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Case Report

Huge keratocystic odontogenic tumour in medically compromised patient–a management dilemma

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ABSTRACT

Keratocystic odontogenic tumour (KCOT) is a cystic lesion of the jaws with tumour behaviour. Its high prevalence rate makes it one of the commonest cystic lesions especially involving the lower jaw. The characteristic histologic features and aggressive nature corresponds to the high recurrence rate associated with KCOT. Lesion expands mostly in an anteroposterior direction and can cause extensive bone destruction before the appearance of any clinical symptoms. The characteristic radiological picture is that of a multilocular cystic lesion with the common differential diagnosis being dentigerous cyst and ameloblastoma. Here we are presenting a case of KCOT of the left lower jaw of size 10.9×7.86×8.54 cm. It is a huge multilocular cystic lesion extending from the right canine region to the left side involving the body, ramus, coronoid and condyle. Various management options are there ranging from enucleation and chemical cauterization to resection and reconstruction depending upon the size of the lesion. In this case we were not able to perform the ideal treatment option for the case because of the multiple drug allergy the patient was having, including most of the general anesthetic agents. Also the patient was not willing for any extensive procedure under general anesthesia. So we had to follow a compromised treatment plan aiming to reduce the size of the lesion, to improve the aesthetics and frequent follow up.

Keywords: KCOT, OKC, CECT, HPR, Aspirate, Enucleation, Resection

INTRODUCTION

KCOT is defined as a benign unicystic or multicystic intraosseous tumour of odontogenic origin with characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive infiltrative behavior. KCOT is a widely occurring tumour in the jaw with high recurrence rate. It was first described by Mikulicz in 1876, who considered it as a dermoid cyst. Hauer described it as a type of cholesteatoma.[1] Robinson in 1945 explained it as a primordial cyst found in the place of a missing tooth, arising from remnants of dental lamina or enamel organ.[2] Based on the histologic appearance Philipsen in 1956 coined the term Keratocyst.[3] Essential histologic features like flattening of basement membrane and pallisading of basal cells was described by Pindborg and Hensen in 1963.[4] Based on the histologic behavior keratocyst was classified as orthokeratinzed and parakeratinized variants. Characteristic histologic picture explains the proliferation kinetics and aggressive behavior. Shear in 2002 explained the difference between parakeratinized and orthokeratinized variant in expression of cytokeratin, epithelial membrane antigen and carcinoembryonic antigen, and described ortokeratinized variant as less aggressive.[5] Based on the aggressive behavior and new information regarding the genetics, WHO in 2005 reclassified OKC as KCOT, keratocystic odontogenic tumour. Large area of bone involvement, high recurrence rate, and distinct histopathology, disregulation of PTCH gene, association with NBCC and other abnormalities and carcinoma arising from OKC explains the tumour behavior of KCOT. Here we are discussing a case of

huge KCOT involving the mandible, with issues in proper management due to large size and medical conditions of the patient prohibiting general anesthesia.

CASE REPORT

A 76 year old male patient reported to our department with swelling in the left side of face since 15 years. He had marked aesthetic deformity due to the swelling. The lesion was progressively enlarging with occasional pain. Teeth in the lower left arch were removed 10 years before. He had approached multiple centers for treatment, and was finally referred to our center. Definitive management of the pathology was not possible due to complicating medical issues depriving general anesthesia. In some of the centers they have tried aspirating the lesion, which showed a mild remission for a period of one week with further collection of cystic fluid and returning to the previous size. The patient had history of multiple drug allergies and on allergy test showed reaction to most antibiotics, analgesics, general anesthetic agents and muscle relaxants making it least possible to do the procedure under general anesthesia. Also the patient seemed to have low pain threshold, as it was difficult for him to tolerate the procedure of aspiration of lesion under local anesthesia.

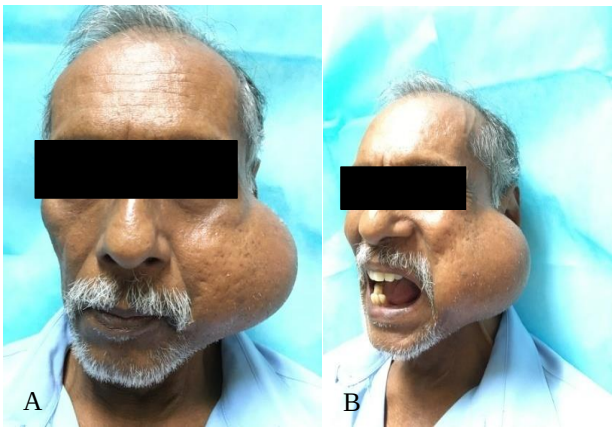


Figure 1: (A) Frontal view; (B) Profile view.

