Type or surgery depends on number of factors

1. Type of pathology—Benign/Benign aggressive
2. Extent of involvement—e.g. Involvement of sinus/pterygoids/infratemporal fossa
3. Site of tumour—Anterior maxilla/Posterior maxilla

Based on this the following excision options are possible:

1. Marsupialization into Oral cavity/Maxillary Sinus
2. Enucleation/Curretage
3. Enbloc resection—Low level/High level maxillectomy

Access incisions:

1. Transoral accesss
2. Weber Ferguson incision—Good for anterior tumours
3. Mandibulotomy access approach—This provides excellent access for posterior tumours especially when access to the pterygoids/intratemporal region is required

Repair:

1. Primary closure
2. Packing cavity to allow healing by secondary intention with a Healing plate
3. Obturator
4. Local flap—e.g. Temporalis
5. Free flap reconstruction

28.5 Reconstructive Modalities After Surgical Resection of Ameloblastoma

Need for Reconstruction:

1. For restoration of movements and equilibrium of mandible
2. For maintenance of normal occlusal plane, floor of the mouth and tongue's anatomical position
3. For restoration of near normal feeding
4. For acceptable aesthetics and function.
5. For more favorable social acceptance
6. To establish the arch form, width and alveolar height.
7. To establish the bone continuity and maintain facial contours

28.5.1 Timing of Reconstruction

Immediate reconstruction is usually performed with the help of microvascular-free flaps, harvested from fibula, scapula, iliac crest and ribs. For the reconstruction of the mandible, free fibula flap is the treatment of choice. It is superior to iliac crest graft. Scapula flap for maxillary reconstruction may be a good alternative because of its long pedicle and good bone quality. For soft tissue reconstruction, the radial forearm-free flap is the choice of treatment. Most of the surgeons still prefer to use Titanium reconstruction plate for reconstruction when free flaps are not possible as an immediate reconstruction modality. Delayed reconstruction (second stage) can be performed for reconstruction with the help of a titanium reconstruction plate. The immediate reconstruction significantly improves the patients' health-related quality of life, many patients prefer immediate reconstruction [40].

Lawson et al. [41] reported 90% success rate with delayed reconstruction in comparison to immediate, which is 46% using non-vascularised bone grafts.

Autogenous bone graft selection (Vascularised free flaps vs non-vascularised bone graft) depends on following factors

1. Experience of the surgeon
2. Contour and size of the defect
3. Size and quality of the soft tissue available

The HCL classification [42, 43] is used as an aid in classifying mandibular defects. The defect from canine to canine, i.e. the central defect is designated as 'C'. The lateral segment defect excluding condyle designated as 'L' and when condyle is included in resection with lateral mandible the defect is designated 'H' (Fig. 28.2). The importance of this classification indicates that reconstruction of lateral defect can be done by straight bone segment while defect located centrally required osteotomies.

28.5.1.1 Case Scenario 1 (Fig. 28.3a–g)

A 38 year old male patient reported to our outpatient department with the chief complaint of painless swelling of lower jaw since last 6 months (Fig. 28.3a). History of present illness represents swelling initially was of lemon size and increasing gradually to reach the present size, extending from right side of the body region of the mandible to the ramus region of the opposite side. On intra-oral examination, a diffuse swelling was present from one side of the molar region extending to the other side of the molar region of the mandible (Fig. 28.3b).

The orthopantomogram (Fig. 28.3c) and the CT scan (Fig. 28.3d) showing the multilocular lesion extending from the right side of the first molar to the ramus region of the contralateral side. The incisional biopsy of the lesion confirmed the diagnosis of solid/multicystic ameloblastoma. Under general anaesthesia, the resection of the lesion was done, the complete resection of lesion was confirmed in the specimen radiography (Fig. 28.3e). The reconstruction of the

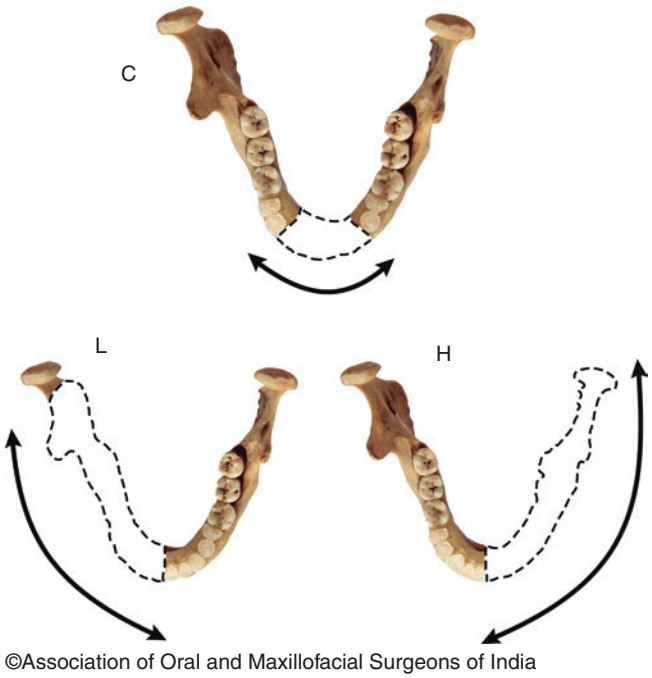

Fig. 28.2 The HCL Classification

defect was done with the help of microvascular free fibula flap and with the titanium reconstruction plate (Fig. 28.3f), post-operative OPG representing reconstruction (Fig. 28.3g).

28.5.2 Bone Graft Substitutes

The calcium phosphate cement (Hydro set) is the commonly used material, which converts in situ to hydroxyapatite, serving as an effective osteointegrative and osteoconductive material [44]. The other implantable material options include high-density porous polyethylene implants [45]. Using rapid prototype models, the custom-made implants are commonly used for the reconstruction of maxillofacial defects with good results [46].

The titanium mesh tray filled with autologous cancellous bone blocks fixed with the residual segment of bone and also titanium mesh cage filled with fresh bone marrow, recombinant human bone morphogenic protein (BMP) and xenogenic bone mineral are used for reconstruction of mandibular defect. The BMP is the key activator of bone induction [42].

Recent advances in mandibular reconstruction include transport disc distraction osteogenesis, modular endoprosthesis and tissue engineering. Dental implants are commonly used in autogenous bone grafts for the rehabilitation of masticatory functions [42].

28.6 Unicystic-Type Ameloblastoma

Robinson and Martinez [47] first described Unicystic Ameloblastoma (UA) in 1977.

In 1988, Ackermann GL et al. [48] reclassified UA with prognostic and therapeutic implications into three types

Type 1: Unilocular cystic lesions lined by epithelium exhibiting features of ameloblastoma.
Type 2: Epithelial nodules arising from cystic lining and projecting into the cyst lumen.
Type 3: The presence of invasive islands of ameloblastomatous epithelium in the connective tissue wall of the cyst and these islands may or may not be connected to the cyst lining.

28.6.1 Clinical and Radiographic Features

Most of the UA clinically and radiographically resemble denigerous cysts in behaviour. Embedded teeth are associated with some UA and hence resembles residual or primordial cysts.

UA most commonly occurs in the second and third decade of life and have predilection to mandible [48]. UA many times are associated with mandibular third molars. There will be well-corticated unilocular and often pericoronal radiolucency

In some cases, root resorption can occur.

28.6.1.1 Case Scenario 2 (Fig. 28.4a, b)

A 14 year old female reported with swelling on the left side of the ramus region for the past 3 weeks. OPG (Fig. 28.4a) and PA view (Fig. 28.4b) showed a large unilocular radiolucent lesion involving the whole of the ramus of the left mandible. Tooth bud of lower left third molar was absent. The lesion seemed to arise from the impacted tooth bud of lower left second molar and the lesion was also involving the roots of lower left first molar. Under the provisional diagnosis of a dentigerous cyst, the lesion was enucleated under anaesthesia and primary closure was done and the impacted tooth bud was removed in this case. The histopathology report was that of an unicystic ameloblastoma with mural changes. The child was on follow-up with no evidence of recurrence.

