Review Article

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Osteo-odonto-keratoplasty: A Review

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Abstract

Objectives: To discuss the historical background of osteo-odonto-keratoprosthesis (OOKP), patients' selection criteria, preoperative assessment of the patients, OOKP technique in detail and its modifications along with the complications involved.

Data Sources: Data for this narrative review were identified by searches of MEDLINE, Current Contents, PubMed, and references from relevant articles using the search term 'osteo-odonto-keratoprosthesis', 'canine', 'eye', 'blindness' and 'Sjogren's syndrome'. Articles published in English medical literature containing studies and case reports on osteo-odonto-keratoprosthesis are included in the present review.

Conclusions: OOKP technique is a complex artificial corneal (keratoprosthetic) transplantation procedure in which an autologus dental root-bone lamina complex and buccal mucosal graft are used to mount a poly-methylmethacrylate optical cylinder, as an artificial cornea. OOKP surgery is usually the last resort for end-stage corneal blindness, where other treatments have failed, or will not be successful.

Introduction

Corneal opacification is the second commonest cause of world blindness affecting an estimated 10 million people [1]. The visual rehabilitation of such patients is a challenging task. Not only the conventional corneal graft but a wide variety of materials have been tried earlier as artificial corneal implants, including metals, ceramics, glass, plastics and biological tissue, for the anchoring skirts or plates or haptic [2,3]. For the optic, only medical grade polymethylmethacrylate (PMMA) has been used since the last 50 years [4]. The three most commonly used keratoprosthesis (KPs) are the Dohlman collar button type [4], the Osteo-Odonto keratoprosthesis (OOKP) by Strampelli [5], Falcinelli in Italy and Liu in UK [6] and the Pintucci biointegrable KP by Pintucci in Italy [7,8].

The original OOKP technique was pioneered by the late Italian surgeon Benedetto Strampelli MD in 1963 [5,9]. OOKP is an eterotopic autograft and the term stands for osteo (bone)-odonto (tooth)-kerato (cornea) prosthesis (artificial device) [10] and thus the technique is also known as "a tooth for an eye" technique [11,12]. Recent innovative changes to the original Strampelli technique have further reduced the incidence of failures to less than 10% of cases [13].

OOKP surgery is usually the last resort for severe, end-stage corneal blindness, where other treatments such as corneal grafts [14], stem cell and amniotic membrane transplantations [15,16] and other forms of ocular surface reconstruction surgery have failed, or will not be successful [9].

Historical background

The first artificial corneal implant was placed in a human eye by Nussbaum, 150 years ago [3]. Strampelli (1963) [5] had noted that gutta-percha will remain in the root canal of a tooth indefinitely, but will be rejected if implanted into soft tissues. So it would seem probable that if a plastic acrylic implant could be held in a piece of the patient's tooth and bone, and the whole placed in a corneal envelope, the tooth and bone would form an autograft picture-frame for the acrylic, and so prevent its extrusion [17]. The technique was largely abandoned in the 1970s due to poor results. An Italian surgeon, Dr. Gian Carlo Falcinelli made numerous improvements in the 1980s. Over the past few years,

the procedure has undergone further refinements, which are referred to as the Rome-Vienna Protocol [18].

Patient selection criteria

Patients with bilateral corneal blindness resulting from severe end-stage Stevens-Johnson syndrome [5,6,9], Lyell syndrome [19], ocular cicatricial pemphigoid, chemical or thermal burns, end stage trachoma, severe keratitis, dry eyes, multiple failed grafts and graft-versus-host disease and consequences of perforating injuries may be considered for OOKP surgery [20]. The other causes of dry eye are ectodermal dysplasia, ionizing radiation damage, cicatrizing conjunctivitis from topical medication, congenital trigeminal nerve hypoplasia, linear IgA disease and nutritional deficiency [20,21]. Though Goosen et al. [22] and Chammartin et al. [23] have reported cases of bilateral OOKPs, the vision of only one eye should be corrected. The better eye, with poor vision is rehabilitated [21]. However patients who are satisfied with their level of vision, children under the age of 17, eyes that have no perception of light, evidence of phthisis, advanced glaucoma or irreparable retinal detachment should be excluded [21].

