ORAL & IMPLANTOLOGY

case report

UNICYSTIC AMELOBLASTOMA: REHABILITATION WITH CHIN GRAFT HARVESTED AND IMPLANT-SUPPORTED FIXED PROSTHESIS

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SUMMARY
Objective. The objective is to present a clinical case of a 38-year-old male with a maxillary unicystic ameloblastoma treated by means of tumor block resection followed by chin-harvested graft placement in order to place two dental implants for esthetic and functional rehabilitation.
Methods. Ameloblastoma is a benign odontogenic tumor characterized by local aggression and a high rate of recurrence; the latter partly depends on how it is treated. Complete resection of the tumor, which usually prevents recurrence, produces bone defects of varying size that must be reconstructed later on. In most cases this is done using bone grafts and implant-supported prostheses. Grafts harvested from the chin are relatively easy to obtain and enjoy a fairly uneventful post-operative with few complications; they are suitable for cases in which the defect generated by resection is of small size.
Results. Functional and esthetic rehabilitation and the tumor has not relapsed during a 7-year follow-up.
Conclusion. Tumor block resection followed by chin-harvested graft placement and dental implants is a safe treatment for patients with unicystic ameloblastoma.

Key words: ameloblastoma, chin graft, dental implants.

Introduction

Ameloblastoma, together with odontoma, are the most common odontogenic benign tumors in the maxillary and mandible. Ameloblastoma is characterized by locally aggressive growth, and a high rate of relapse. Its incidence is around 0.5 cases per million subjects per year although higher incidences have been reported in some African and Asian countries (1, 2). It usually appears between the ages of 30 and 60 years and shows no predilection for gender (3). In 2005, the World Health Organization produced a classification of odontogenic tumors, which recognized four sub-types of benign ameloblastoma: solid/multicystic, desmoplastic, extraosseous/peripheral, and unicystic (4). This entity was first described in 1977 by Robinson and Martínez (5), appearing in radiographs as an

unilocular, macroscopic, cystic, clinical entity, which best responds to conservative treatment. Unicystic ameloblastoma (UA) appears in younger patients than other ameloblastomas, and even in children (1, 2, 6). The average age when UA appears is 18.7 years according to Robinson and Martínez (5), or 26.9 years according to other Authors (7).

In spite of good response to conservative treatment, up to 30.5% of UAs treated by enucleation relapse, compared with 3.6% treated by resection, making resection the UA treatment of choice (6, 8).

The bone defect produced by resection of the tumor must be remedied by means of bone regeneration techniques; of these, block bone grafting predominates. When the defect is small, a block bone graft harvested from the chin can be placed and shows an excellent risk/benefit ratio (9).

The aim of this study is to present a case report of block resection of an unicystic ameloblastoma in a 38-year-old man, and rehabilitation of the bone defect with an *en bloc* chin graft and placement of an implant-supported prosthesis.

Case report

This is the case of a 38-year-old male who was attended the clinic because of a relapsed unicystic ameloblastoma treated one year earlier by enucleation. The patient brought with him a panoramic radiograph showing a radiolucent image extending from the maxillary left premolar area, which had increased in size since enucleation (Figure 1 a). The clinical examination reveal painless, and did not detect swelling or any reported symptoms or any other local sign, such as mucosa disorders or bone expansion. Tomographic study confirmed the presence of a round radiolucency area lesion in the maxillary measuring approximately 1.5x1.5 cm (Figure 1 b). To minimize the risk of further relapse, and having received the patient's informed consent to proceed, the lesion was block resected with safe margins under general anesthesia, at the request of the patient, suturing the soft tissues edge to edge (Figure 2). Anatomopathological study confirmed diagnosis of a plexiform unicystic ameloblastoma (Figure 3).

Five years later, a block graft was harvested from the chin to reconstruction the defect caused by tumor resection, filling any gaps with particulate bone and immediate esthetic prosthesis (Figures 4, 5, 6). After six months, the patient showed no evidence of local recurrence on follow-up computed tomography (Figure 7), so it was decided to place 2 implants in this area and were placed at positions 24 and 25 [Normon HI 3.75x11.5 mm (Tres Cantos, Madrid, Spain)] and, after the osseointegration period, their respective fixed prosthetic crowns (Figure 8 a, b). Seven years since tumor resection, no signs of recurrence have appeared and the implants continue in perfect state (Figure 9).

