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British Journal of Oral and Maxillofacial Surgery 57 (2019) 861–865

BRITISH Journal of Oral and Maxillofacial Surgery

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Contribution to oral and maxillary surgery and surgical technique of osteo-odontokeratoprosthesis in Japan

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Received 8 October 2018; accepted 1 July 2019
Available online 2 August 2019

Abstract

Osteo-odontokeratoprosthesis (OOKP) is a technique invented by Strampelli in 1963, in which the patient's own tooth root is used to support an optical cylinder. It uses an autologous tooth-bone-periodontal complex to mount an optical cylinder, which is stabilised by overlying autologous buccal mucosa. OOKP involves two, staged procedures done by ophthalmologists and oral surgeons, and the main contribution from the oral surgeon is during the first stage. To date we have done nine first-stage, and completed eight second-stage, OOKP operations in Japan with a mean follow-up of eight years and 11 months by modifying the original method of the oral surgery. All OOKP procedures were unilateral, and canines were selected as the donor teeth. Patients developed ocular blindness as a result of Stevens-Johnson syndrome, ocular cicatricial pemphigoid, and chemical and thermal burns to the cornea and ocular surface. All eight patients who completed the second stage have been stable, and there have been no major perioperative or postoperative oral complications. The patients' visual acuities were stable with no serious complications. Here we report the technical details of the oral contribution to OOKP.

Keywords: OOKP; role of the oral surgeon; interdisciplinary management by ophthalmologists and oral surgeons

Introduction

Osteo-odontokeratoprosthesis (OOKP) is one of the methods used to reconstruct the cornea with an artificial cornea, which involves an autograft that replaces the cornea with a polymethyl methacrylate (PMMA) cylinder mounted on the tooth-bone-periodontal complex.[1] This treatment was first described and documented in 1963 by Strampelli, and then later modified by Falcinelli around 10 years later.[2,3] OOKP is currently done in only a few areas worldwide: India, Singapore, the UK, the US, and Japan.[4–7] Several types of keratoprostheses have been tried in the past to restore sight in Stevens–Johnson syndrome or ocular cicatricial pemphigoid, but the only one to achieve clinical, long-term satisfaction has been the OOKP. It is a useful treatment for not only for Stevens-Johnson syndrome or ocular cicatricial pemphigoid, but also for multiple failed grafts, thermal burns, and chemical burns, which are not amenable to penetrating keratoplasty.

It is a two-stage operation, and to date we have done nine first-stage OOKP operations in Japan.[8] Here we report on the clinical process and contributions of oral and maxillofacial surgery to OOKP.

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https://doi.org/10.1016/j.bjoms.2019.07.001

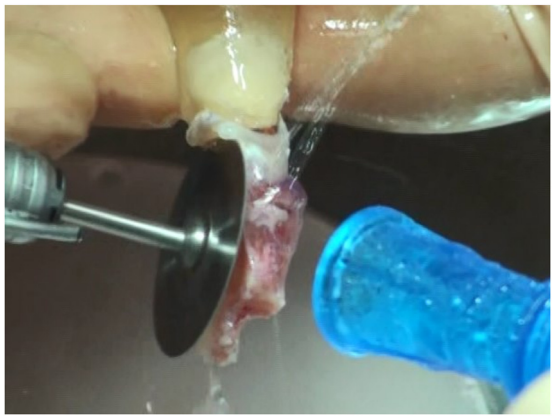

Fig. 1. The gingival mucosa was cut, and the tooth-bone-periodontal complex harvested. After the tooth-bone-periodontal complex had been fixed on the tooth-fixing device, the complex was shaved into the lamina to a thickness of 4 mm using a diamond disc.

Fig. 2. During fixation of the optical cylinder, cylindrical rubber was used to fix the end of the optical cylinder to make it technically usable.

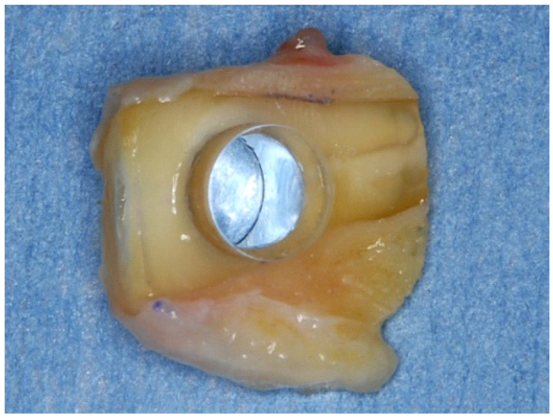

Fig. 3. Photograph of the harvested lamina of the tooth-bone-periodontal complex with the mounted optical cylinder.

