Strampelli's osteo-odonto-keratoprosthesis. Clinical and histological long-term features of three prostheses

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Abstract

The histological features are reported of osteo-odonto-acrylic laminae removed from three patients who for differing underlying causes received Strampelli's osteo-odonto-keratoprotheses (OOK) 20, 16, and 12 years previously. It appears that preservation of the alveolar-dental ligament plays a definitive role in the maintenance of the prosthesis. If this tissue undergoes necrosis as a consequence of an inflammatory disease 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.

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Accepted for publication
29 August 1991

Osteo-odonto-keratoprosthesis (OOK) constitutes a valid option in the management of patients with opaque corneas in whom penetrating keratoplasty is not feasible.¹⁻⁸ This technique was developed by Strampelli in 1963⁹ and is now well recognised despite being, wrongfully in our opinion, unduly neglected. Indeed this prosthesis has stood the test of time although the complexity of the surgical technique does represent a disadvantage.

The removal of three prostheses 20, 16, and 12 years, respectively, after implantation for different causes has allowed us to investigate the preservation status of the osteodental laminae after prolonged periods.

Patients

CASE 1

The male patient aged 54 years had suffered from

bilateral corneal opacities due to lime burn since 1960. In 1968 bilateral OOK was performed. In the right eye a small incisor tooth was used to support the optical device. Because of the smallness of the tooth the fixed lens could only be 1.75 mm in diameter. Postoperative visual acuity was 1/10 and remained so for 20 years after implantation when the right osteo-odontoacrylic lamina (ODAL) was removed and a new one, provided with a larger lens (2.75 mm), was implanted. After this second operation visual acuity improved to 5/10. The ODAL in the left eye was still in position but visual potential had been lost because of secondary glaucoma.

CASE 2

In 1970 a male patient, aged 50 years, lost the whole left eye and the right eye cornea became opaque as the result of alkali burns. In 1973 a right OOK was successfully performed with a postoperative visual acuity of 6/10. The clinical course was unremarkable until the beginning of 1988 when the right eye was affected by repeated inflammatory episodes which did not regress after the administration of broad spectrum antibiotic therapy and anti-inflammatory agents. Due to this complication the patient's visual acuity had decreased to only light perception. Finally 1 year after the onset of the ocular inflammation the lamina had to be removed because of the development of a mucosal ulcer and a white dense membrane behind the implant.

CASE 3

The patient was a 46-year-old female suffering from Hallerman-Streiff syndrome since birth with bilateral blindness. In 1978 a left OOK was performed leading to no substantial visual improvement. In January 1990 acute inflammation of the periprosthetic tissues occurred which did not respond to treatment. Two months later, with the symptoms persisting and the visual acuity not having recovered, the prosthesis was removed and sent for histological examination.

Pathology

The ODALs thus obtained were fixed for 24 hours with 4% buffered formalin and glycol methacrylate embedded with no prior decalcification. Sections of 1-2 µm were cut with a carbon tungsten knife and stained either with routine methods (haematoxylin-eosin, azur II methylene blue, May-Grunwald-Giemsa) or with histochemical methods (PAS for glyco-

Figure 1 Mature bone and medullary cavities containing aggregates of pigmented cells circumscribe the site of lens. (Haematoxylin and eosin, ×23.)

Figure 2 Alveolar dental ligament is replaced by a chronic inflammatory tissue. Mononuclear phagocytes with eosinophilic globules in the cytoplasm are seen. (Haematoxylin and eosin, $\times 90$.)

Figure 3 Chronically inflamed alveolar dental ligament lined by non-keratinising squamous epithelium. (Haematoxylin and eosin, $\times 23$.)

Figure 4 Strampelli's osteo-odonto-keratoprosthesis.

proteins, Masson Fontana for melanin and Perl's for iron).

Sections obtained from the ODAL removed after a 20-year period (case 1) showed predominantly mature lamellar bone with minimal immature woven bone. Focal moulding characterised by inactive lacunar and marginal resorption and lack of osteoclasts and osteoid tissue were present. Fragments of dental tissue were connected to the bone by fibroligamentous tissue. Bone and tooth circumscribed an optically empty hole presumed to have contained the lens (Fig 1). Neither necrotic nor inflammatory foci were seen in these tissues. However in the medullary cavities numerous mast cells at the periosteal site were observed as well as aggregates of large cells with round nuclei and voluminous cytoplasm containing brown granules that proved positive when submitted to Fontana staining. Free pigment granules were also detected in adjacent tissues.

