Reconstruction of mandibular defect using patient specific peek implant – An approach to aesthetic reconstruction

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ABSTRACT
Odontogenic keratocyst (OKC) is a developmental cyst arising from the enamel organ or the dental lamina. It commonly affects males in their 2nd - 4th decade and is associated with a high recurrence rate of 2.5 – 62 %. The treatment of OKC depends on the size
of the lesion, with conservative approaches like enucleation with or without adjuvant therapies, marsupialisation indicated in smaller lesions. For extensive lesions, however, en bloc resection is the choice of treatment. The reconstruction of continuity defect formed following en bloc resection has always remained a question for the surgeons, with options being free tissue transfer, titanium implants and patient specific implants. Polyetheretherketone (PEEK) is one such biocompatible alloplastic material recently introduced for the reconstruction of such discontinuity defects in the maxillofacial skeleton. We present a case of reconstruction of discontinuity defect following en bloc reception of OKC of right mandibular body and ramal region using PEEK.

**Keywords:** Odontogenic Keratocyst; Mandibular reconstruction; Patient specific implant; DICOM; PEEK.

1. INTRODUCTION

Odontogenic keratocyst (OKC) is an enigma to the surgeon owing to its behaviour. Since the time it was first described by Shear, it has been of interest to the surgeon and pathologists alike (M, n.d.). The aggressive behaviour lead to its classification of tumour in 2004, however with new found evidence it was reclassified as a cyst in 2014. OKC is associated with a high recurrence rate (2.5 - 62 %) which is theorized to be due to the fragility of cystic lining and the presence of daughter cells (Shear, 1994) (Brannon, 1976) (Forssell et al., 1988). Various treatment options have been suggested in the literature, such as enucleation, enucleation with chemical cauterization, enucleation with peripheral ostectomy for small lesion. For extensive lesions, aggressive approaches like en bloc resection are done. Newer treatment modalities like Cryotherapy and 5-Flourouracil have also been researched. The reconstruction of the defect formed after such aggressive treatments have always raised a question to the surgeon. With advancement, the free tissue transfer and patient specific implants using Polyetheretherketone (PEEK) polymer have gained popularity (Rudman et al., 2011). We present a case of reconstruction of such a mandibular defect created following resection of the cystic lesion using patient specific PEEK Implant.

2. CASE REPORT

A 28 year old man reported to Oral Surgery OPD, Sharad Pawar Dental College & Hospital, Sawangi, Wardha, with a chief complaint of pain over right side of face. There was history of associated swelling which was initially small in size and rapidly increased to the presenting size of 8 x 4cms in greatest dimensions (Fig 1a). The patient had a difficulty in mastication and there was history of pus discharge from the lower right back region since 15 days, which prompted the patient to seek treatment. On palpation, the inspector findings were confirmed and an egg shell cracking with frank pus discharge was noted. The lesion was seen extending from distal of 44 to the ascending border of ramus. A wide bore 16-gauge needle was used to aspirate the cystic content which was then sent for cyt pathological examination which was suggestive of cholesterol clefts. An OPG was done, which showed a well-defined, multi-locular lesion, displacing the roots of the mandibular molars of the right side. An impacted molar tooth was seen in association with the cystic lesion (Fig 1b). A CT scan of Head was done which was suggestive of a lobulated, lytic lesion expansile lesion of approximate size 7 x 4.6 x 3.5 cms in the right side of mandible involving the body, ramus and coronoid process, with scalloping borders and cortical thinning of the mandibular cortices (Fig. 1c) .Based on the clinical and radiographic findings, a provisional diagnosis of cystic lesion was developed with differential diagnosis formulated of Odontogenic Keratocyst, Ameloblastoma, Dentigerous cyst, and aneurysmal bone cyst.