Patients and methods

Stage 1

The first stage includes two procedures: first, the tooth-bone-periodontal lamina is prepared and placed into the submuscular pocket under the orbicularis oculi and, secondly, the buccal mucosa is harvested and sutured to the sclera bounded by the insertion sites of the rectus muscles.

During the first stage, the patient's tooth-bone-periodontal complex was harvested and shaped into a thin rectangular lamina form. Selection of the donor tooth was carefully considered taking into account adequate size, shape, healthy pulp without dental caries, single-rooted tooth, and a periodontal complex with healthy surrounding alveolar bone. We assessed the suitability of the tooth using clinical and radiological examination by panoramic radiography and computed tomography (CT) at initial presentation, and the canine was selected in all patients. Our criteria for CT take into account a tooth-bone width of 7.5 mm (a tooth-bone lesion of at least 2.0 mm should surround the 3.5 mm optical cylinder), healthy pulp without dental caries, single-rooted tooth, and no periodontal ligament space.

The gingival mucosa was cut during preparation of the tooth-bone-periodontal complex, and then the tooth-bone-periodontal complex was cut and harvested using a surgical saw, with careful attention to the roots of adjacent teeth. After the tooth-bone-periodontal complex had been harvested, Teruplug® (Olympus Terumo Biomaterials) was applied to accelerate wound healing of the cleft to cover the defect. The tooth-bone-periodontal complex was then shaved to become the lamina (4 mm thick), and a hole was drilled through the dentine. To prevent the tooth crown from slipping while the tooth-bone-periodontal complex was shaved, we used a tooth-fixing device instead of tooth extraction forceps during the preparation period, as previously reported (Fig. 1).[8] The tooth-bone-periodontal complex that is used as a base material for the PMMA optical cylinder should include the surface dentine of the tooth-bone-periodontal complex to aid adhesion of the optical cylinder. The gingival mucosa was left attached to the alveolar bone, and the epithelium of gingival mucosa was carefully scraped off using a surgical knife and removed, as these soft tissues help the tooth-bone-periodontal complex fix the recipient nascent tissue after retrieval in the second stage.

For preparation of the hole, the tooth-bone-periodontal complex was drilled on the dentine surface, not the bony surface, by gradually changing the size of the fissure bars. After preparation of the final hole (3.5 or 4.0 mm in diameter), the optical cylinder (Morcher GmbH) was inserted into the lamina and adhered and fixed by using 4-methacryloxyethyl trimellitate anhydride in PMMA initiated by tri-n-butyl borane (4-META/MMA-TBB) resin (Super-Bond C&B®, Sun Medical). For this procedure, cylindrical rubber was fixed on the end of the optical cylinder to make it technically usable (Fig. 2). The harvested lamina with the optical cylinder was then washed with saline (Fig. 3). After being sprayed with fibrin glue (Beriplast®; CSL Behring KK), the prepared lamina was placed into the submuscular pocket

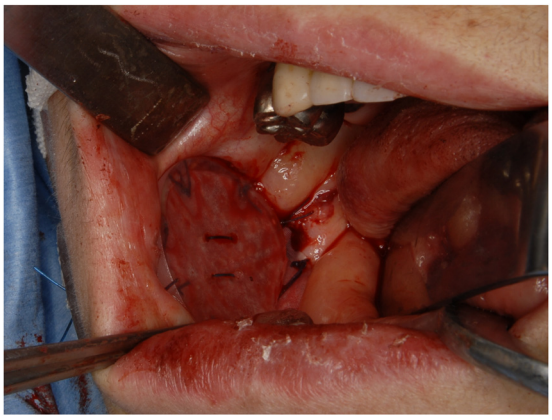

Fig. 4. The buccal mucosa (a circle 3 cm in diameter) was grafted from healthy cheek mucosa. After grafting, artificial dermis (Terudermis®) was applied to the buccal mucosal defect. The surgical wound was layered with silicone sheets between the oral side and the dermis to help wound healing.

under the orbicularis oculi for fibrovascular tissue recovery on the osteo-odontal surface for the second stage.

Next, a full-thickness buccal mucosal graft 3 cm in diameter was harvested (excluding muscle) from cheek mucosa, paying attention to avoid injury to the orifice of the parotid duct. The mucosal graft was soaked in antibiotic solution before the graft was sutured over the corneal and scleral surfaces of the eye. Artificial dermis (Terudermis®, Terumo Corp) was then applied to the buccal mucosal defect, layered with 3 mm silicone sheets between the oral and dermis side to help wound healing of the buccal mucosa (Fig. 4).