Pre operative assessment

Each patient should undergo detailed evaluation by a team of ocular and oral surgeons and radiologists who form a surgical team. This evaluation should include the detailed history and aetiology of loss of vision and a comprehensive ocular and oral examination.

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Ophthalmic assessment [21]

Preoperative examination involves determining an intact and functioning retina and optic nerve. The degree of dry eye is noted, although a severe dry eye is not a contraindication unlike other forms of KPs. The conjunctiva and cornea are examined and evidence of stem cell failure, metaplasia, or dysplasia is noted. Thinning of the cornea and evidence of previous corneal perforation, iris adhesion, and degree of vascularization are also taken into consideration.

Oral assessment

The oral assessment must take into account both the buccal mucosal graft donor site and a selection of an appropriate tooth to form a dentine/bone lamina. Buccal mucous membrane can be classified as being normal, scarred or diseased [9]. Some diseases affecting buccal mucous membrane are recurrent aphthous ulceration, leukoplakia, lichen planus and oral submucous fibrosis. Detrimental habits like smoking and betel nut chewing should be discontinued as it will compromise tissue quality.

The vitality of the teeth to be used with caries or restorations should be tested using an electropulp tester. Panoramic and periapical radiographs are taken; to estimate the length of the root in bone. The healthiest and best-positioned tooth with best shape and size and good covering of alveolar bone is selected. After evaluation, patients are graded on the basis of their dental condition as being suitable for OOKP, at risk of complication or failure, or unsuitable for OOKP [9]. Oral hygiene and periodontal bone loss should be assessed.

The most suitable tooth for modified preparation of the osteodental lamina is the maxillary canine[9] because it has the longest and largest root with the greatest quantity of alveolar bone [19]. Contraindications for using a particular tooth include non-vitality, previous root canal therapy, inadequate bone investing the root, peri-radicular pathology and close proximity of adjacent teeth [9]. Other single-rooted teeth can be used in the absence of a canine [19]. The choice of upper or lower canine depends on the proximity of the maxillary sinus and the mental foramen. In lower canine harvesting, buccal plate is occasionally thin and the lingual muco-periosteum is difficult to preserve. There is a risk of antrum perforation in case of upper canine [21].

In the majority of patients, an autologous osteodental lamina is used. Occasionally when there is no suitable tooth available or in case of edentulous patients an allograft is considered [17]. A tooth from a first-degree relative with the highest number of compatible HLA antigen sites can be used [19] but long term results studied by Liu et al. [20] showed that HLA-matched allografts suffered a higher rate of laminar resorption. Nail (onycho-keratoprosthesis) can be used instead of a tooth but it has the disadvantage that it is dead tissue, but if it is taken from the nail root, it is liable to grow in the eye. Casay et al. [17] have used a chondro-keratoprosthesis, a piece of cartilage removed from the 8th costal cartilage and have reported their best results to date. Some surgeons found tibial bone ie temprano- keratoprosthesis (TKP) as a better option [16,24].

Viitala R et al. [25] have tried synthetic analogue (bioceramics) instead of OOKP and found that at normal physiologic pH the degradation of bioceramics was equivalent to tooth and bone. However at pH of 6.5 to 5 associated with infectious and inflamed tissue, the degradation rate of bioceramics was much higher when compared to bone and teeth.

Psychological assesment [21]

Patients have to understand that they may require multiple procedures and that there is a significant risk of serious complications including loss of the eye. The patient must commit to life-long follow-up, and not have unreasonable expectations of outcome and cosmesis.