Discussion

Ameloblastoma is a benign odontogenic tumor that constitutes 1% of all oral tumors (10). It presents locally aggressive growth and a high tendency to relapse, two characteristics that make a sharp contrast with its benign histologic status (11). Up to 80% of ameloblastomas appears at the mandibular angle or mandibular ramus. They show no predilection for gender and most are diagnosed in patients aged 30-60 years (1, 3). In the present case, the UA appeared in the maxillary; this is extremely unusual given that extensive literature reviews such as Lau and Samman (6) do not mention a single UA in the maxillary, while the age when this case appeared – 38 years – is older than cases reported by other Authors (5, 7).

In comparison with other ameloblastomas, UA presents a series of special characteristics but its radiological aspect is such that it can be easily confused with a dentigerous cyst or keratocyst, a situation that makes histologic confirmation of diagnosis essential, as in the present case (6).

Ackermann et al. (12) redefined the diagnostic

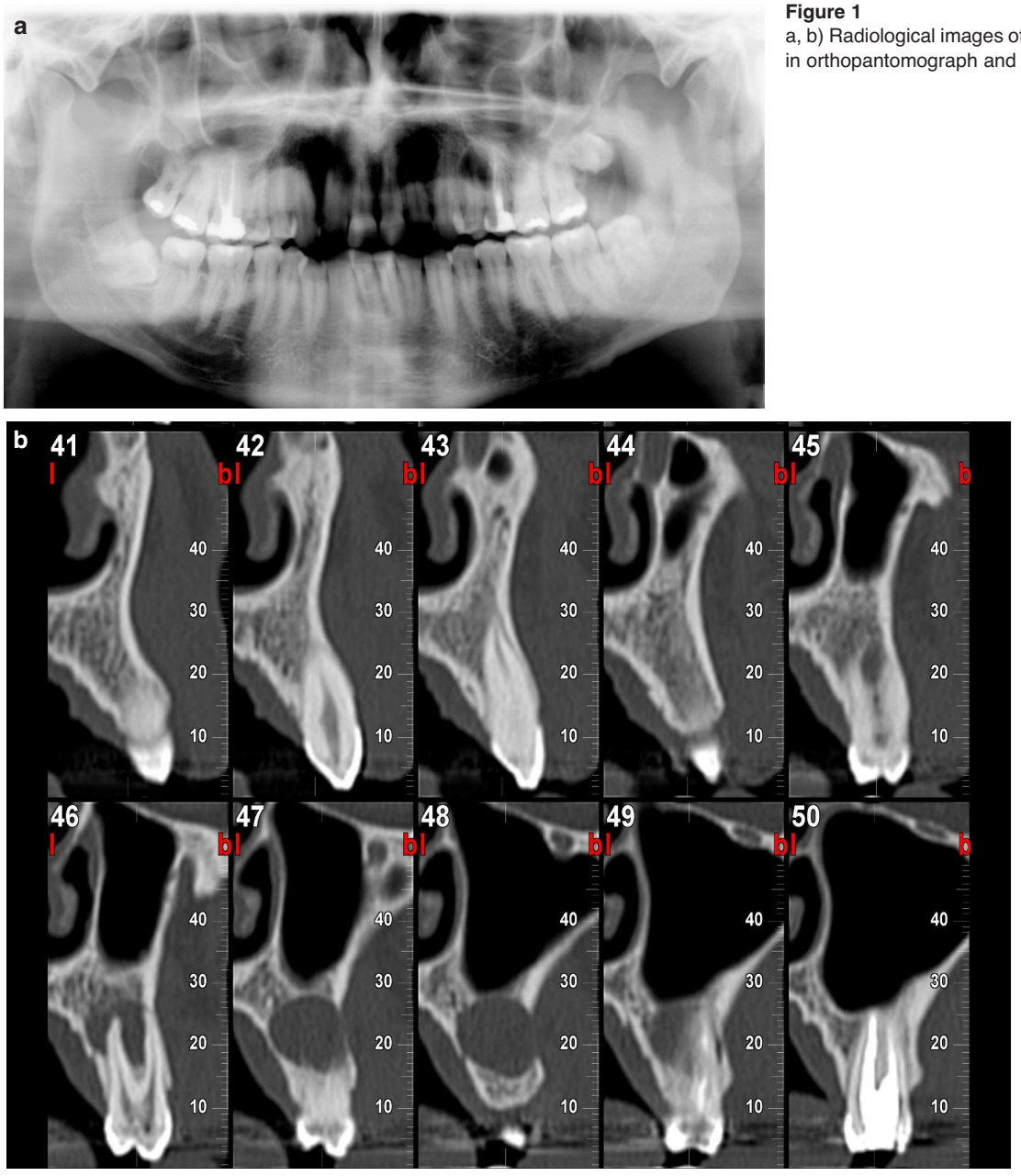

Figure 1
a, b) Radiological images of the lesion in orthopantomograph and scan.

criteria originally proposed by Robinson and Martínez (5) classifying UAs as consisting of three sub-types according to their prognostic and therapeutic implications: Type 1 – A unilocular cystic lesion lined by epithelium; Type 2 – A nodule arising from the cyst lining, projecting into the lumen of the cyst, and comprising odontogenic epithelium with a plexiform pattern which closely resembles that seen in the plexiform ameloblastoma; Type 3 – The presence in the connective tissue wall of the cyst, of invasive islands of ameloblastomatous epithelium which might (type 3b) or might not (type 3a) be connected to the cyst lining. The same Authors propose excisional biopsy to treat unilocular lesions, the mode of treatment applied in the present case.

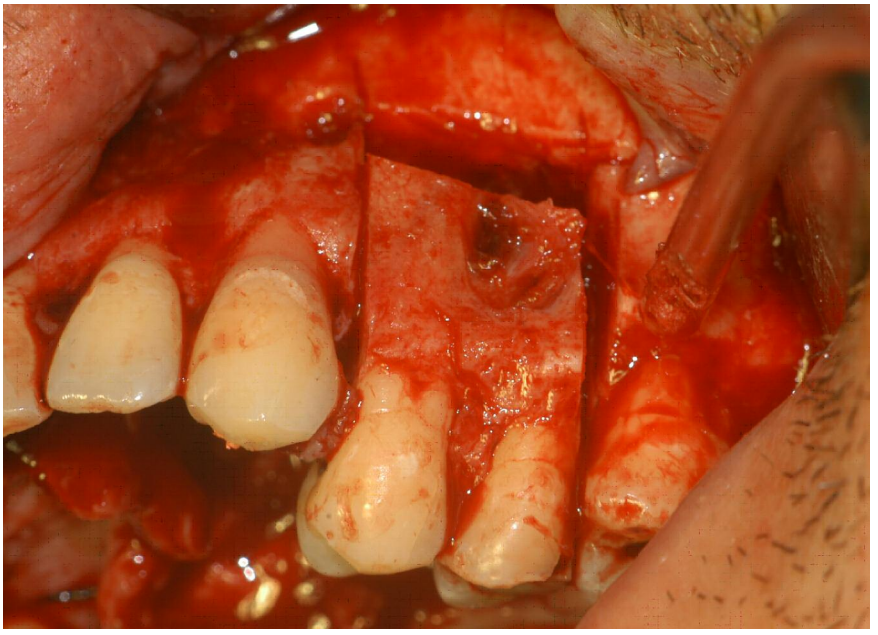

Figure 2
En bloc resection of the tumor.

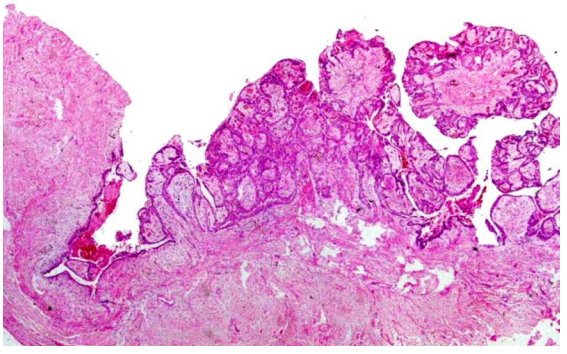

Figure 3
Histological image of the tumor.

