3D CT Evaluation of Unicystic Follicular Type Ameloblastoma Treated By Demineralized Human Bone Graft: A Case Report¶

DEMİNERALİZE İNSAN KEMİK GREFTİ İLE TEDAVİ EDİLEN UNİKİSTİK FOLİKÜLER TİP AMELOBLASTOMANIN 3B-BT İLE DEĞERLENDİRİLMESİ: VAKA RAPORU

Melahat ÖĞÜTCEN-TOLLER*, Nergiz TAŞKAYA-YILMAZ*, Nilüfer ÇAKIR-ÖZKAN*, Emel UZUN*, Vildan KASAP*, Filiz KARAGÖZ**

* Ondokuz Mayıs Üniversitesi Diş Hekimliği Fakültesi Ağız, Diş, Çene Hastalıkları ve Cerrahisi AD,
** Ondokuz Mayıs Üniversitesi Tıp Fakültesi Patoloji AD, Samsun, TURKEY

Summary

Purpose: Ameloblastoma is the most common true odontogenic tumor of the jaws. They are usually diagnosed between the third and fifth decades of life. Mandibula is more affected than maxilla. Radiographically, ameloblastomas form rounded, well-defined, multilocular, cyst-like radiolucent areas with well-defined margins. Histopathologically, it is classified as follicular, plexiform, acanthomatous, granuler cell, desmoplastic, basal cell, clear cell ameloblastomas and papilliferous keratoameloblastomas. In this paper, a case of a large cystic ameloblastoma of follicular type is reported.

Case report: A 29- year old woman was referred to our clinic with a complaint of recurring pain and swelling over the left side of the mandible. Clinically, the left side of the mandible was swollen and it was tender on palpation. On radiographic examination, a well-defined, unicystic radiolucency associated with the crown of impacted mandibular third molar was found to extend from the left mandibular molar area to the subcondylar region. The tumour was excised and the diagnosis was made histopathologically.

Conclusion: In our case the large bone defect filled with demineralized human bone graft (DFDBA, Putty, Grafton) resulted in succesful ossification of the cavity. No recurrence was noted on long term follow up.

Key Words: 3D CT, Unicystic follicular type ameloblastoma, Demineralized human bone graft

T Klin J Dental Sci 2004, 10:43-48

Özet

Amaç: Ameloblastoma çenelerin en sık görülen gerçek odontojenik tümörüdür. Genellikle hayatın üçüncü veya dördüncü dekatında teşhis edilirler. Mandibula maksilladan daha sık etkilenir. Radyografik olarak iyi sınırlı, multiloküler, kist benzeri radyolüsent alanlar meydana getirirler. Histopatolojik olarak, folliküler, pleksiform, akantomatöz, granüler hücreli, dezmoplastik, bazal hücreli, clear hücreli ameloblastomalar ve papilliferöz keratoameloblastomalar olarak sınıflandırılır. Bu makalede, unikistik foliküler tip ameloblastoma rapor edilmiştir.

Vaka raporu: 29 yaşında bayan hasta kliniğimize mandibula sol tarafında tekrarlayan ağrı ve şişlik şikayetiyle başvurdu. Klinik olarak mandibulanın sol tarafında şişlik ve palpasyonda hassasiyet mevcuttu. Radyografik muayenede, gömülü mandibular üçüncü molar diş kronuyla ilişkili, sol mandibular bölgeden subkondiler bölgeye uzanan iyi sınırlı unikistik radyolüsent alan gözlendi. Tümör eksize edildi ve teşhis histopatolojik olarak konuldu.

Sonuç: Vakamızda, demineralize insan kemik grefti ile doldurulan büyük kemik defekti kavitenin kemikle dolmasıyla iyileşti. Hastanın uzun dönem takiplerinde nüks gözlenmedi.

Anahtar Kelimeler: 3B BT, Unikistik foliküler tip ameloblastoma, Demineralize insan kemik grefti

T Klin Diş Hek Bil 2004, 10:43-48

Ameloblastoma is the most common epithelial odontogenic tumour, comprising 1 % of tumours and cysts arising in the jaws (1,2). It is a locally aggressive odontogenic neoplasm in which ameloblastic differentiation is present (2,3). Ameloblastomas grow slowly and typically cause no symptoms until a swelling becomes noticable (4).

Radiographically ameloblastomas typically form rounded, cyst-like, radiolucent areas with

moderately well defined margins and characteristically appear multilocular (5).

Histopathologically, ameloblastomas are classified as follicular, plexiform, plexiform unicystic, acanthamatous, granuler cell, desmoplastic, basal cell, clear cell ameloblastomas and papilliferous keratoameloblastoma (4).