On histological examination, various situations may be found such as shown in Fig. 28.5

1. The ameloblastomatous epithelial lining
2. An ameloblastoma nodule projects into the lumen (Luminal Ameloblastoma)

3a. Ameloblastoma islands present in the connective tissue wall of an apparently non-neoplastic cyst.
3b. Proliferation of ameloblastoma into the connective tissue wall from cystic lining. (One of the Mural variant)

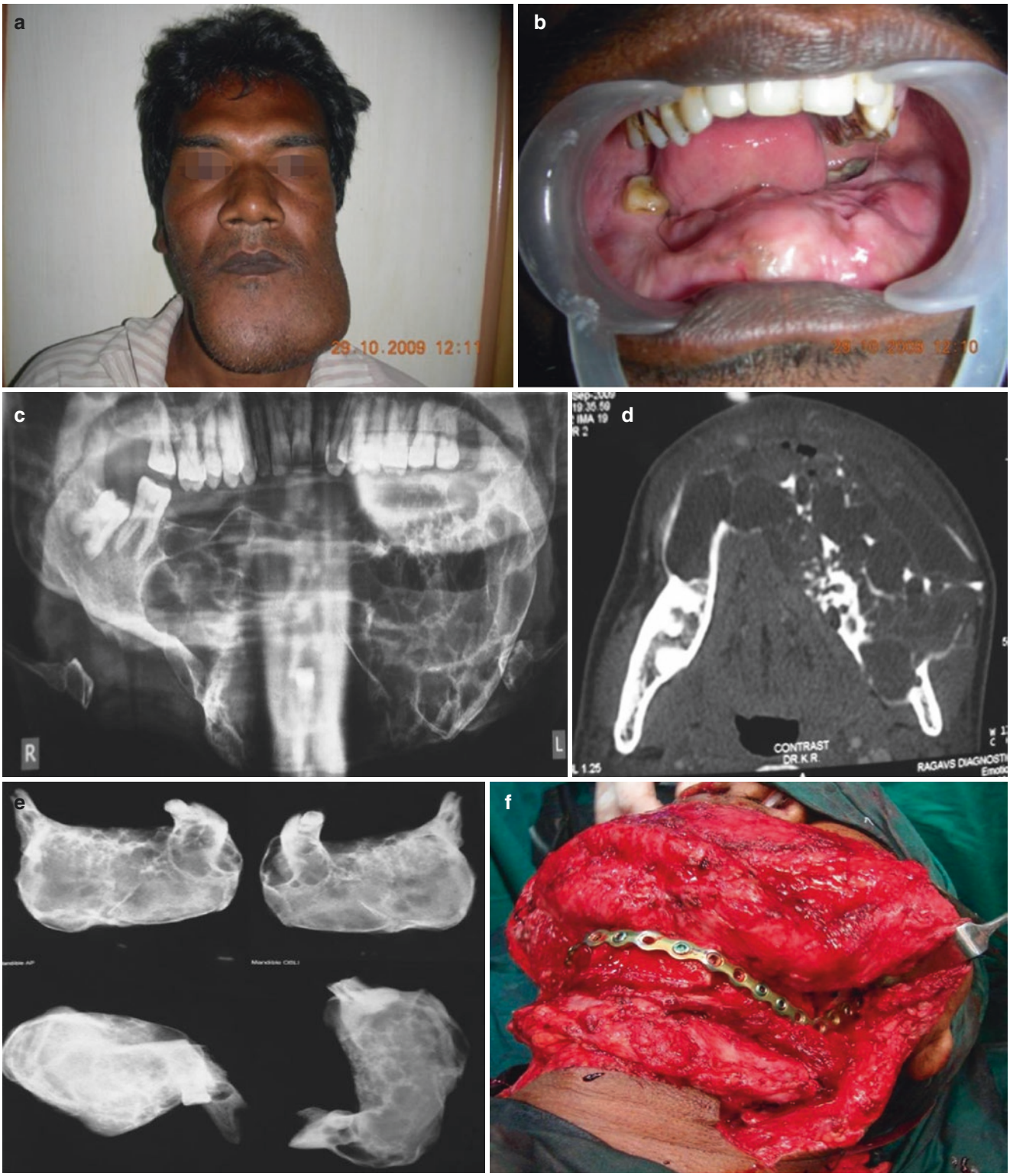

Fig. 28.3 (**a**) Pre-operative extra-oral view. (**b**) Pre-operative intra-oral view. (**c**) Pre-operative OPG. (**d**) Pre-operative CT Scan of the patient. (**e**) Specimen Radiography. (**f**) Clinical picture showing reconstruction with the reconstruction plate. (**g**) Post-operative OPG

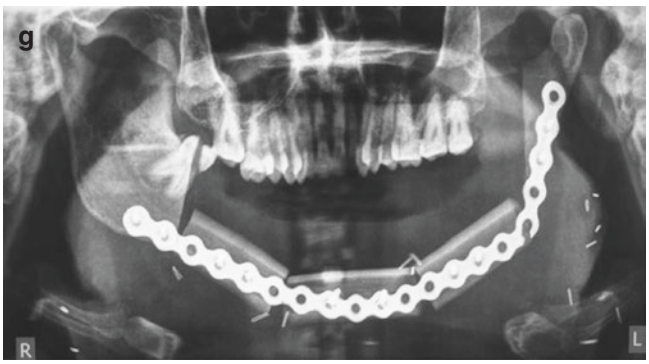

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Fig. 28.3 (continued)

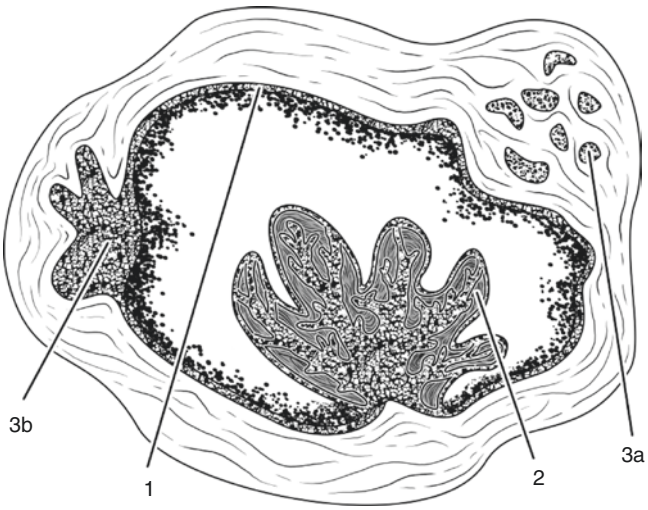

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Fig. 28.5 Various situations of Unicystic Ameloblastoma found on Histological examination. 1. The ameloblastomatous epithelial lining. 2. An ameloblastoma nodule projects into the lumen (Luminal Ameloblastoma). 3a. Ameloblastoma islands present in the connective tissue wall of an apparently non-neoplastic cyst. 3b. Proliferation of ameloblastoma into the connective tissue wall from cystic lining. (One of the Mural variant)