On extraoral examination the patient showed a 12×15 cm size lesion in the left side of lower jaw extending from the upper zygomatic arch region to the lower border of mandible, and anteroposterior extention from the angle of mouth to the posterior border of ramus. Patient had paresthesia in the region of lower lip, mouth opening was normal. Occassional pain was reported especially towards the posterior ramal region. The overlying mucosa appeared normal in colour and texture, no local rise in temperature and showed mild tenderness on palpation, fluctuancy of the lesion was noted. Intra orally the upper arch seems normal where as the lower arch was partially edentulous with swelling and fistula in the posterior buccal sulcus near the retromolar region. Discharge of dark brown coloured fluid was present introrally through the fistula. Aspiration of the lesion was done on the day the patient reported and the histopathology report was suggestive of inflammed odontogenic lesion. On behalf of the medical history and the history of drug allergy patient cosultation was done with the anesthetist,since the patient was allergic to both depolarising and nondepolarising muscle relaxants and general anesthetic agents like propofol and thiopentone sodium, patient was on high risk category for general anesthesia Patient was also not willing to undergo any procedure under general anesthesia.

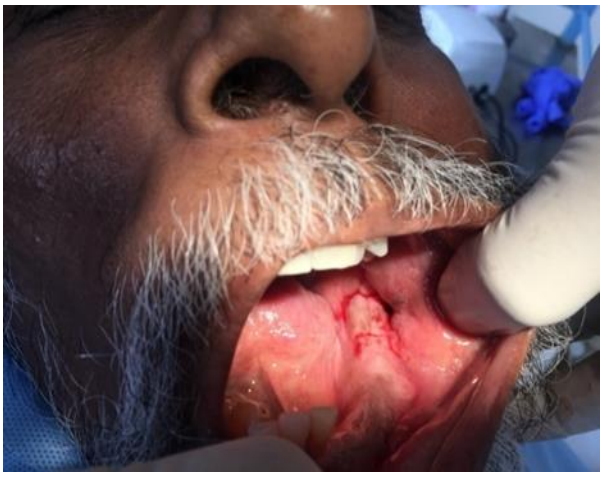
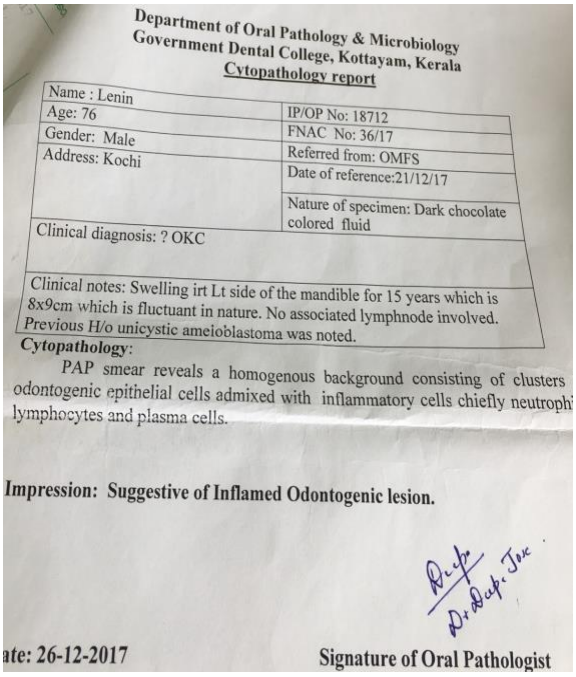


Figure 2: Lower arch.

The patient had a series of radiographs with him including the OPG and CT scan. In the OPG a huge multilocular cystic lesion of the left mandible from the 43 region extending to the symphysis, body, ramus and condyle of the left lower jaw.



Department of Oral Pathology & Microbiology
Government Dental College, Kottayam, Kerala
Cytopathology report

Name : Lenin	IP/OP No: 18712
Age: 76	FNAC No: 36/17
Gender: Male	Referred from: OMFS
Address: Kochi	Date of reference:21/12/17
	Nature of specimen: Dark chocolate colored fluid
Clinical diagnosis: ? OKC	

Clinical notes: Swelling irt Lt side of the mandible for 15 years which is 8x9cm which is fluctuant in nature. No associated lymphnode involved. Previous H/o unicystic ameloblastoma was noted.