Based on the clinical and radiographic findings and histopathologic appearance, three categories of ameloblastoma have been designated to help manage patients because individual categories exhibit differences in behavior and recurrence rates and do not require the same extent of surgical management. The three categories or types are conventional, unicystic, and peripheral (extraosseous) (3).

Bone grafting is frequently used to augment bone healing with numerous approaches to reconstruction or replacement of skeletal defects (6).

Three-dimensional (3D) computed tomography (CT) is a valuable technique for the planning of skeletal, plastic and maxillofacial surgical procedures. Anatomic measurements can be made from three dimensional display images and manufactured life-sized models, and spatial relationships may be analyzed from desired perspectives (7).

In this article a case of unicystic follicular type ameloblastoma of the mandible treated by surgical excision and demineralized human bone graft placement using 3D CT reconstructions for pre-and postoperative assessment purposes is presented.

Case Report

A 29-year old woman was referred to our clinic with a complaint of recurring pain and swelling over the left side of the mandible. Antibiotics helped to alleviate the symptoms but they recurred after a while.

Clinically, the left side of the mandible was swollen and it was tender on palpation. Antibiotic therapy was started with 600 mg i.m. clindamycin for seven days followed by aspiration of the intralesional fluid from where was similar to pus.

On radiographic examination, a well-defined, unicystic radiolucency associated with the crown of impacted mandibular third molar was found to

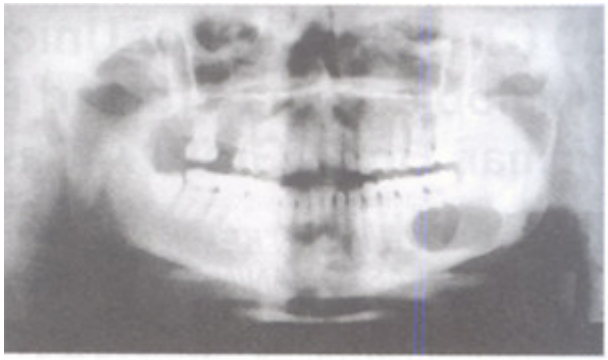


Figure 1. The cystic lesion extending from the left molar area to subcondylar region is seen on panoramic x-ray.

extend from the left mandibular molar area to the subcondylar region. The distal root of the mandibular first molar and both roots of the mandibular second molar were found to be resorbed on panoramic x-ray (Figure 1). Preoperative 3D CT reconstructions were made in order to obtain real dimensions and extent of the lesion (Figure 2). A provisional diagnosis of unicystic ameloblastoma with secondary infection was made.

The left mandibular first molar was treated endodontically. Since pathologic fracture of the mandible might occur intra or postoperatively, interdental wiring for a possible bimaxiller fixation

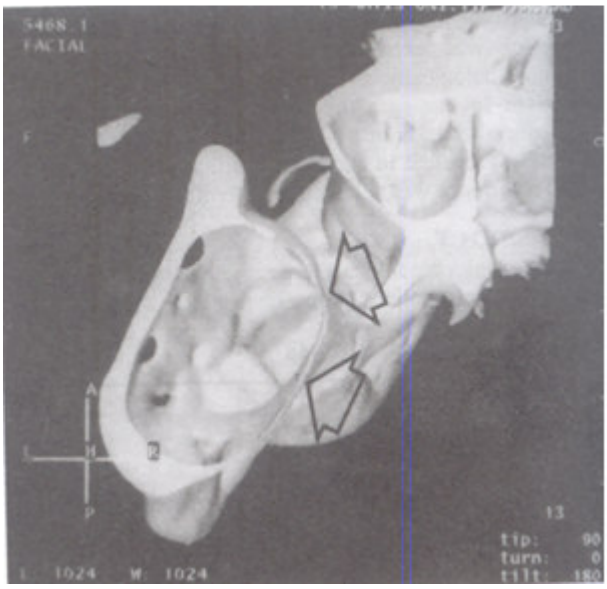


Figure 2. Preoperative 3D CT reconstruction of the lesion. Arrows show the lingual expansion.