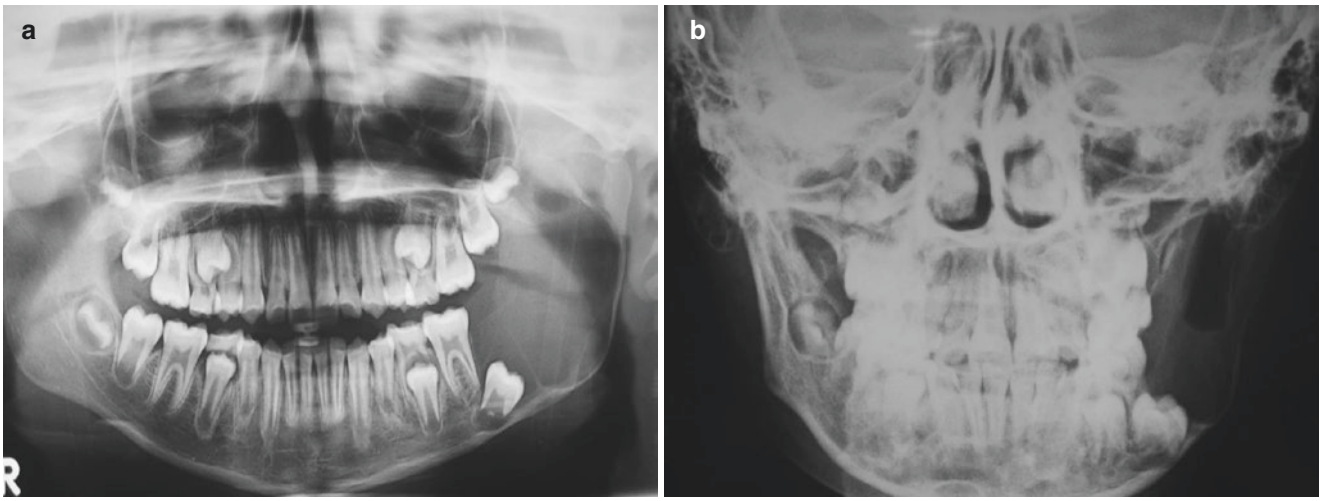

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Fig. 28.4 (**a**, **b**) OPG and PA view showing Unicystic Ameloblastoma

28.6.2 Management

UA treatment is controversial and can be grouped into

- Enucleation
- Marsupialisation or decompression
- Radical resection

Conservative management has been advocated by cakarer et al. in their study on large benign aggressive lesions of the jaws, where they performed decompression followed by curettage if required [49].

Zheng et al. in 2019 published a long-term follow-up study on 116 cases of UA and have concluded that marsupialisation as an effective treatment option for UA. They found the recurrence rate more for the mural sub-types and the predictors for outcomes were resorption of the root, perforation of the cortical bone and histopathological sub-types [50].

Usually, initial treatment for UA is enucleation because they appear clinically as cysts, and the histopathology type is known only in the excision biopsy report as mural changes can be picked by the pathologist only when the full specimen is available. For histologic type showing luminal changes, the treatment of choice is Enucleation, but has to be followed up for 5–10 years. In types where the ameloblastoma infiltrates the adjacent cancellous bone, marginal resection is the treatment of choice after initial Enucleation and curettage [51]. This means that patient has to undergo two surgeries based on the histopathological examination.

There is some controversy on its management, with some authors advocating aggressive management, especially for the mural variant, while others advocating conservative management such as curettage, peripheral ostectomy and various adjuvant materials such as Carnoy's solution and Liquid Nitrogen [33, 52, 53]. If adequate follow-up is possible, the UA of posterior mandible can be treated conservatively with curettage or peripheral ostectomy.

According to LAU and Samman [54], 30.5% recurrence rate reported with enucleation alone. Application of carnoy's solution along with Enucleation decrease the recurrence rate to 16% and the least percentage of recurrence was seen with resection of tumour, i.e. only 3.6%

28.7 Use of Carnoy's Solution in Ameloblastomas

- Stoelinga and Bronkhorst [55] in 1987 suggested the use of carnoy's solution for UA as a chemical cauterisation. The carnoy's solution consists of chloroform 3 ml, absolute Alcohol 6 ml, 1 ml glacial acetic acid, sclerosing agent ferric chloride 1 gm for the management of fistulae and cysts as a fixative.
- The modified carnoy's solution avoids the use of chloroform due to concerns about its carcinogenicity [56]. However, the recurrence rate is lower with application of conventional carnoy's solution after enucleation and curettage in comparison to modified carnoy's solution [57].
- The use of carnoy's solution reduces the risk of recurrence by fixing the residual tumour tissues after enucleation of UA with mural invasion, but conventional multicystic ameloblastoma and some UA are unlikely to be effective by carnoy's solution. The recurrence experienced more in patients treated with Enucleation alone without the application of carnoy's solution.
- The solution is applied for 5 min with the help of cotton applicator or gauze soaked in carnoy's solution in the bony cavity. Irrigation with normal saline is done after that. The nerve and vessels should be avoided by contact of the carnoy's solution as much as possible. The method and duration of application of carnoy also is contentious where different regimes have been proposed by various authors.

28.7.1 Case Scenario 3 (Fig. 28.6a–i)

Case scenario 3 is provided to show that conservative management with adjunctive procedures will give reasonably good results thereby avoiding major resective and reconstructive surgeries. A 21-year male reported with gradually growing swelling right side body mandible (Fig. 28.6a). OPG (Fig. 28.6b) showed an unilocular expansile lesion in right side body mandible with resoprtion of the associated roots. There was intra-oral vestibular swelling and it was fluctuant on palpation due to thinning of the labial cortex. CT views (Fig. 28.6c–e) show the lesion to be unilocular, expansile with loss of both labial and lingual cortices. An incisional biopsy was performed, which gave the report as Unicystic ameloblastoma. This could be considered as an aggressive form as there was root resorption. Considering the age of the patient, a less-radical approach was taken, which involved excision of the lesion by raising an intra-oral crevicular mucoperiosteal flap (Fig. 28.6f), extraction of all the involved teeth (Fig. 28.6i), performing a peripheral ostectomy and applying carnoy's solution (Fig. 28.6g) and also excising the overlying mucosa, which was in direct contact with the lesion. The excised cystic lesion was quite thick walled and an in toto enucleation was possible (Fig. 28.6h). A reasonable thickness of the lower border of mandible was left behind and as the alveolar part of labial and lingual cortices was removed, excess tissue was available for achieving a tension-free primary closure. The postoperative OPG at one and half years (Fig. 28.6j) shows no evidence of recurrence and prosthetic rehabilitation performed. The patient is recurrence free for past 7 years.

28.7.2 Case Scenario 4 (Fig. 28.7a–f)

This case scenario is given to show that at times complex resection and reconstructive procedures may be avoided in amelobalstomas considering, the patients age and general health and other social background. In such cases where a conservative approach is used, the adjunct measures of peripheral ostectomy and carnoys solution will serve as adjunct methods to give an overall good prognosis (readers are also advised to refer chapter 27 on odontogenic cysts for use of these adjunct measures in odontogenic keratocyst).

A 67 year old male patient presented with loosening of teeth and swelling of gums in the anterior mandible