Procedure: The technique involves three procedures stages [9]

Stage 1a: Tooth ostectomy, preparation of the osteo-odonto lamina, cementation of the optic cylinder in the lamina, and submuscular implantation in the contralateral infraorbital area

Stage 1b: Removal of the ocular surface, harvesting of a full-thickness buccal mucosal graft, and coverage of the ocular surface with the mucosal graft

Stage 2: Retrieval of the keratoprosthesis, lifting of the buccal mucosal graft and implantation of the keratoprosthesis into the eye

Stage 1: The procedure involves the preparation of an osteo-odonto-lamina tooth by harvesting a single rooted tooth [9,21]. If the surgeon does not find the lamina's surface large enough to allow the insertion of an optical cylinder, two teeth can be extracted to prepare two laminae, which are then glued together with acrylic resin to prepare a larger surface [19]. The resulting alveolar defect is covered with adjacent mucosa. In Japan, surgeons are using artificial mucous membrane graft to cover the defect to accelerate wound healing [26]. Casey [17] used half of the root after apicectomy and filled the remaining root part.

Prepare a rectangular lamina with the central part comprising two third of the lamina and a thin cemental rim. The peripheral one third part is of alveolar bone. The cementum and alveolar bone are attached to each other by intact periodontal ligament, as seen in the normal tooth histological structure [19]. Stoiber J et al. [27] studied OOKP histologically and found that the preservation of alveolar-dental ligament seems to be essential for the maintenance of the integrity of the OOKP. Ideal lamina should be of a size measuring 12 mm x 6 mm x 3 mm [9]. A hole of an average diameter of 3.70 mm (range, 3.3-4.0 mm) is prepared leaving an edge of dentine of 1 to 1.5 mm [19].

A PMMA optical cylinder is inserted into the hole and cemented to the osteodental lamina by means of biocompatible acrylic resin or PMMA cement [9,19]. Some operators discourage the use of dental cement as it may act as an irritant [17]. The optical characteristics of the PMMA optical cylinder are, mean intraocular diameter, 4.1 mm (range, 3.6-4.6 mm); mean extraocular diameter, 3.65 mm (range, 3.3-4.0 mm); mean length, 7.75 mm (range, 7.25-8.25 mm); mean radius of the convex extraocular surface, 16 mm; mean radius of the convex intraocular surface, 6.5 mm; refractive index, 1.49; and equivalent power, 50.8 diopters [28,29].

Hull et al. [30] have concluded that it is possible to increase the theoretical maximum visual field through these cylinders. The optical cylinder with wider diameter offers a wider visual field for the patient [17].

The prepared complex is inserted beneath the orbicularis oculi muscle just below the contralateral lower orbital rim, for a period of two and half to four months to allow for osteo-odontal tissue recovery and formation of a fibrovascular covering over the lamina [9,19]. (Figure 1)

A buccal mucous membrane graft which is preferred to cover the ocular surface as it contains stem cells. It has proliferating capability and is adapted to high bacterial load [21]. The buccal graft of full thickness mucosa without including muscle and large enough (approximately
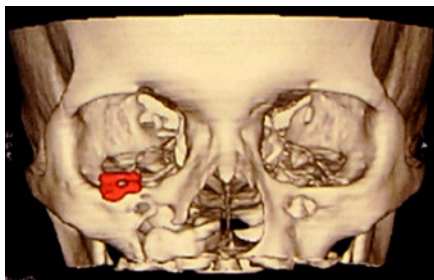
Figure 1: CT scan showing the prepared lamina complex just below the contralateral lower orbital rim. It is kept for a period of two and half to four months to allow the tissue to recover and formation of a fibrovascular covering over the lamina.

3cm in diameter) to extend from medial to lateral canthi and from upper to lower lid fornices is harvested taking care to avoid parotid duct orifice and lower buccal sulcus (mental nerve injury) [9]. It will be vascularized by the time of Stage 2 surgery and will thus provide the blood supply to the bone part of the OOKP lamina. The graft is soaked in an antibiotic solution like cefuroxime until required. An oval piece of buccal mucosa is prepared [21].