The thick keratinised ocular surface membrane was then cut and separated to expose the cornea and sclera. The rectus muscles were separated by hook and sutures, and the grafted buccal mucosal sutured to the sclera bounded by the insertion sites of rectus muscles to create a new ocular surface.

Stage 2

About 2–4 months after the first stage, the lamina of the OOKP was retrieved from the submuscular pocket, and excess soft tissue removed from the bone surface. All soft tissue on the surface of the dentine was removed. The buccal mucous membrane was then cut, and the Flieringa ring was sutured. The central 5.0 mm of the cornea was trephined. We pulled out the whole iris, and extracted the lens with extracapsular cataract extraction followed by open-sky anterior vitrectomy.

The posterior part of the optical cylinder was inserted through the central corneal hole. The lamina was then sutured onto the cornea and sclera. The mucosal flap was replaced and sutured after cutting a hole to allow the protrusion of the anterior part of the optical cylinder. A cross-sectional diagram of the OOKP after the second stage operation is shown in Fig. 5.

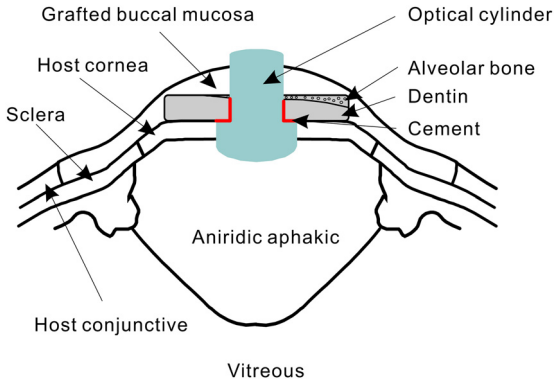

Fig. 5. Diagram of the harvested tooth-bone-periodontal complex lamina placed between the grafted buccal mucosa and the conjunctiva.

Results

We have done first-stage OOKP operations (on four men and five women, aged 34-77 years) in Japan (Table 1). Five and two patients, respectively, had developed blindness as a result of Stevens-Johnson syndrome and ocular cicatricial pemphigoid, and one patient each had chemical and thermal burns to the cornea and ocular surface.

The donor tooth for OOKP was the canine (three upper and six lower) for all patients, because all nine patients had adequate canine teeth. The buccal mucosa was not damaged in any patient, and did not affect harvesting of the graft, although some patients had mucocutaneous disease.

Nine patients successfully had the first stage of OOKP; all were unilateral. The operative complications were exposure of adjacent roots in two cases. No oronasal perforation or sensory disturbance of the lower lip was caused by damage of the mental foramen. None of the patients had trismus, although all nine patients developed submucosal scar bands in the donor site of the buccal mucosal graft. All patients had prosthodontic restorations. No implants have been inserted so far because of the large alveolar bone defect after harvesting, although they could clearly contribute to a simplification of treatment compared with removable or tooth-fixed dentures.

Eight of nine patients completed second stage OOKP operations with the longest follow-up being 14 years and 11 months (mean follow-up: 8 years, 11 months). One patient (case 4,Table 1) who did not have a second stage OOKP operation developed primary myelofibrosis during the waiting period between the first and second stage operations and died. All eight remaining patients have been stable with no problems, including dislocation or extrusion of the optical cylinder. However, there were minor complications related to the optical cylinder, such as tilting, buccal mucosal overgrowth, and glaucoma tube shunt. To date, there have been no important or sight-threatening keratoprosthesis-related complications. All patients were thought to have attained their possible visual potential after OOKP (Table 1).

Table 1
Details of nine patients who had a first stage osteo-odontokeratoprosthesis in Japan.

Case No.	Diagnosis	Age/ sex	Operated eye	Harvested tooth (No.)	Optic diameter	Best visual acuity	Current visual acuity	Complications	Duration of follow-up (years/months)
1	SJS	49/F	Left	33	3.5	1.2	0.5	None	14y11m
2	SJS	8/F	Left	33	3.5	0.6	0.04	Optic cylinder tilting	14y1m
3	SJS	63/M	Left	33	4.0	0.2	0.1	Mucous overgrowth	13y9m
4	SJS	77/F	Left	43	3.5	–	–	None	–
5	OCP	72/F	Left	23	3.5	0.04	0.04	None	11y7m
6	Chemical	46/F	Left	23	3.5	1.2	0.6	Glaucoma tube shunt	7y9m
7	SJS	59/M	Right	3	4.0	1.0	1.0	None	6y5m
8	Thermal	34/M	Left	13	3.5	HM	HM	None	1y11m
9	OCP	64/M	Left	43	3.5	0.5	0.5	None	8m

SJS = Stevens-Johnson syndrome, OCP = ocular cicatricial pemphigoid, Chemical = chemical burn, and Thermal = thermal burn.