With regard to the therapeutic options for treating UA described in the literature, they are mainly based on conservative techniques such as enucleation with curettage or marsupialization, which reduces the size of the tumor in preparation for less aggressive surgery afterwards; this type of treatment is for types 1 and 2 as described above. Complete removal with safety margins is reserved for treating type 3 UA, as in the present case, or in cases of relapse of types 1 and 2. Safety margins have been established according to Carlson and Marx (13) who showed that ameloblastoma can extend 2.3-8 mm beyond the radiographically visible tumor edge, which means that having performed enucleation of the tumor, ameloblastoma cells may remain in the area favoring recurrence, as in the case of the type 3 UA (described by Ackerman) (12).

UA relapse is less common than with other types of ameloblastomas, occurring in 0.2-12% of cases (8) and according to Lau and Samman (6) block resection is the treatment mode with the lowest incidence of recurrence: 3.6%. Enucleation followed by application of Carnoy's solution presents an 18% incidence of relapse, rising to a 30.5% incidence of relapse after enucleation alone. These percentages justify the treatment option applied in the present case.

With regard to the time when recurrence appears following elimination of the tumor, Reichart et al. (14) affirm that 50% appears within the 5 first years after intervention but may appear at any time up to 33 years after treatment. In the present case, en bloc chin graft for later prosthetic rehabilitation was performed 5 years after UA resection, the interval during which most relapses occur.

En bloc resection of UA or any other type of

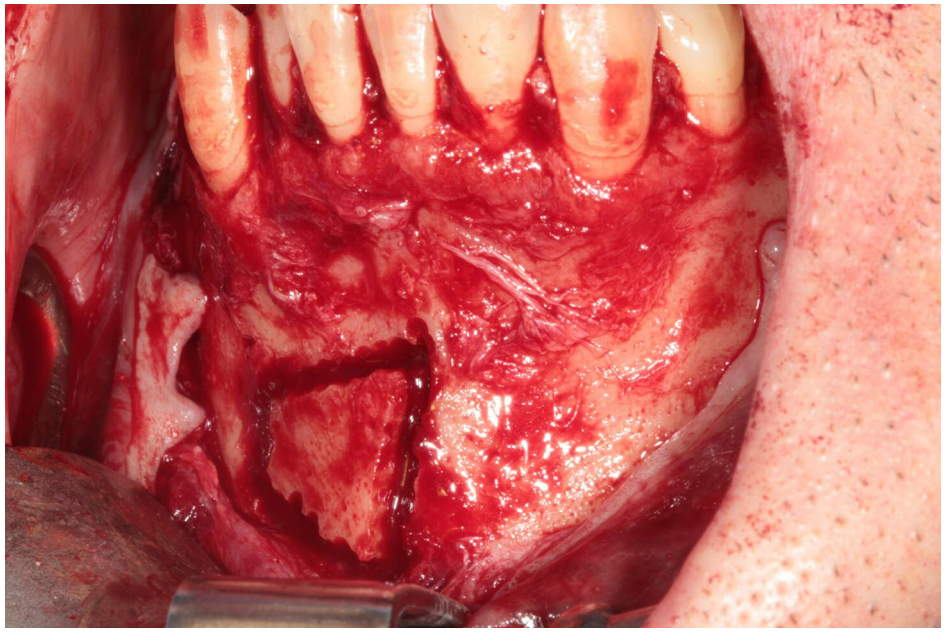

Figure 4
Graft removed from chin.