If the eye is very dry or there is a risk of the mucous membrane graft not taking, it may be better to perform Stage I surgery in two steps. The mucous membrane graft to the eye is done first (stage IA), and it is only when the graft has been shown to be well established before the patient is readmitted for tooth harvesting and preparing an OOKP lamina (stage IB) [21].

Stage 2: Stage 2 surgery is carried out two to four months after Stage 1 [9,31]. The time interval helps graft and lamina to heal and vascularize properly. The soft tissue invests into the pores of the lamina and the lamina recovers from thermal damage and infection if present.

It is retrieved and inspected for adequate size. The fibrovascular capsule is trimmed away from the optical surfaces of the PMMA optical cylinder.

The buccal mucosal graft is reflected superiorly and cornea is exposed [21]. Corneal trephination (5 mm-diameter) is done to create a central opening in the eye. The iris is completely removed. The keratoprosthesis is then placed into the opening, which serves as a strong biological skirt to secure suturing of the keratoprosthesis in position [9]. The buccal mucosal graft is sutured over the implant and trephined in the centre to allow the anterior aspect of the optical cylinder to protrude [9,21]. (Figure 2 and 3)

One month after surgery, a cosmetic prosthesis covering the external ocular surface can be given [19]. Main outcome measures to be considered are visual acuity, field of vision, anatomical integrity and stability and complications related or unrelated to the OOKP technique [31].

Donald et al. [31] performed OOKP surgery on 15 patients, with a mean follow-up of 19.1 months. Eleven patients (73.3%) attained a stable best spectacle-corrected visual acuity of at least 20/40 or better, whereas 9 (60%) attained stable 20/20 vision. Four patients achieved a visual potential ranging from 20/100 to counting fingers vision.

The follow up of patients for 10 years by Michael et al. [24] yielded following statistics:

- 10-year anatomical survival: 66% for OOKP and 47% for osteo-keratoprosthesis,

- 2-year functional survival: 63% for OOKP and 49% for osteo-keratoprosthesis ,

- 10-year functional survival: 38% for OOKP and 17% for osteo-keratoprosthesis, with functional survival defined as best corrected visual acuity above 0.05.

Fong et al. [32] used electron beam tomography (EBT) in imaging of OOKP to identify early bone and dentine loss. They concluded that it is important to monitor regularly the dimensions and stability of the OOKP lamina as it will help detect cases that are at risk of extrusion of the optical cylinder and consequent endophthalmitis. They also found out that EBT have excellent resolution and speed and recommend regular scanning of the lamina in all patients.

Hille et al. [33] have implanted a total of 35KPs, 29 with biologoic haptic (25 OOKP and 4 TKPs) and 6 KPs with biocompatible haptic. There was no significant difference between the various types of KPs concerning the best postoperative visual acuity. During long term follow up, only 1 of the KPs with biologic haptic (TKP) was lost compared with 4 out of 6 KPs with biocompatible haptic (P<0.0001). Thus they concluded that OOKP leads to the best results in the long-term follow up.

Percorella I et al. [34] biopsied the junction between the osteodental acrylic lamina and surrounding modified oral mucosa in 7 patients and were examined by light microscopy. Six of the 7 corresponded microscopically to conjunctiva. Typical oral mucosa could still be observed overlying the osteodental acrylic lamina. Thus they concluded that the production of local regulatory factors is a possible explanation for the survival of oral mucosa over the osteodental acrylic lamina, whereas their absence in distant areas may have induced the oral mucosa to transdifferentiate into a conjunctival-type lining. Alternatively, conjunctival regrowth from forniceal stem cells should be taken into consideration.
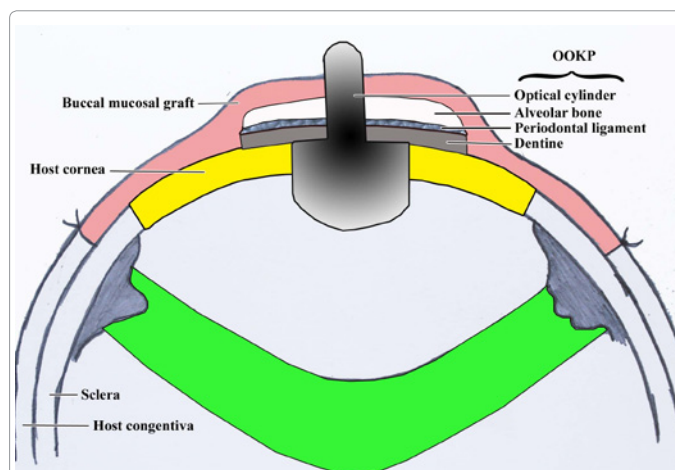