Cytopathology:

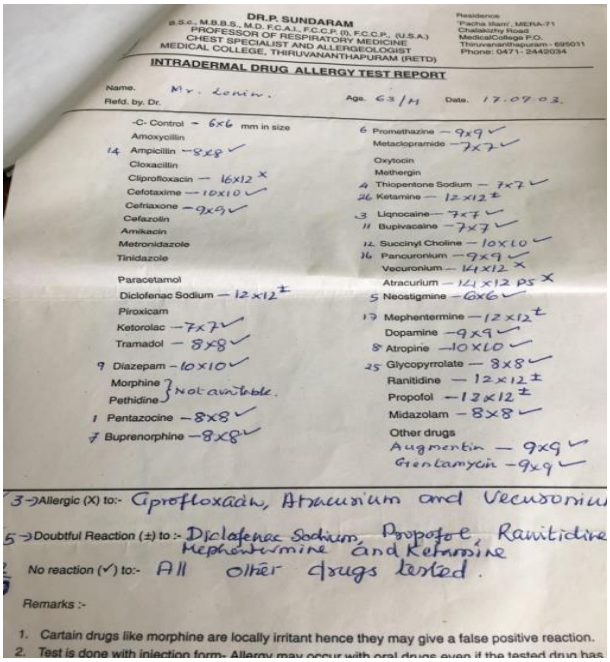
PAP smear reveals a homogenous background consisting of clusters odontogenic epithelial cells admixed with inflammatory cells chiefly neutrophi lymphocytes and plasma cells.

Impression: Suggestive of Inflamed Odontogenic lesion.

Dr Asst. Prof

ate: 26-12-2017 Signature of Oral Pathologist

Figure 3: HPR of aspirate.



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INTRADERMAL DRUG ALLERGY TEST REPORT

Name. Mr. Lenin. Age. 63/M Date. 17.09.03.
Refd. by, Dr.

-C- Control - 6x6 mm in size

Amoxycillin
14 Ampicillin - 8x8 ✓
Cloxacillin
Ciprofloxacin - 16x12 X
Cefotaxime - 10x10 ✓
Ceftriaxone - 9x9 ✓
Cefazolin
Amikacin
Metronidazole
Tinidazole

Paracetamol
Diclofenac Sodium - 12x12 ±
Piroxicam
Ketorolac - 7x7 ✓
Tramadol - 8x8 ✓
9 Diazepam - 10x10 ✓
Morphine } Not available.
Pethidine }
1 Pentazocine - 8x8 ✓
7 Buprenorphine - 8x8 ✓

6 Promethazine - 9x9 ✓
Metaclopramide - 7x7 ✓
Oxytocin
Methergin
4 Thiopentone Sodium - 7x7 ✓
26 Ketamine - 12x12 ±
3 Lignocaine - 7x7 ✓
11 Bupivacaine - 7x7 ✓
12 Succinyl Choline - 10x10 ✓
16 Pancuronium - 9x9 ✓
Vecuronium - 12x12 X
Atracurium - 12x12 PS X
5 Neostigmine - 6x6 ✓
17 Mephentermine - 12x12 ±
Dopamine - 9x9 ✓
8 Atropine - 10x10 ✓
25 Glycopyrrolate - 8x8 ✓
Ranitidine - 12x12 ±
Propofol - 12x12 ±
Midazolam - 8x8 ✓

Other drugs
Augmentin - 9x9 ✓
Gentamycin - 9x9 ✓

3 → Allergic (X) to:- Ciprofloxacin, Atracurium and Vecuronium
5 → Doubtful Reaction (±) to :- Diclofenac Sodium, Propofol, Ranitidine, Mephentermine and Ketamine
No reaction (✓) to:- All other drugs tested.

Remarks :-
1. Certain drugs like morphine are locally irritant hence they may give a false positive reaction.
2. Test is done with injection form. Allergy may occur with oral drugs even if the tested drug has

Figure 4: Allergy test report.

A characteristic soap bubble appearance was seen in the symphysis and body region, with root resorption of 41, 42 and displacement of 43. The lesion was close to the lower border of mandible in the posterior region. In the anterior region lower border is intact with about 1 cm of bone; in the upper border the lesion was abutting the bony margin. No clear definition of inferior alveolar nerve canal is seen. Upper left posterior tooth also showed displacement suggesting encroachment of the lesion into upper left alveolus.