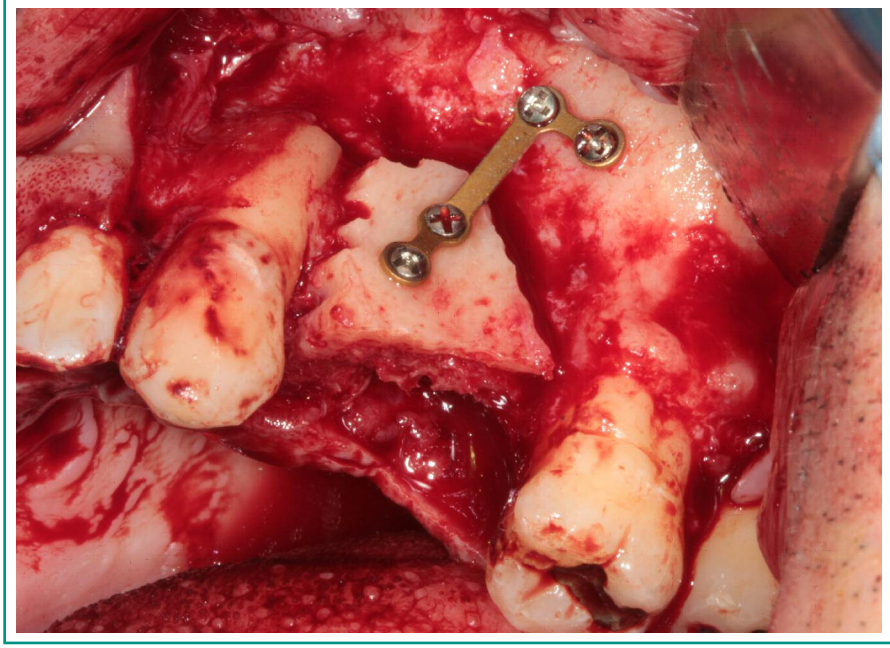

Figure 5
Regeneration of the bone defect 5 years after tumor resection.

ameloblastoma produces bone defects of variable size that must be reconstructed in order to restore lost function and esthetics and to minimize the psychological impact of the surgical outcome (15). The most commonly used donor sites for bone reconstruction in the oral cavity are the mandibular ramus, mandibular symphysis, and maxillary tuberosity; the most frequently occurring complications deriving from bone harvesting can be treated locally or, in the case of more complex complications, by additional surgery. Most published cases have used

Figure 6
Suture and immediate esthetic prosthesis.

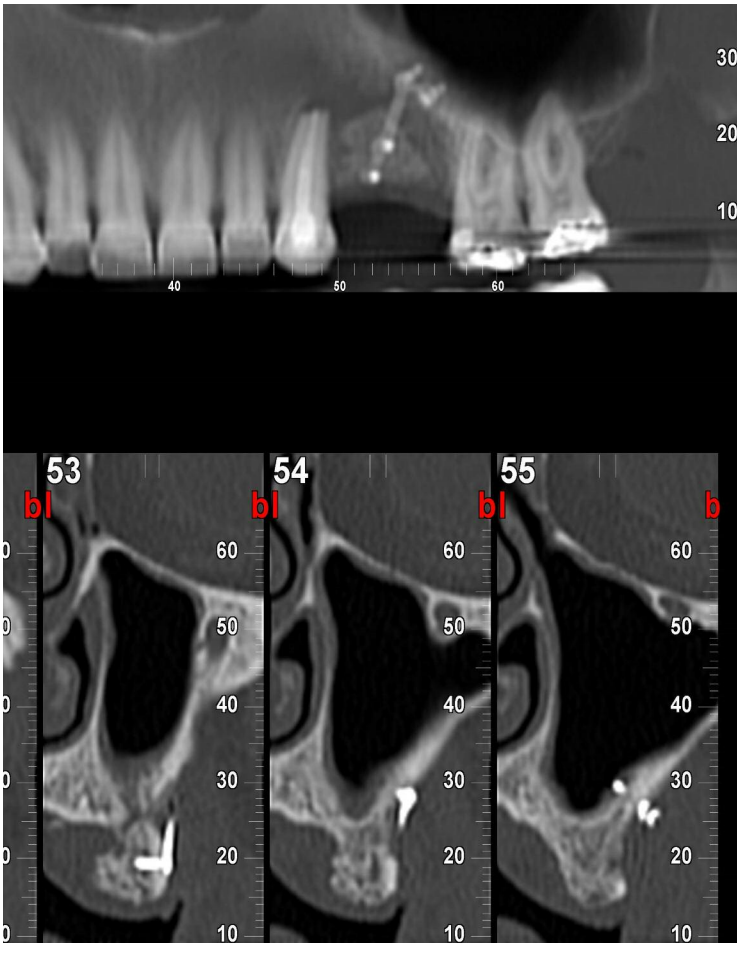

Figure 7
No evidence of local recurrence on follow-up computed tomography.