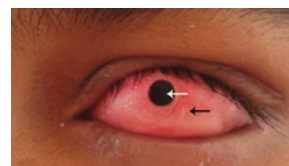**Figure 2:** Schematic diagram showing OOKP and its placement in the eye.

Figure 3: Eye with OOKP showing optical cylinder (white arrow) and surrounding buccal mucosal graft (black arrow).

Percorella I et al. [35] demonstrated elastic and precursor fibers and distribution of collagen types I to VI in bone and dental tissue which were exposed to the ocular surface.

Ricci R et al. [36] showed that preservation of the alveolar-dental ligament plays a definitive role in the maintenance of the prosthesis. If this tissue undergoes necrosis, the implanted material is eventually lost. However, when no such event occurs the OOK is well preserved and well tolerated even 20 years after implantation.

Complications

Ocular complications

1. Forniceal and tarsal conjunctival cicatrization
2. Ocular surface inflammation
3. Continuation of primary disease process resulting in shallow fornices, upper or lower lid cicatricial entropion and wide palpebral apertures
4. Buccal mucous membrane thinning, ulceration or necrosis
5. Globe perforation
6. Lamina infection: Caiazza et al. reported early and recurrent bacterial infections (Staphylococcus epidermidis) which resulted in destruction of lamina
7. Lamina resorption: Study by Liu et al. showed that the main factor resulting in anatomical failure was OOKP lamina resorption
8. Vitreous haemorrhage
9. Choroidal and retinal detachment
10. Glaucoma or macular disease : reported incidence is up to 75% [16]
11.. Fistula formation
12. Extrusion of the optical cylinder: Results of the Strampelli's OOKP-technique show good long-term results with 2% loss of prosthesis over 27 years [16]. According to Falcinelli et al. 18 years after surgery, the probability of retaining an intact OOKP was 85% [17]. A significant risk of 19% has been estimated by Liu et al. [22]
13. Retro implant membrane formation
14. Mild inferior optic tilt
15. Increased intra ocular pressure
16. MRSA colonization
17. Periostitis of the lamina

Oral complications

1. Trismus resulting from submucosal scar formation
2. Buccal mucous membrane infection

Figure 4: Oro-antral fistula as a complication of OOKP procedure.

3. Fracture of tooth will render it useless as a lamina for the optical cylinder
4. Small part of the tooth if left behind in the bed will act as a nidus for infection
5. Once the tooth is extracted it will result in cosmetic and functional problems like lisping while speaking and extrusion of liquids while eating
6. Palatal or lingual bone fracture
7. Exposure of roots of adjacent teeth due to poor healing: Damage can be avoided by using sharp blades and careful technique
8. Oroantral perforation leading to fistula formation: The damage to the maxillary sinus can be avoided by not making the apical cut too high but if the root of the tooth is very close to the maxillary sinus, this complication is unavoidable. (Figure 4)
9. Oronasal perforation
10. Damage to mental nerve leading to lip paresthesia in case of mandibular canines

Systemic complications

1. Complications related to local and general anesthesia [38]
2. Immunosupression because of corticosteroid administration

Other complications

Other complications not directly related to device insertion include

1. Retinal detachment related to silicone oil removal
2. Endophthalmitis related to endoscopic cyclophotocoagulation

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