Patient was then subjected to incisional bone biopsy to establish a final histopathological diagnosis. The biopsy specimen showed a cystic space lined by nonkeratinized stratified squamous epithelium and surrounding connective tissue capsule. The epithelium appeared corrugated with 4-5-layer cell thickness. Palisaded tall columnar epithelial cells with hyperchromatic nuclei were the component of basal cell layer. The epithelium & connective tissue interface was flat and there was incomplete formation of the rete ridges which were the characteristic features of an Odontogenic keratocyst Considering the histopathological diagnosis and the extent of bone destruction, conservative surgical treatments would have resulted in thin lower border predisposing to pathological fracture, and hence an en-bloc resection of the lesion under general anesthesia was planned. As the patient was of young age and the resection would have lead to the formation of a continuity defect, the reconstruction was then considered using either a free tissue transfer or a custom made implant using PEEK. The patient was presented with both the options and after consideration of the resulting donor site morbidity from free tissue transfer, he opted for a custom-made PEEK implant.

For the fabrication of a PEEK implant, DICOM (Digital Imaging and Communications in Medicine) were sent to the PSI (Patient Specific Implant) manufacturer, who then created 3-D images using CAD/CAM software (fig 1d). A virtual surgery defining the limits of the resection was performed and a mirror image of the contralateral mandible was superimposed on the defect. This was then used to fabricate a PSI using Steriolithography. A two piece PEEK implant was fabricated which was sterilised prior to its use (Fig 2b).
Under general anaesthesia, en-bloc resection of the lesion from 44 to disarticulation of condyle of was done and the PEEK implant was used for reconstruction of the defect. The implant was secured to the bone using titanium implants and screws and closure was done (Fig 2a, 2b, 2c, 2d). Formalin fixed specimen was subjected to Histopathological examination. Post operatively, the patient was advised soft diet for a period of 3 months. The patient is on a follow up of 2 years and no complications regarding the implant or recurrence of the disease has been noted (Fig 3a, 3b).

**Figure 1** 1a – Pre-operative frontal profile, 1b – Pre - operative OPG showing destruction of the mandibular ramus and body on right side, 1c – Pre- operative CT Head (Axial Section) showing destruction of the labial and lingual cortices in the right posterior region, 1d – Steriolithographic model showing the thinning of labial and lingual cortices

**Figure 2** 2a – Intraoperative picture showing the lesion over right mandibular ramus and body region, 2b – Resected specimen and Patient specific implant fabricated out of PEEK, 2c – Resected specimen Radiograph, 2d – Patient Specific implant fixed over the defect created following resection
3. DISCUSSION

The term Odontogenic Keratocyst (OKC) was coined by Philipsen in 1956. The nature and the histological variants were fully described in 1963 by Pindborg and Hansen (Anthony Pogrel and Jordan, 2004). It is a developmental cyst derived from the enamel organ or the dental lamina (Bataineh and Al Qudah, 1998) (B Motwani et al., 2011). It has been one of the most controversial pathologies since its initial description. The WHO in 2004 designated it as a Keratocystic Odontogeic Tumor (KCOT) and defined it as “A benign uni- or multi- cystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinised stratified squamous epithelium and potential for aggressive, infiltrative behaviour”. However, in 2017, The WHO reclassified this tumour as a cyst on the basis of re-evaluation of the clinical, radiological and biological features (Slusarenko da Silva et al., 2019).

OKC accounts for 2–11 % of total cysts of the jaw. These may occur in solitary or as a multiple as a part of the syndrome – Nevoid Basal Cell Carcinoma (NBCCS). Multiple OKCs without any associated features are considered to be an incomplete form of the NBCCS (Mendes et al., 2010). OKCs affect males more commonly than females and are usually diagnosed in the 2nd–4th decade of life. These are more commonly located in the mandible than the maxilla. There is a higher prevalence in the body, angle and ramus of the mandible than in the anterior region. Unlike ameloblastoma, there is only anteroposterior expansion of the cortical bone (B Motwani et al., 2011; Mendes et al., 2010). Clinically these present as painful swelling and in cases of a superimposed infection these are often associated with draining sinus. A similar finding was noted in the above mentioned case with the patient complaining of a slow growing swelling over two years.