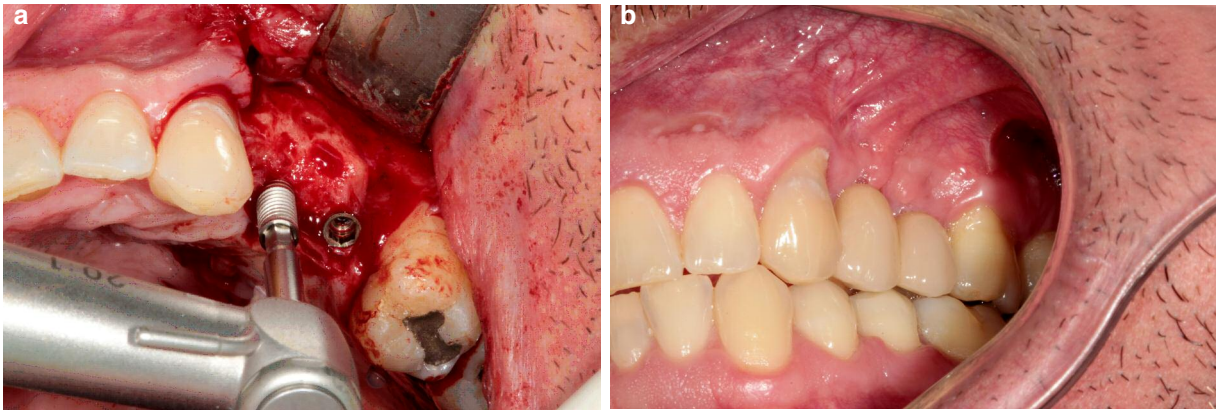

Figure 8
a) Implant placement and prosthetic rehabilitation; b) radiological aspect after 7-year-follow-up.

Figure 9
Clinical aspect of prosthetic rehabilitation.

grafts harvested from the iliac crest to reconstruct the bone defect, although these were large resections resulting from the removal of large multicystic ameloblastomas (10, 16, 17).
According to Misch et al. (18), the minimum alveolar crest dimensions required for implant insertion are 5 mm bone thickness and 10 mm height. When these dimensions are smaller, it is necessary to perform some reconstructive procedure, in which grafts harvested from the upper maxilla undergo a resorption index of around 10%, while those of mandibular origin undergo 5% (19). The choice of donor site will ultimately depend on the bone volume required to deal with the defect. Bone harvested from the chin is indicated for small and medium-size bone losses, equivalent to four teeth in width and two in height as put forward by Misch et al. (18), so the present case constituted an ideal indication for this type of *en bloc* graft. The chin-harvested *en bloc* graft offers biological and immunological advantages in comparison with xenografts, allografts or alloplastic materials as it has viable cells (for osteogenesis), bone morphogenetic protein (for osteoinduction), and a bone matrix (osteoconduction) (19-21). The chin offers additional advantages as a donor site due to its accessibility, ease of harvesting (even under local anesthesia), minimal morbidity, and an absence of visible scarring (22). Its main complication – incisor and mental nerve damage – can be reduced to 14% of cases by using the harvesting technique described by Pommer et al. (23) in 2008, which maintains an 8 mm safety margin from the incisor roots apex and the graft harvest site and 5 mm between the site and the mandibular base and mentonian foramen respectively. In the present case, no sensitive postoperative deficit was produced after bone harvesting.
Complete resection followed by rehabilitation by means of a bone graft does not eliminate the possibility of tumor relapse, which (as various Authors have affirmed) can affect the graft (24, 25). This is explained by the remnants of ameloblastoma, which can infiltrate the graft.

But to date, 7 years after complete resection of the tumor, our patient has not presented any signs or symptoms of relapse.

Conclusions

Esthetic and functional rehabilitation of the bone defects produced by tumor resection of a unicystic ameloblastoma, by means of a chin-harvested *en bloc* bone graft and dental implants constitutes a good therapeutic alternative in cases where the defect is of small size. *En bloc* resection of the UA is the treatment option with the lowest incidence of relapse although it is advisable to allow 5 years to pass between resection and rehabilitation using bone grafts, this being the period when most relapses occur.

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