The inflamed ODAL removed after 16 years of implantation (case 2) was devoid of bone which had broken into pieces during removal. Microscopically the tooth displayed no morphological alterations. However next to it a chronic inflammatory reaction was observed, mainly consisting of plasma cells and lymphocytes, that was almost totally surrounded by non-keratinising squamous epithelium (Fig 2). This epithelium having focally penetrated the inflammatory tissue showed multiple areas of ulceration in which necrosis and polymorphonuclear leucocytes were seen. Interspersed within the chronic inflammatory cells were a few mononuclear phagocytes showing one or more eosinophilic PAS-negative anisometric globules in the cytoplasm. Occasionally similar globules could be observed in the extracellular matrix.

Histological sections obtained from the third patient were composed of dentine, woven bone, and soft tissues. Normal dentine prevailed; it showed a central round aperture where the lens was located. In the contiguous totally viable bone tissue physiological resorption activity was evident. Bone was partially covered by non-keratinising squamous epithelium with focal interposition of chronic mononuclear infiltrate rich in plasma cells. Around the tooth there was well vascularised and chronically inflamed fibrous connective tissue partially lined by non-keratinising squamous epithelium (Fig 3). Among the inflammatory cells numerous mast cells were present.

Discussion

The operative technique is certainly complex inasmuch it involves the performance of different operations which take up a period of 6 to 12 months. The optical device consists of a 3 mm thick lamina obtained from a tooth root and alveolar dental ligament of the patient; an acrylic lens is inserted in the lamellar section (Fig 4). The OOK technique has been described previously in detail.^{48,9}

Briefly, Strampelli's OOK offers three advantages over the alloplastic prostheses: (1) permanent fixation of an acrylic lens; (2) insertion of the mucosal epithelium onto the

alveolar dental ligament; (3) long-term retention of the prosthesis due to its complete biological compatibility as far as other pathological conditions do not occur.

As the anterior surface of the ODAL protrudes over the surrounding tissues epithelial proliferation is stimulated. However when the epithelium reaches the alveolar dental ligament the proliferation ceases. In this way the lens will not be covered by a membrane and the epithelium will not proliferate behind the optical cylinder to promote fistula formation, secondary intraocular infection, hypotone, uveitis, visual loss, and probable extrusion of the ODAL. Long-term retention of a keratoprosthesis mainly depends on whether it behaves as a graft or a simple implant. On the other hand mesoprostheses (like keratoprostheses) will not be retained unless a true biological connection is established with the tissue at the point at which it is inserted – namely the epithelium.

In the literature few authors have reported their direct experience with Strampelli's OOK^{2,3,10} and some series related only to small numbers of cases.

Our series is composed of 75 patients on whom Strampelli operated between 1964 and 1975 and who have subsequently been controlled by the present authors; 86 additional operations were performed at our centre from 1968 to 1990. Of these six were performed in 1990. On the whole the incidence of ODAL necrosis was very low (3.8%) and was associated with loss of the alveolar dental ligament. In particular necrosis had occurred in only one out of 155 cases within the first 10 postoperative years while in five instances it occurred 10 to 20 years later. It seems that no rejection of the prostheses occurred because of the compatibility of the dental support.

However other potential causes of blindness should be noted especially secondary glaucoma unresponsive to medical or surgical therapy, fistula formation, and occasionally, retinal detachment.

Compared with Strampelli's OOK other kinds of keratoprostheses with synthetic support frames, in addition to the aforementioned complications, carry with them the risk of extrusion of the implant as retention is ensured only by pure mechanical contact.

Cardona refers to an extrusion rate of 21.3% with the through-and-through keratoprosthesis due to several causes¹¹; other authors using the same technique confirm this possibility.¹²⁻¹⁴ Moreover, with Choyce's two-piece perforating keratoprosthesis epithelial erosion around the optical cylinder is to be expected in 21.6% of cases.¹⁵

Some authors performing Strampelli's OOK report a high incidence of extrusion from an unspecified cause or visual loss due to secondary glaucoma and eye infection.¹⁰ According to our experience and to the experience of other authors,³ implant necrosis does not occur provided the tooth is correctly removed along with a viable alveolar dental ligament.