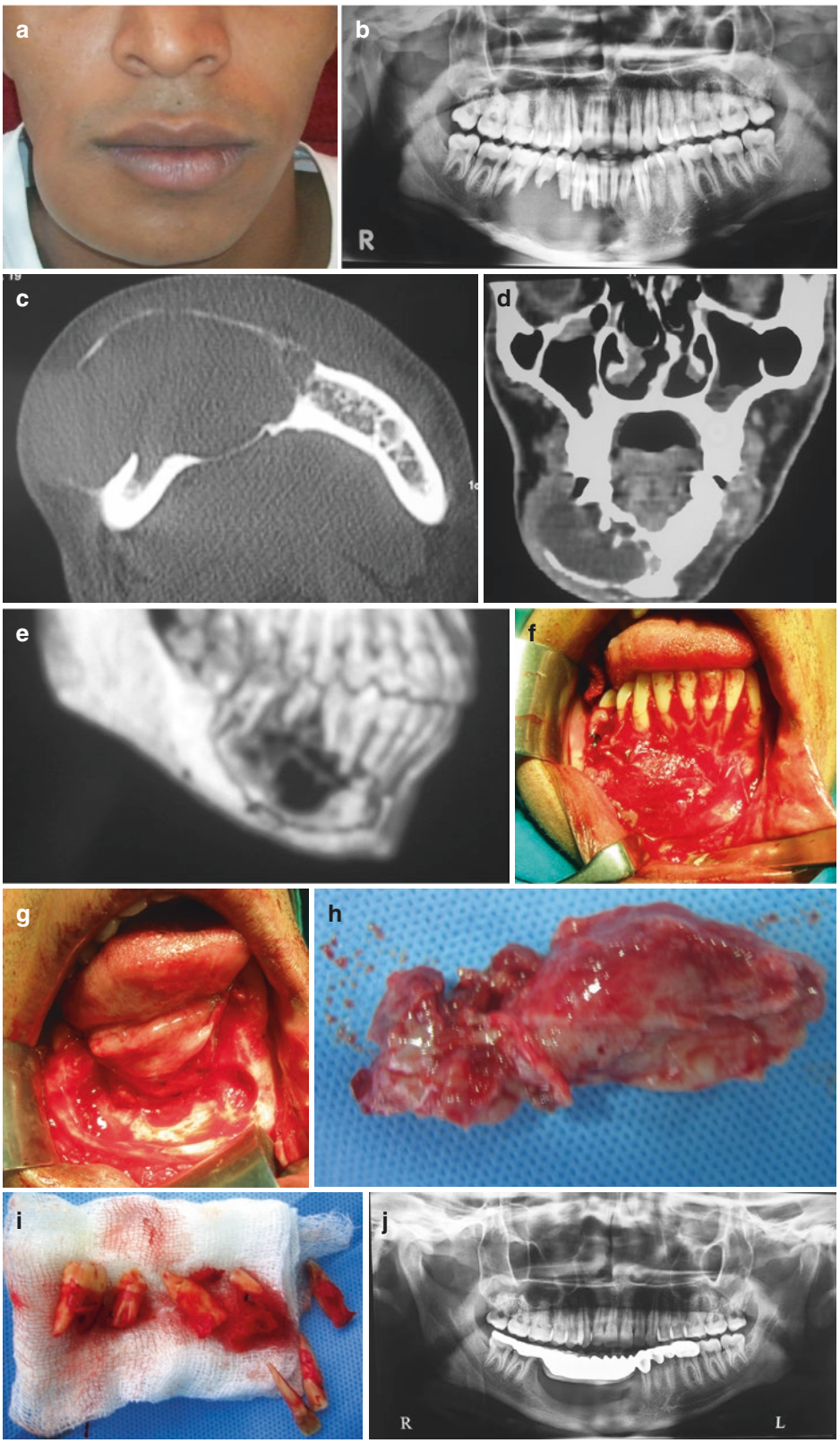

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Fig. 28.6 (**a**) Clinical image showing swelling right body mandible. (**b**) opg showing the radiolucent lesion right body mandible with root resorption. (**c**) Axial CT image showing the expansile lesion with thinning of the labial cortex. (**d**) Coronal CT image showing the expansile nature of the lesion. (**e**) 3D CT showing the erosion of both labial and lingual cortical walls. (**f**) Intra-oral intra-operative view showing the lesion exposed by crevicular flap. (**g**) Intra-oral view of the surgical bed after extraction of teeth, excision of lesion and peripheral ostectomy. (**h**) Image of the excised cystic lining, with thick walls. (**i**) Images of the extracted teeth showing the root resorption, (**j**) post operative radiograph after prosthetic rehabilitation

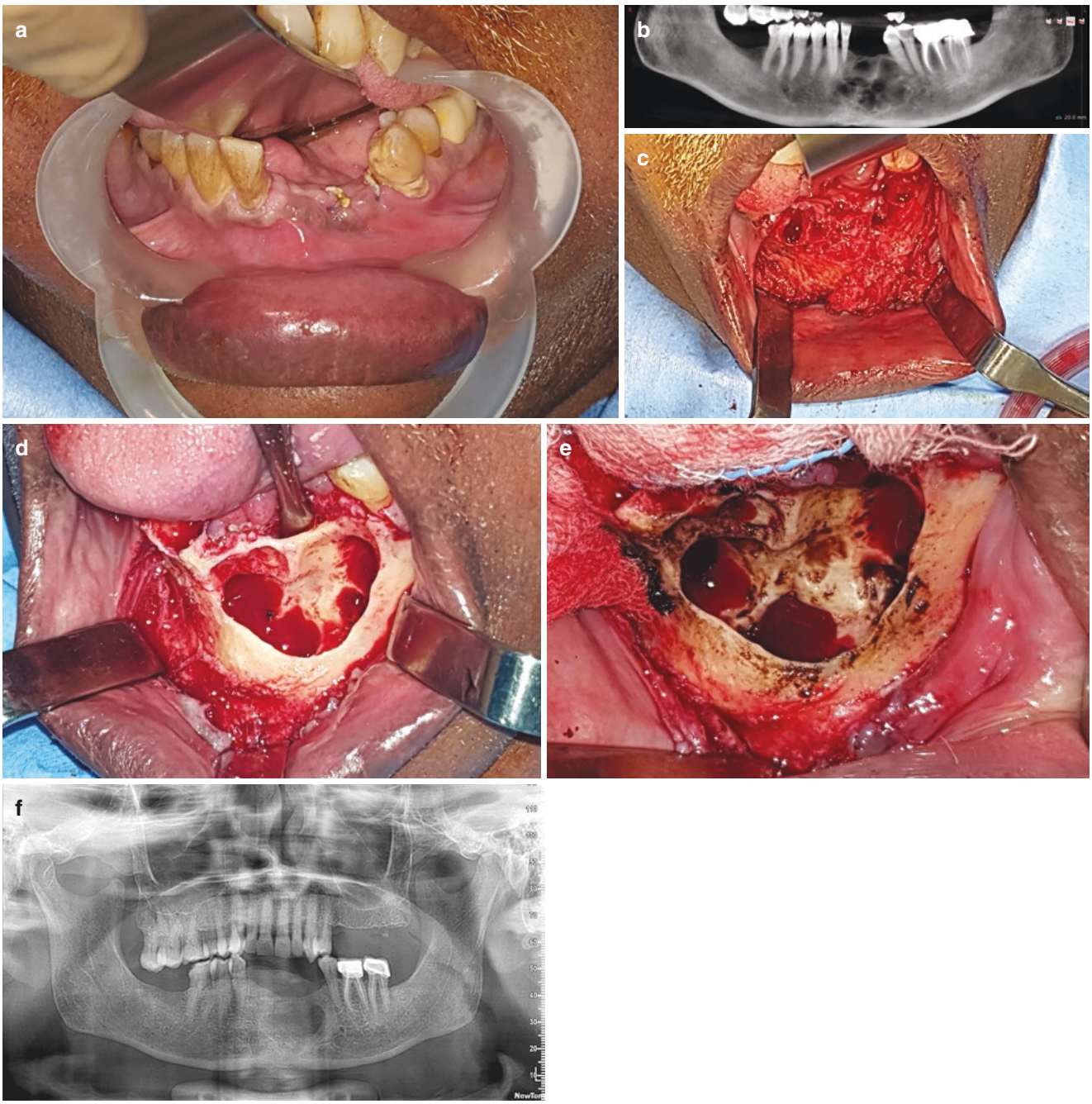

Fig. 28.7 (**a**) loosening of teeth and expansion of the gums in the anterior lower mandible. (**b**) A OPG and CBCT showing multilocular radiolucency in the anterior mandible. (**c**) Lesion exposed (**d**) lesion excised and peripheral ostectomy done (**e**) carnoys solution applied to surgical bed (**f**) sixteen month post operative OPG

(Fig 28.7a). A CBCT scan (Fig 28.7b) confirmed the presence of a multilocular radiolucency in between right lateral incisor and left first premolar region. An incisional biopsy confirmed a diagnosis of a plexiform ameloblastoma.

Considering the patient's age and general health, it was decided to follow a conservative line of treatment to avoid the need for complex reconstruction.

The lesion was exposed under general anesthesia (Fig 28.7c) and the involved teeth and tumour lining was removed. A Large round bur was then used to remove 3mm of bony walls around the locules left by the lesion (Fig 28.7d). The lesion was treated with modified carnoy's solution for 3 minutes (Fig 28.7e) followed by irrigation with normal saline and primary closure achieved.