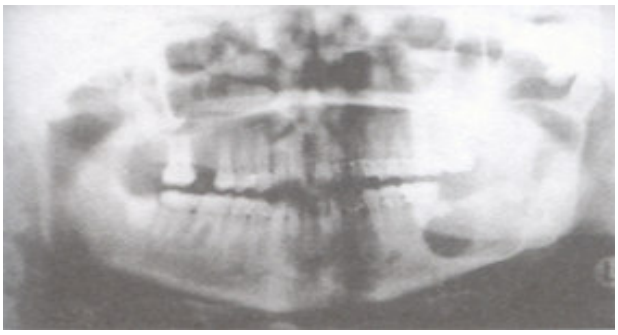


Figure 3. Preoperative panoramic x-ray of the patient after endodontic treatment of the left mandibular first molar and interdental wiring.

was made before operation (Figure 3). The cystic lesion was excised totally in a conservative manner under general anaesthesia with nasotracheal entubation. The impacted left mandibular third molar was removed in the same operation. The remaining large bone cavity was filled with 5 cc demineralized human bone graft (DFDBA, Putty, Grafton; Figure 4). The bone defect healed perfectly. The bone cavity reduced significantly after 3 months and it was filled with bone almost completely after one year on CT reconstructions (Figures 5,6). No recurrence was noted during the post operative 18 months period (Figure 7). The patient is still under long term follow-up.

Histologically, columnar ameloblastic tumor cells with reversed nuclear polarity surrounding loosely arranged polygonal cells resembling stellate reticulum within fibrous stroma in a follicular pattern were observed on the wall of the cystic lesion. Final diagnosis of mural ameloblastoma of follicular type was made (Figure 8).

Discussion

Ameloblastoma is a true neoplasm of enamel organ-type tissue which does not undergo differentiation to point of enamel formation (8). It is the most common neoplasm of jaws, ranging in occurence between 0.78 and 36,5 % (4,5,9,10). Men and women are equally affected (11). Eighty percent of ameloblastomas occur in the posterior mandible (5,12). In our case, the tumor was seated in the mandibular ramus and posterior body of the mandible. It is usually first recognised between the ages of 30 and 50. It is rare in children and old people (2,4,5,11). In developing countries ameloblastomas occur in younger people (11). Christos et al (13) found that the mean age at diag-

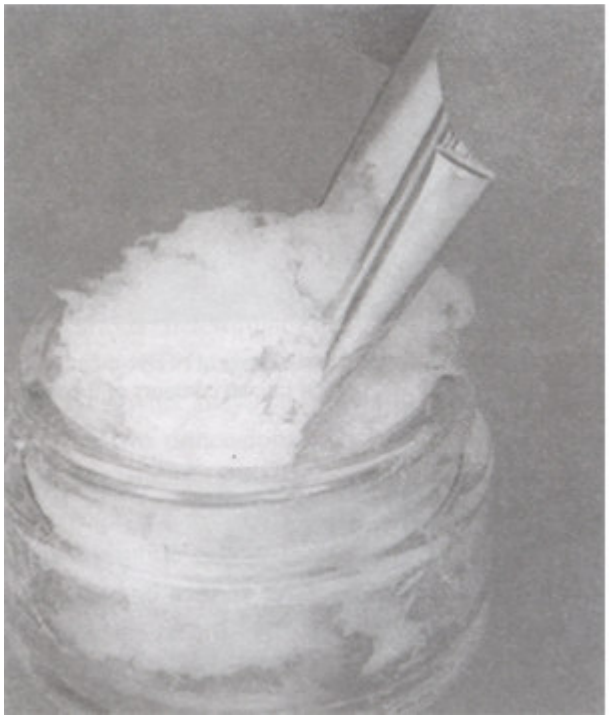


Figure 4. Demineralized human bone graft used for filling of the remaining bone cavity.

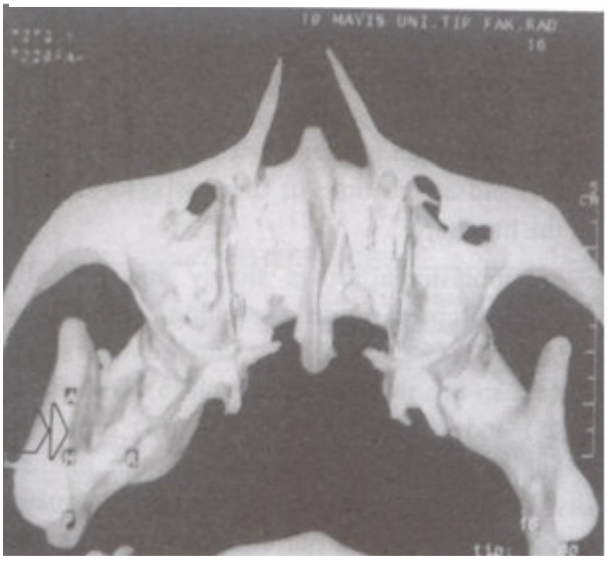


Figure 5. 3D CT reconstruction of jaws at three months after operation. Arrow shows the reduced bone cavity.