Radiographically these may appear as small, ovoid or round, radiolucent lesions often with multi-lobulated, scalloped and defined margins. These are difficult to distinguish from other lesions (M, n.d.). In about 25–40% of the cases an impacted tooth is seen in association with the lesion, hence making it difficult to differentiate from a dentigerous cyst (Marcelis et al., n.d.). These lesions often lead to displacement of the tooth roots in the area involved and/or the inferior alveolar nerve bundle. Histologically, OKCs include a thin epithelial lining, consisting fewer than 6 cell layers arranged in a corrugated or wavy pattern resembling a picket-fence or a tomb stone appearance. Daughter cells which are islands of epithelium, along with thin, irregular bungles of collagen are present underlying the epithelium in the connective tissue (Anthony Pogrel and Jordan, 2004)(M, n.d.).

OKCs have a high recurrence rate, with 2.5 – 62.5 % cases recurring in the first 5 years after the first surgery (Shear, 1994; Brannon, 1976; Forssell et al., 1988). Brannon (Brannon, 1976) suggested that the remnants of epithelium or cell rests of cystic tissues were the most tenable factors for recurrence. Vedtofte and Praetorius observed that the overlying mucosa contained remnants of the cystic lining and when failed to remove, lead to recurrence (Vedtofte and Praetorius, 1979; Emerson et al., 1972). Given the nature of this lesion, various surgical modalities have been suggested in the literature ranging from simple conservative measures...
such as Marsupialisation to aggressive modalities such as resection. The recurrence of the lesions with marsupialization has been reported to be ranging from 9 – 60% (Marker et al., 1996; Forsell et al., 1988). Larger lesions are however best treated by resection with or without continuity defects as these are said to have the lowest recurrence rated of all surgical modalities i.e. 0% (Morgan et al., 2005; Chuong et al., 1982).

Reconstructive surgery for complex maxillofacial defects remains a challenge for even the most experienced surgeon (Rudman et al., 2011). Free graft transfer, reconstruction using Titanium implants and PEEK has been described in the literature. Free tissue transfers fulfil both the aesthetic and functional requirements at the cost of a second surgical site with additional strain on the patient’s physical, mental and social aspects. Reconstruction using titanium plates however fails to fulfil both these requirements (Rudman et al., 2011; Scolozzi, 2012). Autologous bone grafts are a good reconstructive option for small defects (<3 – 4cm) with non-complex shapes. Larger defects, however, require free tissue transfer, causing increased donor site morbidity, insufficient donor material – related problems and bone resorption. The use of alloplastic material such as MEDPOR, Titanium, PEEK helps over these issues.

PEEK is a penultimate polyaromatic linear polymer. It was first developed in 1978 and used in aircrafts and turbine blades. The use of PEEK in surgical field was started by the late 1990s, where it was used to replace metal implants in spine and orthopaedic surgery (Honigmann et al., 2018). It is mechanically strong, non-allergic and non-magnetic and does not undergo exothermic reactions. It has good biocompatibility and is radiolucent. The patient specific implants fabricated using PEEK can be easily trimmed intra-operatively and provide permanent long-term results. PEEK as it does not undergo exothermic reactions, it can withstand repeated autoclave sterilizations. The density is 1.30g/cm2 and the elasticity of PEEK (GPa – 8.3, MPa – 139) is comparable to that of cortical bone (GPa – 17.7, MPa – 133) (Scolozzi, 2012; Järvinen et al., 2019) (“Advanced Biomaterials,” n.d.). The infection rates with the use of PEEK implants was noted to be 8.3%, which was comparable with other alloplastic material (Järvinen et al., 2019). The disadvantage of PEEK is the high cost of manufacturing. Considering the age of the patient and the extent of resection, we opted for a patient specific implant fabricated using PEEK which showed no evidence of post-operative complications.

4. CONCLUSION

PEEK (Polyether ether ketone) is a biocompatible, non-magnetic, non-allergic material with its density and elasticity comparable to cortical bone. Therefore, custom made implant of PEEK material is a good option for reconstruction of discontinuity defects of the maxillofacial skeleton. It helps in restoration of the function with minimal disturbances of the aesthetics and prevents donor site morbidity.

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Conflict of interest
There are no conflicts of interests.

REFERENCE