Excision pathology confirmed the diagnosis of a Plexiform ameloblastoma. The healing was uneventful. A 16 month post operative radiograph showed good bony healing (Fig 28.7f) without any recurrence and the patient is on long term follow-up.

28.8 Extraosseous/Peripheral Ameloblastoma (PA)

The tumour was first described by Kuru [58] 1911. The PA is defined as the tumour having intra-osseous ameloblastoma characteristics histologically but occurs in the soft tissues overlying the tooth-bearing areas of mandible and maxilla. In comparison to the intra-osseous solid/multicystic ameloblastoma, it is the extraosseous counterpart [59].

PA representing only 1–5% of all ameloblastomas considering it as a very rare odontogenic tumour. It is also called ameloblastoma of soft tissue, mucosal origin ameloblastoma and gingival ameloblastoma. It arises from the remnants of the odontogenic epithelium with in the lamina propria gingival or from the gingival epithelium of the basal cell layer.

It is painless, firm and exophytic growth with a smooth, granular and warty surface. Most commonly located in the oral mucosa or the gingiva. It occurs in wide age range groups, i.e. from 1 to 92 years with more than 64% of cases occurring in the fifth to seventh decade of life. In edentulous areas, it affects the alveolar mucosa. The ratio of 2.4:1 is noted in mandible: maxilla. Intra-osseous ameloblastoma rarely extends to the gingival tissues and merge with gingival epithelium creating PA of the exophytic type [60]. Histopathologically same histomorphic cell types of odontogenic epithelium seen in solid/multicystic variant as consist in PA.

28.8.1 Differential Diagnosis

1. Peripheral odontogenic fibroma
2. Peripheral variant squamous odontogenic tumour

It does not show invasive behaviour and the treatment of choice will be conservative excision with adequate disease-free margins. The recurrence is low, but patients are to be kept on long-term follow-up.

28.9 Metastasising (Malignant) Ameloblastoma

'Malignant Ameloblastoma (MA)' term was proposed by Slootweg and Muller [61] in 1984. According to them, it is well-differentiated ameloblastoma that metastasises but which maintains the characteristic cytologic features of original tumour [62]. The ameloblastic tumour that undergoes malignant cytologic transformation; the term 'Ameloblastic carcinoma' is used.

Metastasis is the only factor that distinguishes ameloblastoma from MA. Its clinical behaviour is helpful in diagnosing the tumour.

Lung shows maximum 75% metastatic deposits, 15% each by spine and cervical lymph nodes followed by cervical lymph nodes and spine (15% each) [63]. Small bowel, liver, skull, brain and kidneys were the other locations, which show metastases but with lower incidence [64].

MA is very rare with an occurrence rate of 2% of all benign ameloblastomas [65]. The age ranges from 5 years to 74 with 34.4 years mean age. 1:1.2 is the male to female ratio. The majority cases were localised in the mandible. The survival time ranges from 3 months to 5 years after the appearance of metastases. Most MA are histologically plexifom type but not significantly different from the metastatic type in their histologic and cytologic features [66].

The initial tumours were treated with Enucleation and curettage while advanced with resection (block or segmental). Open thoracotomy is indicated in discrete and isolated lung metastases and wedge resection. Occasionally, chemotherapy gives successful results. Inoperable metastatic deposits can be treated with radiation therapy but having an unpredictable response [67, 68].

28.10 Squamous Odontogenic Tumours

28.10.1 Introduction

Squamous odontogenic tumour (SOT) was first described by Pullon et al. [69] as a rare benign odontogenic tumour in 1975. It affects all the age group and equally occurs in both the jaws. SOT histologically characterised by squamous epithelial islands, which are surrounded by mature connective tissue stroma. The SOT is occasionally misdiagnosed as squamous cell carcinoma, keratocanthoma, ameloblastoma and verrucous carcinoma. It is hamartomatous epithelial proliferation, arising probably from cell rests of Malessez [70]. The differential diagnosis of SOT may be acanthomatous and desmoplastic ameloblastoma variants, squamous cell carcinoma (well differentiated).

28.10.2 Definition

"SOT is a locally infiltrative, benign neoplasm consisting of islands of well-differentiated squamous epithelium in a fibrous stroma. The epithelial islands shows foci of central cystic degeneration occasionally" (WHO).

28.10.3 Clinical Features [71]

1. Swelling, which is painless
2. Mildly painful gingival swelling
3. Loosening of involved teeth
4. Occurs in the mean age of 38 years with age ranges from 8 to 74 years.

5. Multiple SOT, which involves several quadrant of mouth in few patients have been reported, a family of 3 siblings having multiple lesions has been reported.
6. Absence of periodontal ligament between the lesion & the root of the tooth, suggesting that lesion arises from Malassez rests cell in the periodontal ligament or closely adjacent mucous membrane.
7. Equally affects maxilla and mandible, but most commonly involves incisor-cuspid area in maxilla and bicuspid-molar area in mandible

28.10.4 Radiographic Features [72]

1. Unilocular radiolucency of the triangular or semicircular type located in alveolar bone along the roots' lateral surface.
2. In some cases, vertical bone loss appears
3. The radiolucent area may show ill-defined or well-defined margins
4. Sometimes, tumour appears as an intra-bony pocket between the teeth
5. Peripheral lesions may cause saucerisation of underlying bone, which is likely because of the pressure phenomenon rather than infiltration of the tumour.
6. Few extensive lesions show multilocular radiolucency, pushing the maxillary sinus and involving the mandibular body region.

28.10.5 Treatment and Prognosis

Conservative treatment in the form of local excision, enucleation and curettage may be done for the successful management of SOT. Recurrent and clinically aggressive lesions have been treated with en bloc excision. Extraction of the associated teeth along with the conservative treatment is mandatory. It has a very low recurrence rate [73].

Cortical bone erosion of maxilla and mandible exhibits aggressive biological behaviour. Aggressive treatment should be followed for the lesions, which show early recurrence [74].

28.11 Calcifying Epithelial Odontogenic Tumour

Pindborg in 1955 first introduced the calcifying epithelial odontogenic tumour (CEOT) in the scientific literature [75]. It is well known as 'Pindborg Tumor' since then. CEOT is slow growing, benign and occasional locally invasive odontogenic neoplasm, which is epithelial in its origin.

WHO accepted and adopted the term calcifying epithelial odontogenic tumour (CEOT) in its first edition of 'Histological typing odontogenic tumours, jaw cysts and allied lesions', and recognised it as a distinct entity [76]. It may be extra-osseous or intra-osseous.

28.11.1 Definition

CEOT is a epithelial odontogenic neoplasm, which is locally invasive and is characterised by the presence of amyloid material that may become calcified (WHO)

28.11.2 Epidemiology

It accounts for less than 1% of all odontogenic tumours, hence considered uncommon. It commonly occurs between the age of 8 and 92 years with the mean age of 36.9 years.

The intra-osseous variant occurs in the third, fourth and fifth decade of life in 64% of patients [77]. CEOT has no gender predilection, and occurs equally in both the sexes. Premolar and molar regions are the commonest site of occurrence, although can occur at any site. Anterior gingiva is most commonly affected by the peripheral lesions. Maximum cases reported are of intra-osseous lesions, only 6% arise in extra-osseous locations. Mandible is affected by intra-osseous lesions more frequently than maxilla, with a ratio of 2:1.

28.11.3 Clinical Features

CEOT is slowly growing, painless, expansile and hard bony swelling, which can cause thinning of the cortical bone and infiltration of soft tissue subsequently. It can cause rotation, tipping, migration or mobility of tooth secondary to resorption of roots. In the anterior region, there is also a distinctly uncommon peripheral variant of CEOT, limited to soft tissue only, presenting as a nodular mass on the gingiva.