Biological tolerance of the ODAL was confirmed in our series by histological examination

of three prostheses. The oldest one, in which the prosthesis was changed 20 years after implantation to provide a device carrying a larger optical cylinder, was microscopically still composed of viable and well preserved tissues. When tissues become inflamed or undergo necrosis the implant behaves like an allogeneic material and is eventually extruded. This possibility was demonstrated by histological examination of the second ODAL removed 16 years after implantation. In this instance the alveolar dental ligament was replaced by a chronic inflammatory tissue. Therefore it may be asserted that as long as the grafted tissue remains viable the prosthesis will stay in position. In the ODAL removed prophylactically 2 months after initial inflammation a viable alveolar dental ligament was present, although a moderate to intense chronic non-specific surrounding infiltrate could be observed. Furthermore the dental and bone structures were uncompromised which suggests that long-lasting inflammation is necessary to cause necrosis of the prosthesis.

Strampelli's OOK is a surgical technique offering long-term guarantees of tolerance and steadiness and is the only prosthesis with more than 20 years follow-up. Nevertheless the development of an allogeneic keratoprosthesis capable of lasting as long as an OOK but easier to implant and less traumatic for the patient would be valuable. Some of the newer keratoprostheses appear promising,¹⁶ although they need further experimentation on a long-term basis.

We are grateful to Professor A Garner for constructive criticism and to Miss Silvia Berni for the expert technical assistance.

- 1 Falcinelli G, Missiroli A, Petitti V, et al. Osteo-odonto-keratoprosthesis up to date. In: *Acta XXV Concilium Ophthalmologicum*. Proceeding of the XXVth International Congress of Ophthalmology, Rome, May 4-10, 1986. Amsterdam, Berkeley, Milano: Kugler & Ghedini, 1987: 2772-6.
- 2 Iannetti F. Statistical consideration on Strampelli's osteo-odonto-keratoprosthesis. *Ann Ottalm Clin Ocul* 1974; 100: 57-82.
- 3 Lacombe E. Possibilités actuelles des keratoprothèses. *J Fr Ophthalmol* 1986; 9: 323-9.
- 4 Marchi V. Osteo-odonto-cheratoprotesi (l'intervento fondamentale e gli interventi accessori). Parte I: l'intervento di osteo-odonto-cheratoprotesi. *Ann Ottalm Clin Ocul* 1989; 95: 325-38.
- 5 Marchi V, Fiorimonte F, De Martiis A. Osteo-odonto-cheratoprotesi (l'intervento fondamentale e gli interventi accessori). Parte II: l'intervento di cataratta. Il trattamento delle membrane retrolenticolari. *Ann Ottalm Clin Ocul* 1989; 95: 349-54.
- 6 Marchi V, Fiorimonte F, Manni GL, et al. Osteo-odonto-cheratoprotesi (l'intervento fondamentale e gli interventi accessori). Parte III: gli interventi antiglaucomatosi. *Ann Ottalm Clin Ocul* 1989; 95: 403-21.
- 7 Marchi V. Results of 62 osteo-odonto-keratoprosthesis. *Acta XXV Concilium Ophthalmologicum*. Proceeding of the XXVth International Congress of Ophthalmology, Rome, May 4-10, 1986. Amsterdam, Berkeley, Milano: Kugler & Ghedini, 1987: 1244-9.
- 8 Marchi V. Osteo-odonto-keratoprosthesis. *Ottalmol* 1970; 31-67.
- 9 Strampelli B. Osteo-odonto-keratoprosthesis. *Ann Ottalm Clin Ocul* 1963; 89: 1039.
- 10 Casey TA. Osteo-odonto-keratoprosthesis and chondro-keratoprosthesis. *Proc Roy Soc Med* 1970; 63: 313.
- 11 Cardona H. Keratoprosthesis. *Am J Ophthalmol* 1962; 54: 284-94.
- 12 Barron BA, Dingeldein S, Kaufman HE. Spontaneous unscrewing of a Cardona keratoprosthesis. *Am J Ophthalmol* 1987; 103: 331-2.
- 13 Dhawan Singh, Bansal DC, Avtar Singh. Keratoprosthesis: a clinical study. *Ind J Ophthalmol* 1973; 21: 112-6.
- 14 Rao GN, Blatt HL, Aquavella JV. Results of keratoprosthesis. *Am J Ophthalmol* 1979; 88: 190-204.
- 15 Choyce DP. Results of keratoprostheses in Britain. *Ophthalmic Surg* 1973; 4: 23-32.
- 16 Pintucci F, Pintucci S. Pemfigoide oculare in paziente adentulo. Nuovo tipo di cheratoprotesi. In: L Cappelli, ed. *Atti SOI*. Nuova Casa, Bologna, 1986: 139-45.