Discussion

We successfully did nine first stage OOKP operations in Japan. Postoperative clinical assessments were made to ensure adequate follow-up by the oral surgeon and the ophthalmologist. The prognosis in all cases has been good.

The main contribution of the oral surgeon is during the first stage. Selection of the donor tooth is important, and as reported previously, the ideal tooth should be of adequate size and shape, be single-rooted with healthy pulp, and without dental caries, and have no periodontal complex with healthy surrounding alveolar bone.[1] The pulp should be removed during preparation. Assessment of the suitability of a tooth depends on clinical and radiological examination by panoramic radiography and CT at initial presentation. Berg et al reported that the use of cone-beam CT simplifies preoperative decision-making and the process of OOKP.[9] In addition the surrounding anatomy (such as maxillary sinus or mental foramen) should be assessed for the avoidance of possible complications and to minimise cosmetic and functional defects after the tooth-bone-periodontal complex has been harvested. The canine is therefore generally the best candidate, as it is the most reliable and accessible tooth.[6] The density of the mandibular bone is greater than that of the maxilla, which may subsequently cause differences in alveolar bone resorption of the maxilla and mandible. Long-term postoperative alveolar resorption may therefore differ between upper and lower canine lamina. The lower canine lamina may have an advantage for the long-term prognosis. Conversely, that of the upper canine is generally slightly larger than the lower canine, so the upper canine is more manageable for preparation of the lamina than the lower canine. However, if tooth conditions are similar, then both canines are suitable. In such cases, the state of the remaining teeth after extraction of the canine should be considered.

The tooth-bone-periodontal complex is shaved into the lamina (4 mm thick), and a hole is drilled through the dentine. A resin stick handling device is useful for this. The device reduces accidental iatrogenic injury caused by technical errors by freely holding the tooth extraction forceps, and could prevent gingival damage during preparation of the lamina.[8] Preservation of a periodontal ligament is suggested to prevent the down-growth of gingival tissue that is associated with a possible inflamed tooth and resorption of alveolar bone.[6,10]

After preparation the optical cylinder is inserted into the lamina and fixed using 4-META/MMA-TBB resin. Because the physical stress on the optical cylinder after grafting in the orbital lesion when there are no tears is comparatively less than that in the oral lesion, fixation of the optical cylinder on the lamina should be stable enough to adhere.

We sprayed fibrin glue (Beriplast®) on to the prepared lamina before placing it into the submuscular pocket under the orbicularis oculi. Fibrin glue helps to accelerate recovery of fibrovascular soft tissue on the osteo-odontal surface. This procedure improves subsequent suturing of the soft tissue on the recipient tissue during the second stage. Teruplug®, an absorbable atelocollagen sponge, is used as scaffold material to help wound healing after the tooth-bone-periodontal complex has been removed. Teruplug® comprises a sponge block configuration and is shaped for easy placement in the surgical wound. Filling wound defects reduces bleeding, improves granulation formulation, and protects the wound surface. We had no postoperative complications such as infection of the surgical site or postoperative bleeding. Additionally, artificial dermis (Terudermis®). which is made of atelocollagen, is applied to the buccal mucosal defect, with the expectation of good adhesion and reduction in pain.

The dental implant restoration for a missing tooth can better maintain long-term oral quality of life than a removable denture. In addition, a tooth-fixed denture (dental bridge) requires alteration of adjacent teeth to support the bridge, so a dental implant is a better option as it provides dental restoration without alteration of adjacent teeth, thereby simplifying treatment. However, in our cases, dental implants were not used because of the large alveolar bone defect after harvesting. If requested by the patient, restoration of a dental implant could be done with a bone graft.

OOKP is indicated for scarred corneal disorders with loss of function of the lacrimal gland. OOKP has been done in only a few countries, and is not widely known around the world. Compared with a corneal graft, it is a complex pro-

cess that may lead to postoperative cosmetic problems. It requires interdisciplinary management by ophthalmologists and oral surgeons, and has a long operating time. However, it is a useful option for severe cases as the patient generally has good function without the need for immunosuppressive agents. Further improvements, such as use of an artificial basement membrane instead of harvesting of the tooth-bone-periodontal complex, are currently being developed to enable more effective and less invasive procedures.

Conclusion

We have successfully done nine first-stage OOKP operations in Japan. Eight of the nine patients have completed the second stage, with a long follow-up. Postoperative clinical assessments have been made to ensure adequate follow-up by the oral surgeon and the ophthalmologist. The prognosis in all cases has been good.

Conflict of interest

We have no conflicts of interest.

Ethics statement/confirmation of patients' permission

We did not need ethics approval. We have obtained the patients' permission.

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