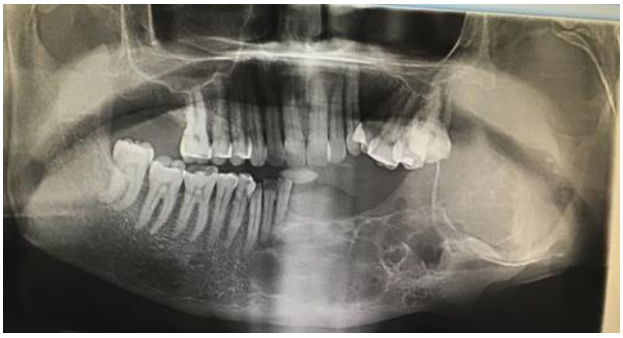


Figure 5: OPG.

Axial and the coronal CECT images also showed large cystic lesion involving the left side of mandible of size 10.9×7.86×8.54 cm extending to the left maxillary alveolus. Lesion is multilocular with soft tissue attenuation in the inferior aspect. Axial section showed areas of cortical destruction with thin fragmented bony wall as seen in the palpatory finding. In CT there is no evidence of perineural invasion. Coronal sections showed no invasion into orbit and nasal cavity. Superiorly there is erosion of left maxillary alveolus.

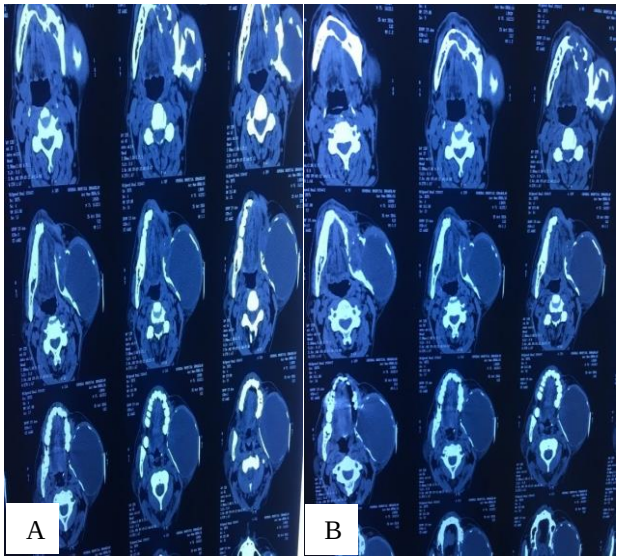


Figure 6: (A) Axial view 1; (B) Axial view 2.

The ideal treatment option is resection with continuity defect of left side of mandible along with involved portion of upper alveolus and soft tissue infiltrated areas followed by reconstruction with free flap. Because of the medical condition and since the patient was not willing to undergo the extensive procedure in general anesthesia, we had to manage the lesion conservatively. We tried intraorally aspirating the fluid content in the lesion in multiple sittings, about 100-150 ml of fluid aspirated at a time. After aspiration the size reduced making the thin fragmented bony wall palpable, but within two weeks the patient reported back with repeated collection. Another option tried was aspirating the lesion and packing intraorally with iodoform gauze. With that slight amount of reduction in the size of lesion was achieved along with a decrease in the rate of fluid reaccumulation. Further we have considered making an intraoral or extra oral incision and suturing a small portion of suction catheter in position to allow dependent drainage and self-aspiration by the patient.

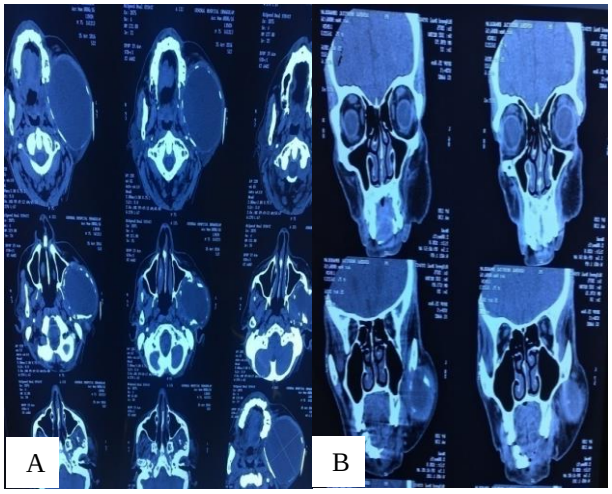


Figure 7: (A) Axial view 8; (B) Coronal view 1.