28.11.4 Radiographic Features

The larger or the mature tumour will be mixed radiolucent—radiopaque, although the early tumour may be completely radiolucent. CEOT is often associated with unerupted teeth. It may be unilocular and cystic in appearance. It can demonstrate a mixture of large and small multilocular spaces described as 'soap bubble' and 'honey comb' in appearance. The radiographic borders in almost all cases between surrounding tissues and tumour appear to be circumscribed and well defined [78].

CEOT on CT examination demonstrating thinning and expansion of lingual and buccal cortical plates with well-defined mass containing scattered radiopaque areas of different size and signal intensity in mandible. Pindborg tumour on MRI reveals predominantly a hypointense lesion on T1-weighted images and mixed hyperintense lesion on T2-weighted images [79].

28.11.5 Treatment

Surgical management is the treatment of choice for CEOT. Conservative treatment in the form of Enucleation or curettage followed by judicious removal of the thin layer of bone adjacent to the tumour is the choice of treatment in small, intra-bony lesions with well-defined borders. However, the tumours treated with curettage and enucleation show a recurrence rate ranging from 15 to 30% after 2–4 years with the overall recurrence rate of 14% [77].

According to Melrose RJ [80], even the small CEOTs are infiltrating in nature. A margin of about 1 cm normal bone should be removed along with the tumour excision. Peripheral tumours are treated with smaller margins 0.5 cm because they are less aggressive. Few recurrences have been reported with the tumours treated with jaw resection. However, the patient should be kept for follow-up upto 5–10 years.

The recurrent lesions and the tumours, which are diagnosed late in their clinical course, which over an extended time becomes larger and extensive (more than 4 cm in size) may not respond well to conservative management-like surgical excision only. Segmental resection such as partial or hemimandibulectomy or hemimaxillectomy will be the treatment of choice. However, it may leave a significant bony discontinuity requiring grafting or extensive soft tissue reconstruction.

The incidence of malignant transformation is very low; however, Veness et al. [81] reported a case of metastatic spread and malignant transformation with CEOT.

28.12 Adenomatoid Odontogenic Tumour

Stafne in 1948, first described the adenomatoid odontogenic tumour (AOT) as an odontogenic neoplasm [82]. It was referred as ameloblastic adenomatoid tumour or adenoameloblastoma initially, because it was considered as a variant of ameloblastoma [83]. The term AOT, which is generally accepted today suggested by Philipsen and Birn [84].

28.12.1 Definition [85]

A neoplasm of locally invasive nature was characterised by ameloblastoma-like islands of epithelial cells in a mature connective tissue stroma. Aberrant keratinisation may be found in the form of ghost cells in association with varying amounts of dysplastic dentin (WHO).

Three variants of AOT are recognised, i.e. Follicular, Extra follicular and peripheral.

The peripheral type arises from the gingival tissues and is very rare. The follicular type is commonly associated with impacted tooth and found in 75% of cases; on the other hand, the extrafollicular type is located between the roots of adjacent teeth and is not related to an unerupted teeth [86].

28.12.2 Clinical Features

- AOT commonly occurs in the anterior region of jaws, particularly the maxilla.
- It occurs commonly in young patients, two-thirds of the cases occurring between 10 and 19 years of age. However, the age ranges from 3 to 82 years [87].
- Females are affected more than males with a ratio of 5.6:1.
- Size of AOT ranges from 2 to 7 cm with more than 60% involving the entire quadrant [88].
- The most common tooth associated with AOT is impacted canine followed by premolars and lateral incisors [89].

Radiographically, the most common appearance will be a well-demarcated unilocular radiolucency associated with unerupted tooth. Sometimes, intra-bony cases show scattered radiopacities within the radiolucency. Intra-oral periapical radiographs found to be better than OPG are best suited for showing discrete calcified deposits [90].

The AOT is usually well-encapsulated tumour so the treatment of choice will be Enucleation and Curettage. The recurrence is extremely rare [91].

28.13 Mixed (Epithelial-Mesenchymal) Origin

28.13.1 Ameoblastic Fibroma

Ameloblastic Fibroma (AF) was first reported by Kruse in 1891 [92]. AF consists of odontogenic ecto-mesenchyme resembling the dental papilla, epithelial strands and nests resembling dental lamina and enamel organ. Dental hard tissues are absent in it. The lesion is referred to as ameloblastic fibro-dentinoma if there is dentin formation. It is a rare odontogenic mixed tumour in which ectomesenchymal and epithelial elements are neoplastic. It accounts only 2% of all odontogenic tumours [93].

28.13.1.1 Clinical Features

AF occurs in young adults and children with an age range from 1.5 to 42 years, with the average age ranging from 14.5 to 15.5 years [94]. It was occasionally reported in middle-aged individuals. There is no gender predilection. More than 50% of patients present with a sign of swelling. Failure of tooth eruption, pain, tenderness and discharge are the other common findings [95]. Posterior mandible is the commonest site of occurrence and first permanent molar and second primary molar areas were involved in more than 70% of cases.

28.13.1.2 Radiographic Features

Multilocular radiolucency with sclerotic margins is the most common radiographic appearance. Unilocular radilolucency is the feature of small tumours, while large tumours extend through the bony cortices. The size typically ranges from 1

to 8 cm [96]. Ameloblastic fibrosarcoma is the differential diagnosis of AF.

Enucleation and curettage of the adjacent bone along with the extraction of affected teeth is the treatment of choice for AF. They occur rarely but required a long-term followup. In order to preserve the teeth involved in the tumour, the chances of recurrence increase after conservative management [97, 98].

The ameloblastic fibro-odontoma is composed of connective tissue characteristic of an ameloblastic fibroma and calcified tissue identifying the tumour as a complex odontoma. The ameloblastic fibro-odontoma diagnosis is based on the histologic evidence of ameloblastic fibroma with active odontogenic epithelium embedded in an embryonal connective tissue. The differential diagnosis of ameloblastic fibroma and ameloblastic fibro-odontoma is based on the absence (ameloblastic fibroma) and presence (ameloblastic fibro-odontoma) of enamel and dentin. A tumour is called ameloblastic fibro-dentinoma when exclusive dentin formation is observed. A tumour is called an ameloblastic fibro-odontoma in the presence of both enamel and dentin. In ameloblastic fibroma, no dental hard tissues are present [99]. AFO is similar to AF described by the WHO, and [they also show inductive changes that lead to both enamel and dentin formations]. Moreover, AFO and AF are defined as hamartomatous lesions and they are believed to be stages of formation of odontoma. That means the above-mentioned lesions should not be considered as distinct entities [100].

28.13.2 Odontoma (Compound, Complex Types)

In 1866, Broca coined the term 'Odontome' [101]. The term 'Odontoma' was given by Thoma and Goldman, to include tumours that were composed of well-differentiated tooth structure [102]. Odontoma defined as a tumour that differentiated and developed enough to produce dentin, enamel and cementum in varying proportions [103].

Compound odontoma is defined by the WHO as a malformation in which all the dental tissues represented in a more orderly pattern than complex odontoma, so that lesions consist of many tooth-like structures.

Complex odontoma is a type of malformation in which all dental tissues are represented and individual tissues are well formed, but occur in a disorderly pattern [104].

Compound-type odontomas are more common than complex odontomas [105]. According to Regezi, the compound odontomas are 37% and complex odontomas were 30% of all the reported odontogenic tumours [106]. Odontomas occur equally in both sexes. They are rarely associated with deciduous teeth but more frequently associated with permanent teeth. Anterior maxilla is the most common site for the occurrence of compound-type odontome while posterior mandible (Fig. 28.8) is affected more commonly in complex odontomes followed by anterior maxilla [107].

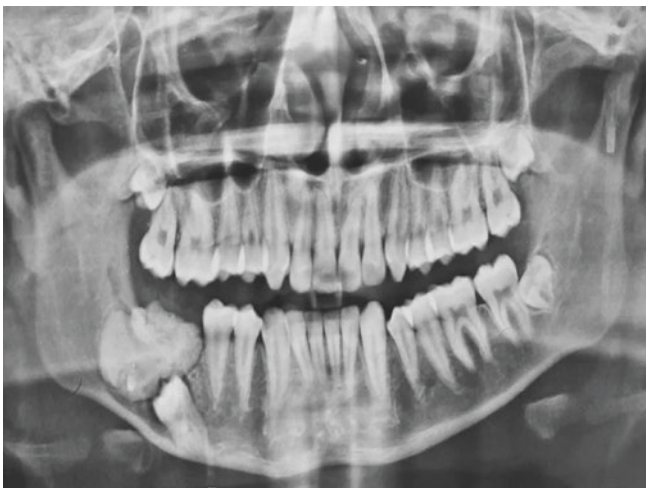

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Fig. 28.8 OPG showing complex odontome in mandible right molar region

They are small, painless and hard mass. Permanent impacted or retained deciduous teeth are present as a common symptom. The second most common complaint is swelling. Complex odontoma may become large and produce asymmetry with bone expansion [108, 109].

On radiological examination, the lesion consists of densely opaque masses of varying size, usually associated with unerupted or impacted teeth. The radiolucent line surrounds the opaque masses. The collections of tooth-like structures of different shapes and sizes are the features of compound odontomes, the teeth are of diminutive sizes. The complex-type odontomes appear as calcified masses that have consistency the same as the tooth structure.

The treatment of choice for odontomes is surgical excision and recurrence is very rare.

28.14 Mesenchymal Origin Tumours

28.14.1 Odontogenic Myxoma

Myxoma is very uncommon. It accounts for 0.5 to 20% of all odontogenic tumours and has a incidence of 0.07/million [110]. It arises from mesenchymal stroma and is a benign, locally aggressive tumour. Bone, soft tissues and most frequently the myocardium are favourite sites for myxomas. Jaws myxomas are both odontogenic and osseous in the origin . Myxoma according to WHO is defined as a locally invasive neoplasm, consisting of angular and rounded cells that lie in an abundant mucoid stroma [111].

28.14.1.1 Clinical Features

The myxomas have a predilection for molar and premolar regions of mandible and maxilla, however they can occur

anywhere in the jaws. One third of myxomas located in the maxilla while two third in mandible. In maxillary myxomas, the cortical expansion and perforation of bone are common and often extend into the sinus [112].

It is reported to have a slight female predilection (1.5:1) while other authors reported equal gender predilection between females and males. Odontogenic myxomas most commonly occur in the second and third decade of life with the average age ranging from 25 to 30 years [113].

The periapical radiographs and OPGs are the first indicators of myxomas of jaws. Odontogenic myxomas radiographically present as unilocular lesions to large multilocular neoplasms, which often cause displacement of teeth but resorb the roots less frequently. The unilocular lesions are small in size on the other hand multilocular myxomas are greater than 4 cm in size. 5% Myxomas have association with unerupted tooth [114].

28.14.1.2 Treatment and Prognosis

Surgical excision is the treatment of choice of odontogenic myxomas. The rate of recurrence is quite high, i.e. nearly 25% during first two years after removal. Atleast 1 cm of medullary bone should be resected along with tumour and always try to involve one tumour-free anatomic barrier at its periphery [78]. Subramaniam S [115] used endoscopes in the resection of pterygoid plates for the complete treatment of odontogenic myxoma. Use of endoscopes eliminates the use of extra-oral incisions.

28.14.2 Cementoblastoma

Dewy in 1927 first described Benign cementoblastoma [116]. It is a rare tumour of mesenchymal origin. Cementum-like tissue formation around the roots of the teeth is its important characteristic.

Cementoblastoma has predilection of mandible with 79.5% and most commonly in the premolar and molar region. They rarely occur in maxilla.

The roots of vital erupted permanent tooth are affected by the cementoblastoma. The most common tooth involved is the mandibular first molar. It has slight male predilection and occurs most commonly in the second and third decade of life. Cementoblastoma can cause teeth displacement, expansion of bone and maxillary sinus invasiveness, and aggressive tumours usually present with the symptoms of swelling and pain [117].

The diagnosis of cementoblastoma is very challenging as hypercementosis is also associated with roots of the teeth, that is the reason why hypercementosis is always included in the differential diagnosis. Cementoblastoma is slow growing, but can cause perforation and expansion of the cortices. The most common symptom is pain [118]. Radiographically the lesion will appear as a radiopaque mass fused with roots of the teeth , surrounded and limited peripherally by a radiolucent halo.

Complete excision of cementoblastoma is the gold standard treatment with extraction of the involved teeth [119]. It must be removed early, otherwise it may continue to grow. Maxillary lesions at times can involve the entire maxillary sinus making the prognosis poor. Recurrence is very rare, however incomplete removal may result in recurrence.

The tooth associated with cementoblastoma may be preserved under the following conditions [120]

1. In slow-growing tumour
2. In asymptomatic patients, with no pain
3. Perforation of cortical plates is absent
4. Patient refusal for teeth extraction

28.15 Recent Advances

Genetic marking has helped identify gene mutations, which may in the future help with histological and clinical management of these lesions. The most promising of these is the BRAF V600E mutation that is present in 90% of ameloblastomas and in the unicystic mural variant, suggesting predisposition to infiltrative behaviour. In the future, gene therapy may be possible using this gene marker [121].

The sonic hedgehog (SHH) and PI3K/Akt/Mtor signaling pathways may soon provide non-surgical options for treatment of ameloblastoma. The tumours that depend on active SHH signalling for growth/survival and maintenance may be susceptible targets for combined chemotherapy with SHH-specific inhibitors together with PI3K, Akt or mTOR blocking agents [122]. Jhamb T and Kramer JM advise to check molecular markers and accordingly decide the treatment plan [123]. Effiom OA et al. reported that explanation of molecular factors that arrange pathogenesis and recurrence of ameloblastoma will lead to new targeted drug therapies and diagnostic markers for ameloblastoma [124].

28.16 Case Scenario 5 (Fig. 28.9a–j)

A 25 year old male patient presented with an expanding lesion around the left upper posterior teeth (Fig. 28.9a). Radiographs suggested a cystic lesion and a CT scan confirmed bony destruction by a multilocular lesion extending from the first molar to the tuberosity (Fig. 29.8b). An incisional biopsy of the lesion confirmed a diagnosis of follicular ameloblastoma.

The patient had excision of the lesion with a posterior partial maxillectomy, with a mandibular split [125] for access (Fig. 28.9c, d). The defect was reconstructed with a temporalis muscle flap (Fig. 28.9e, f, g). The mandibulotomy site was fixed with titanium plates (Fig. 28.9h).