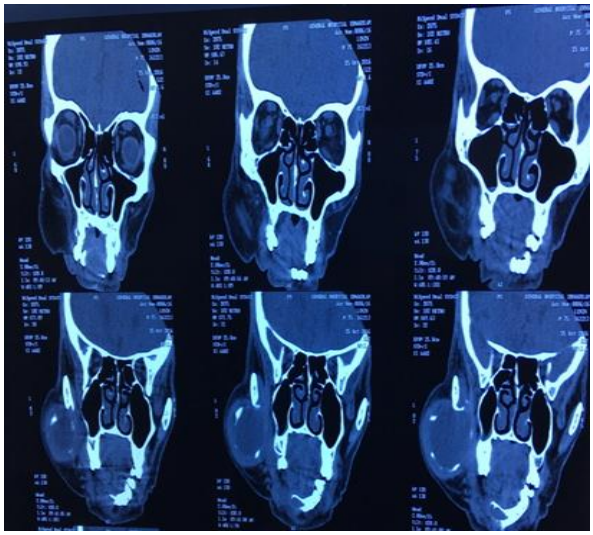
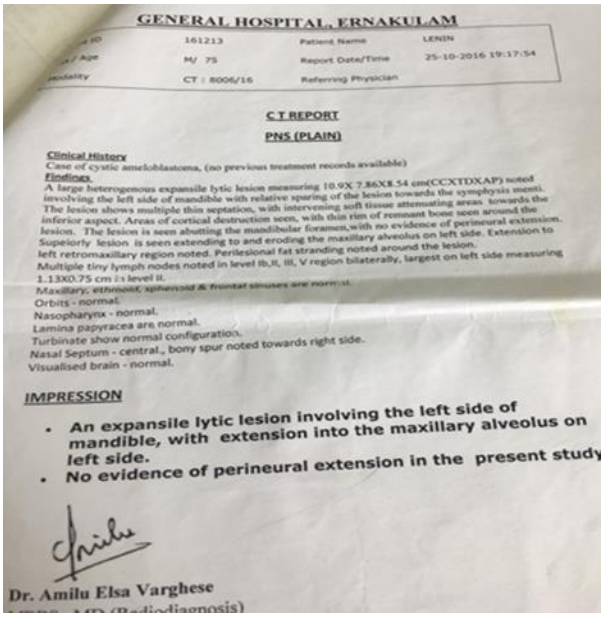


Figure 8: Coronal view 2.



GENERAL HOSPITAL, ERNAKULAM

161213 — Patient Name: LENIN
M/ 75 — Report Date/Time: 25-10-2016 19:17:54
CT : 8006/16 — Referring Physician

C T REPORT
PNS (PLAIN)

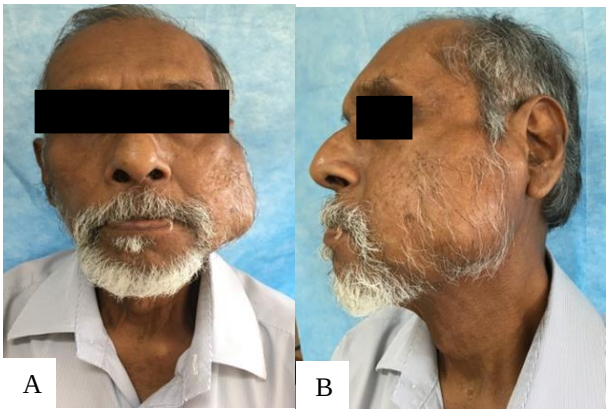
Clinical History
Case of cystic ameloblastoma, (no previous treatment records available)
Findings
A large heterogenous expansile lytic lesion measuring 10.9X 7.86X8.54 cm(CCXTDXAP) noted involving the left side of mandible with relative sparing of the lesion towards the symphysis menti. The lesion shows multiple thin septation, with intervening soft tissue attenuating areas towards the inferior aspect. Areas of cortical destruction seen, with thin rim of remnant bone seen around the lesion. The lesion is seen abutting the mandibular foramen,with no evidence of perineural extension. Superiorly lesion is seen extending to and eroding the maxillary alveolus on left side. Extension to left retromaxillary region noted. Perilesional fat stranding noted around the lesion.
Multiple tiny lymph nodes noted in level Ib,II, III, V region bilaterally, largest on left side measuring 1.13X0.75 cm in level II.
Maxillary, ethmoid, sphenoid & frontal sinuses are normal.
Orbits - normal.
Nasopharynx - normal.
Lamina papyracea are normal.
Turbinate show normal configuration.
Nasal Septum - central., bony spur noted towards right side.
Visualised brain - normal.