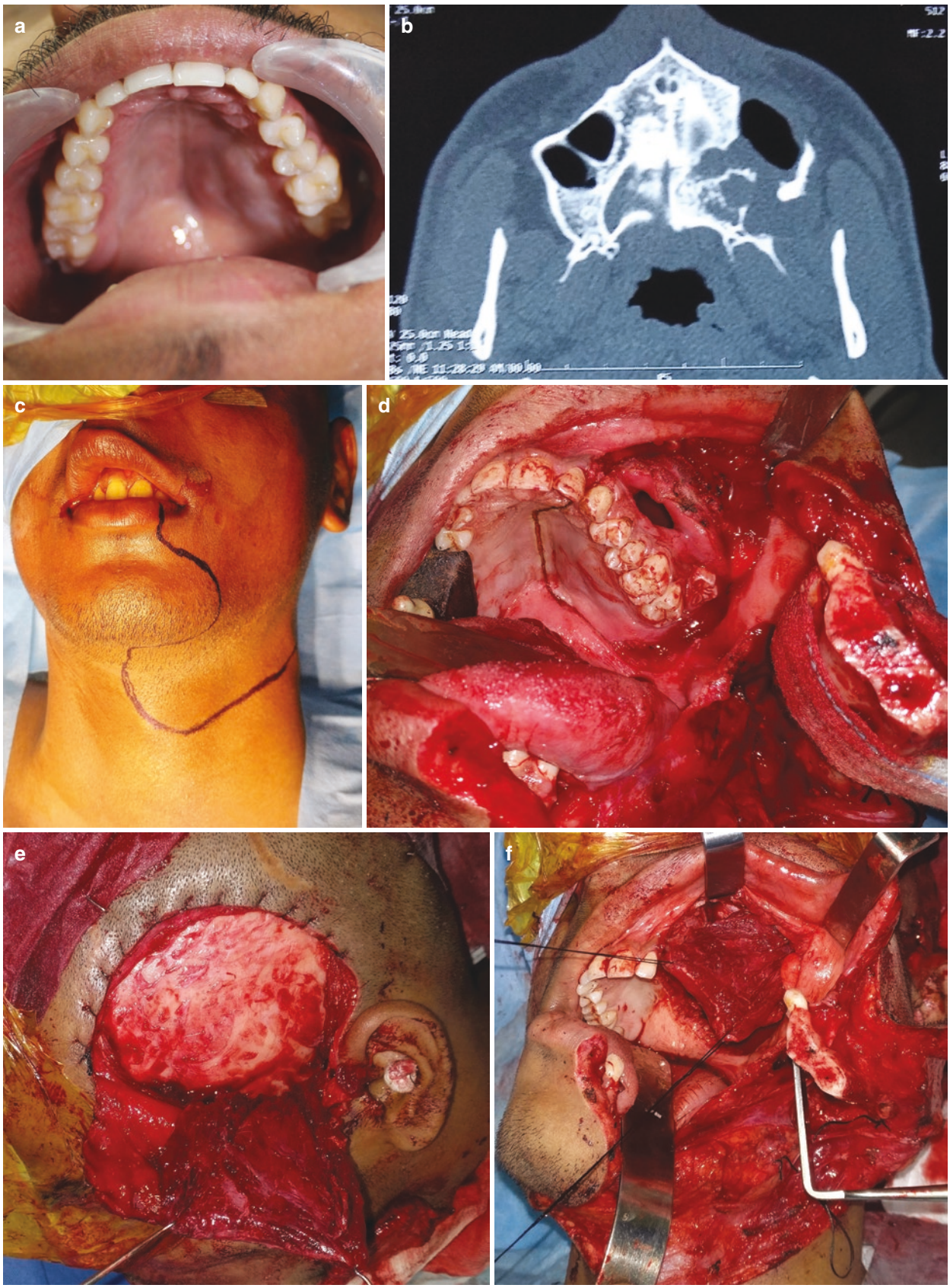

Fig. 28.9 (**a**) expanding lesion around the left upper posterior teeth. (**b**) CT scan confirmed bony destruction by a multilocular lesion extending from the first molar to the tuberosity. (**c**) Incision marked for partial maxillectomy via mandibular split access (**d**) left posterior partial maxillectomy being performed (**e**) temporalis muscle flap being harvested (**f**) temporalis flap being advanced to the partial maxillectomy defect (**g**) Introperative view after suturing of the temporalis flap to cover the defect. (**h**) Fixation done at the mandible split region (**i**) post-operative intra oral view showing good healing (**j**) extra oral view showing good cosmetic and functional result

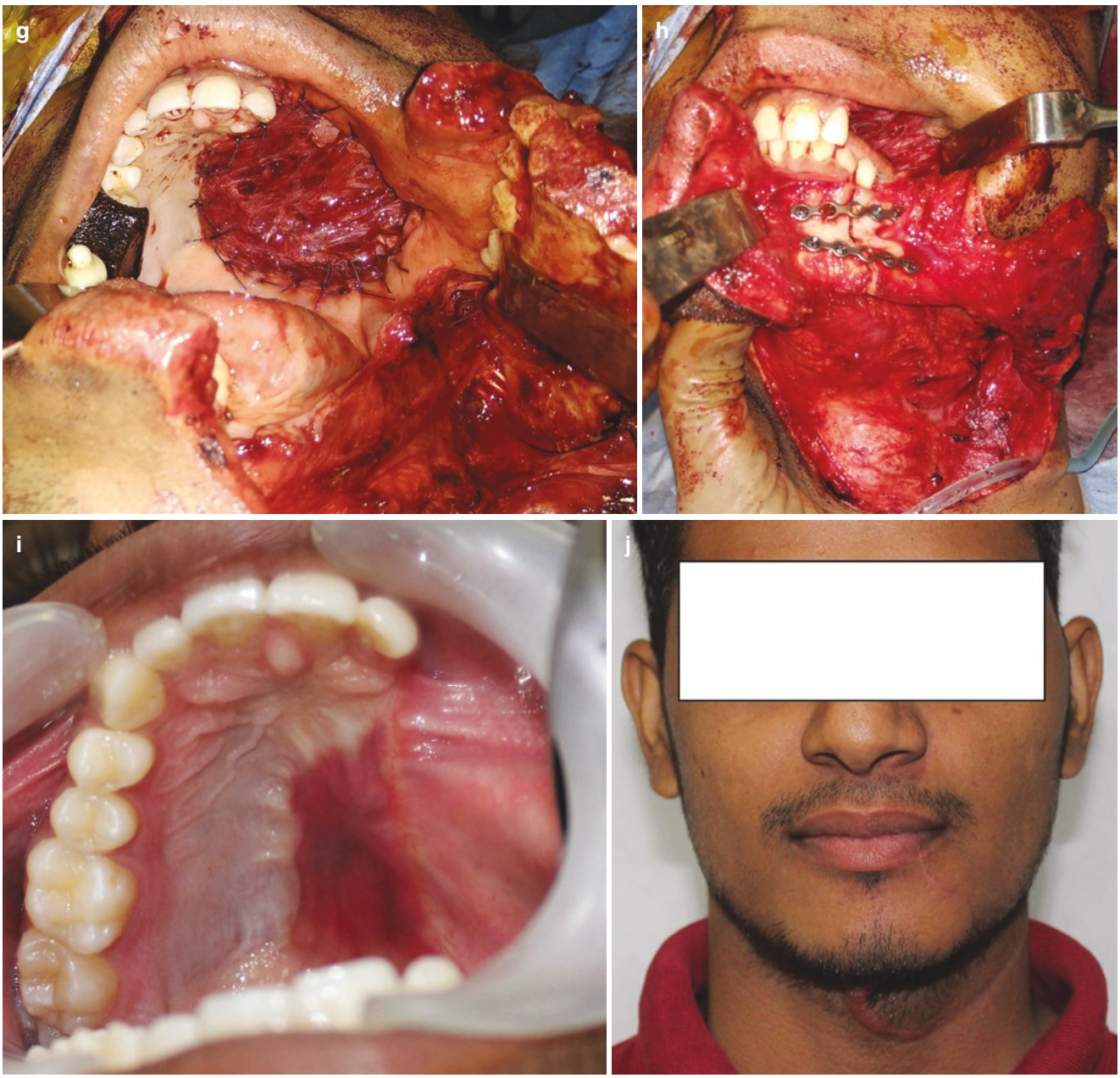

Fig. 28.9 (continued)

The healing was uneventful. Pathology reports confirmed the diagnosis of Follicular Ameloblastoma with clear excision margins. He had an excellent intra oral and cosmetic (extra oral) post-operative result (Fig. 28.9i, j).

Maxillectomy through mandibulotomy approach has been followed by author in a series of cases [125].

The readers are advised to refer chapter 85 of this book for detailed reading on access surgeries and osteotomies for the maxillofacial region.

28.17 Conclusion

Posterior mandible and cuspid areas of maxilla are the most common sites for the odontogenic tumours to occur. However, they can occur anywhere in the tooth-forming apparatus. The detailed history, thorough radiological and clinical evaluation is important in making the probable diagnosis. Conservative and aggressive surgical management has been used according to the size and extent of the tumour.

Proper aesthetic reconstruction should be carried out to avoid facial disfigurement. To check for recurrence, patients should be kept on regular follow-up and ensure long-term successful results.

Acknowledgements for Figs. 28.4 and 28.6 (case scenarios 2 and 3) to suvy manuel

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