IMPRESSION

- An expansile lytic lesion involving the left side of mandible, with extension into the maxillary alveolus on left side.
- No evidence of perineural extension in the present study

Dr. Amilu Elsa Varghese

Figure 9: CT report.



A B

Figure 10: (A) Post Rx frontal view; (B) Post Rx profile view.

DISCUSSION

KCOT arises from the remnants of dental lamina. Incidence is about 5-11% of odontogenic lesions of jaw. Occurs commonly in the 2ND-4TH decade of life, with male predominance of 2:1. Ten percentage of KCOT occurs in association with nevoid basal cell carcinoma syndrome. Main site of occurrence is the angle of mandible (49%) along with the ramus and body posterior to first premolar. In maxilla site of occurrence in the 3rd molar region involving the antrum and anterior region.

Small lesions are diagnosed in routine radiological examination. Since the lesion extends in the medullary cavity along the path of least resistance, patient will be free of symptoms until the lesion reaches large size. As the lesion enlarges following signs and symptoms are noted: missing teeth, displaced teeth, dull sound on percussion, buccal expansion of bone, lingual or palatal expansion is rare, deflection of neurovascular bundle, associated tooth is usually vital. Acute infection presents with pain, swelling, pus discharge, neuropraxia causing labial paresthesia etc. Maxillary lesion due to close proximity to the anturm may cause distortion of the orbital floor and proptosis of the eye. Multiple KCOT is seen in association with NBCCS. High recurrence rate for the orthokeratinized variant. Rate of growth of KCOT is 2-14 mm per year with average around 7 mm/year, as explained by Forssel et al.[6] Nuclear morphometric variables like large number of basal cells and more H 3 thymidine in the basal and suprabasal cells suggest high mitotic value.

Radiologically KCOT may appear as a unilocular or multilocular lesion with smooth or scalloped and sclerotic margin with unequal growth. Buccal cortical expansion and resorption leading to perforation of cortex, whereas lingual cortical expansion is rare. Larger lesions cause resorption of cortical plate and involvement of inferior boarder of mandible, necessitating CT/CBCT for the radiological evaluation. Periapical cyst and dentigerous cyst are the main differential diagnosis due to relation with the tooth apex and impedance of eruption of related tooth. Pathogenesis of KCOT suggest origin from the odontogenic epithelium either from remnants of basal lamina or enamel organ and basal cells of overlying mucosa.

Keratocystic aspirate has certain characteristic features. It appears as thick, creamy, dirty white and viscoid suspension of keratin, with offensive smell and occasional appearance of pus. Examination of smear shows the presence of keratin cells. In electrophoresis low protein content and high albumin- globulin ratio is seen, with total protein less than 4 g/100 ml. Other features are cholesterol crystals, keratin squames, hyaluronic acid, rushton bodies, heparin/chondrotin sulphate etc. Kuusela et al had done immunofluorosence studies suggesting the presence of KCA [keratocystic antigen] and positive stains for P53, Ki-67, bcl-2 etc.[7]

OKC aspirate also contain lactoferrin, more Ig A, plasma cells and crystalline deposits of $CaPO_4$, hyroxyapatite, whitlockite, etc.

Basic histologic features of OKC was given by Pindborg and Hansen in 1963.[4] It includes thin or uniformly thick lining epithelium with no rete pegs, hyperchromatic pallisading cuboidal or columnar basal cell layer, thin spinous layer with direct transition from basal cells with intra cellular odema, para or orthokeratinized corrugated inner cystic surface, thin uninflammed cystic wall. Histologic diagnostic criteria was given by WHO in 2001. According to the criteria the lesion is characterized by a thin fibrous capsule and a lining of keratinized stratified squamous epithelium usually 5-8 cell layer in thickness and generally without rete pegs .Basal layer is pallisaded, polarized with cuboidal or low columnar cells budding into connective tissue. Spinous layer is polyhedral and shows acanthosis. Sub epithelial layer shows increased collagenase activity and decreased inflammatory infiltrate. There is loss of rete pegs with higher number of basal cells and low mean nuclear area of basal cells. Infolding of epithelium into the fibrous cystic wall results in satellite cyst. When inflamed the epithelium loses keratin, thickens and develop rete pegs. On enucleation thin lining may separate from the connective tissue. Thin capsule is free of inflammatory cells contain strands of epithelium that resemble dental lamina cell rest and daughter cyst. Cystic cavity contains desquamated keratin. Malignant transformation rarely occurs as squamous cell carcinoma.

Fibrous capsule of OKC is thin, widely separated by stroma rich in mucopolysacchride. It resemble mesenchymal connective tissue with mild infiltration of lymphocytes and monocytes. If intense inflammation is there then adjacent epithelium develop rete pegs, loses keratin and becomes ulcerative. Epithelial -connective tissue attachment is weak with infolding of epithelium into the fibrous cystic wall. Feature seen in the fibrous capsule are satellite cyst, epithelial rest or islands, proliferating dental lamina, nest of basaloid cells, budding of the cystic lining, ameloblastamatoid features, mucous metaplasia, hyaline bodies, cholesterol clefts, increased mast cells, some may show cartilaginous metaplasia and increased melanin pigmentation.

Ultrastructural study of OKC is done by scanning electron microscopy and transmission electron microscopy. In SEM orthokeratinized variant shows uniform flat surface with leafy squames of orthokeratin, whereas the para keratinized variant shows complex elevation and depression on the cell surface. Under TEM parakeratinized variant shows cytoplasmic inter-digitations, desmosomal junctions etc, orthokeratinized variant shows loose attachment between superficial shreds of orthokeratin and a compact layer of underlying dermis. Collagenase is responsible for the collagenolytic activity causing ready separation of OKC from the supporting tissue. There is an increase in tonofilaments from basal to superficial layers and cytoplasmic organells like mitochondria and golgi bodies have no significant changes. Cells of stratum spinosum has more amount of glycogen. Philipsen et al suggested a complex pattern of para keratinized OKC, having microplications on both upper and deep cell surface and microvilli between microplications[3]. Low magnification SEM of the orthokeratinized variant shows regular, flattened, polyhedral cells with distinct intercellular boundaries, round and ovoid central elevation corresponding to nuclei of cells. Under high magnification the characteristic feature is reticular network of intercommunicating micro ridges surrounding depressions giving a honey comb appearance to the entire surface and the deep surface have microvilli. Fenestrated capillaries and epithelial degeneration or thrombosis is seen and the platelets in thrombosed vessels release growth factor that promotes proliferation of OKC epithelium.

Aggressive nature of KCOT was first given by Pindborg and Hansen with 2.5-62% rate of recurrence mostly occurring in five to seven years projecting the need for long term follow up.[4] With NBCCS the rate of recurrence is 25-65%. Reason for recurrence are multiplicity, satellite cyst, thin and fragile lining epithelium with intrinsic growth potential forming epithelial islands and basal cell harmatomas that arise from the basal cells of oral mucosa. Borg et al have have given the relation between lingual plate perforation and recurrence.[8] Bramon in 1976 described three mechanism of recurrence; (1) incomplete removal, (2) new growth from satellite cyst and odontogenic remnants left after surgery, (3) development of new KCOT in adjacent areas interpreted as recurrence.[9] Mechanism of recurrence as suggested by Voorsmith et al, points to the lining epithelium left behind in the oral cavity after enucleation, daughter cyst or micro cyst in the wall of original cyst and epithelial offshoots of basal layer of oral epithelium for giving rise to new lesion.[10] Recent molecular evidence consider KCOT as a benign cystic neoplasm. Recurrence of peripheral KCOT was explained by Dayan et al.[11]

Recent genetical studies correlates the abnormal function or dysregulation of PTCH , a tumour suppressor gene as the etiology for the occurrence of KCOT in NBCCS and sporadic cases. PTCH and SMO forms a receptor complex that inhibit the growth signal transduction by binding with SHH [sonic hedgehog] ligand. So there is inhibition in growth signal transduction pathway. In PTCH dysfunction proliferation stimulating effect of SMO predominate. Genetically the pathogenesis of KCOT can be explained by 2-HIT mechanism. Allelic loss of 9q22 and dysregulation of tumor suppressor gene are the causes. First hit is the mutation of one allele that is newly inherited [no phenotype effect]. Second hit is the loss of other allele [loss of heterozygocity]. These two will cause the dysregulation of oncoprotien Cyclin D_1 and P 53.

Enlargement of KCOT is mainly assumed to be due to raised osmolarity in the cystic fluid and various mechanisms causing bone resorption. Protein content and the products of cell lysis increase the osmolarity of cystic fluid. Others believe in mural growth in the form of epithelial proliferation. Ahlkrs et al explained about the infolding of epithelial lining into the capsule and collagenolytic activity in the fibrous capsule causing bone resorption. Inflammatory infiltrate has a negligible role in the growth of KCOT, whereas glycos-aminoglycans like chondratin sulphate, heparin sulphate and hyaluronic acid increases osmotic and hydrostatic pressure. IL-1, TNF, MMP, tenascin, fibronectin, collagen IV, myofibroblast and parathyroid hormone related protein have a role in collagenolytic activity leading to cyst growth. DNA aneuploidy of the lining epithelium can be assessed to predict the change of OKC to squamous cell carcinoma. Immunofluorosence staining of the dysplastic epithelium shows the loss of blood group antigen A and B.

Various management options of KCOT have been discussed in literature. There are reports of carcinoma arising from KCOT and recurrence of KCOT in bone grafts. Surgical management options are enucleation along with curettage, peripheral ostectomy, cryotherapy, chemical cauterization, marsupialisation and decompression, resection with or without continuity defect are some of the treatment options. Marsupiali-sation denies the tumour character. Ghali et al in 2003 suggested that as with any other odontogenic lesions take a proper history, physical examination and radiological evaluation, for larger lesions CT/CBCT is desirable.[12] Differential diagnosis is done by biopsy either incisional or excisional preceded by aspiration. Consider size, location and behavior of lesion prior to biopsy. For larger lesions take biopsy from multiple sites. For proper treatment evaluate the size and extend of lesion, age, location of lesion, proximity to vital structures, radiological evaluation, perforation and soft tissue involvement, in case of recurrence time since previous treatment.

In 2002 Nakimra et al suggested marsupialization and decompression, as the procedure releases the pressure in the cyst cavity.[13] It enhances the filling of defect by new bone there by decreasing the size of cyst, saving the roots, antrum and inferior alveolar nerve. The lesion seems to be less aggressive during the course of marsupialization. The need for second surgery and chance of recurrence are the disadvantages. Bramely had given various treatment options in 1971, based on the size of the lesion.[14] For small cystic lesions with regular spherical outline enucleation via intraoral approach is ideal. Enucleation via extraoral approach was suggested for larger and less accessible lesions with regular spherical outline. Unilocular lesion with scalloped or locculated periphery and small multilocular lesions need resection maintaining the continuity of inferior or posterior boarders via a combined intra and extraoral approach. If the cystic lesion is adherent to the overlying mucosa or muscle, it is excised along with marginal resection and the defect is either closed primarily or left for secondary healing. Along with it hyroxyapatite, autogenous bone graft, corticocancellous chips, allogenous bone powder, chips and blocks can also be used. Large multilocular lesions with cortical perforation needs resection with primary or secondary reconstruction using reconstruction plates and bone grafts.

Stoelinga has proposed enucleation and chemical cauterization using carnoy's solution along with strict follow up to look for recurrence.[15] Voorsmit has shown the mean depth of bone penetration of Carnoy's solution is about 1.54 mm after 5 minutes. Most accepted treatment options are enucleation and cryosurgery, enucleation and chemical cauterization and radical excision. Future treatment aims at molecular level. Cyclopamine is a plant based alkaloid that inhibit the SHH pathway activity. Zhang et al in 2006 have told about the intralesional injection of SMO protein antagonist.[18]

CONCLUSION

Since the time of explanation there has been confusion about the cystic or tumour nature of KCOT. Initially described as odontogenic keratocyst was later reclassified as keratocystic odontogenic tumour by WHO in 2005, now being reclassified as cyst in new WHO classification 2017 as that hypothesis like clonality is considered insufficient. But there is no doubt regarding the aggressive nature of the lesion. Depending upon the size of the lesion different treatment modalities are selected as described above. For extensive lesions as described in this article the ideal treatment option is resection and reconstruction, because of the compromising medical condition we had to manage the lesion conservatively.

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Conflict of interest: None declared
Ethical approval: Not required

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