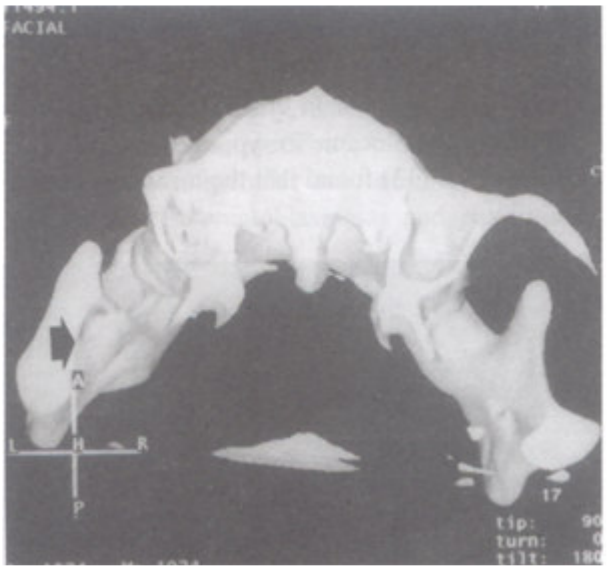
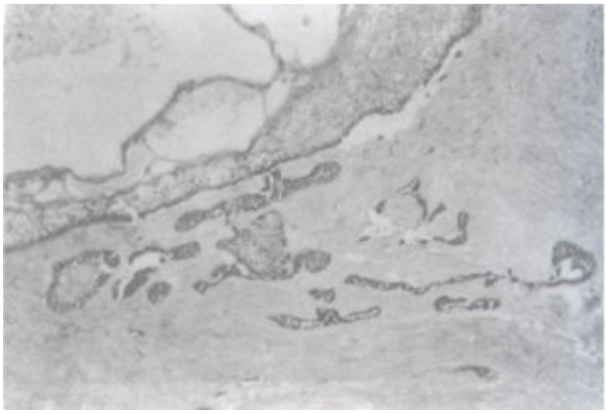


Figure 6. 3D CT reconstruction of jaws at twelve months after operation. Arrow shows normal lingual contours of the ramus, almost symmetrical to the other side.

Figure 7. Panoramic x-ray taken 18 month postoperatively shows complete bony healing.

nosis was 26 years in their series. Our patient was 29 years old at the time of diagnosis, in keeping with the literature.

Ameloblastoma grows slowly and typically causes no symptoms until a swelling becomes noticeable (2,4,14). Occasionally, small tumours may be first seen in a routine radiograph (5). Kim SG et al. (15) and Christos CG et al. (13) described swelling as the most common symptom in their series. If there is no secondary infection, the tumor is rarely painful (4). In our case, pain was caused by secondary infection and antibiotic treatment alleviated the pain.



Figures 8. Ameloblastic tumour cells surrounding stellate reticulum within fibrous stroma leads to the diagnosis of follicular ameloblastoma (H&Ex200 magnification).

Ameloblastoma have three categories as conventional, unicyctic and peripheral in terms of tumour behaviour. Unicystic type most often is discovered in the second and third decades of life (3), as was the case presented. In an analysis of cases of unicystic ameloblastomas, Gardner and Corio (16) reported that all formed in the mandible and males were affected twice as frequently as females. Its typical radiographic presentation is that of well-defined, unilocular radiolucency associated with the crown of an unerupted tooth, usually a mandibular third molar. The definitive diagnosis of the unicystic ameloblastoma requires correlation of the clinical findings of a cyst at the time of surgery and the histopathologic findings of unicystic structure lined by ameloblastic epithelium (3). The definition of unicystic ameloblastoma is important. It is based on two features: the lesion must be unilocular clinically and radiologically; secondly, it must appear on microscopic examination as a single cystic lesion with the epithelial lining consisting of ameloblastoma. Solid masses of tumor cells may also extend into the lumen, or islands of tumor may infiltrate the fibrous wall (4). Our case had similar clinical, radiological and histopathological properties.

It is generally believed that histological variations in ameloblastomas do not effect tumor behavior. Recurrence rate of follicular ameloblastoma was reported to be 29,5 % (4,5). However, more impor-

tant than histologic subtypes in terms of behavior, is whether the tumour is multi-or unicystic (4). Unicystic ameloblastoma can often be treated succesfully with less aggressive surgery than that needed for multicystic ameloblastoma (17).

Treatment is by wide excision, preferably taking up to 2 cm of apparently normal bone around the margin. Complete excision is curative but anything less is followed by recurrence (5). Conservative surgical management such as curettage has been used for the unicystic ameloblastoma, as the neoplastic epithelium is confined to the lumen of the cyst (3,12). For the same reason, follicular unicystic ameloblastoma was removed in a conservative manner with resulting success after 18 months in the case presented. Postoperative follow-up is most important in the therapy of ameloblastoma, because more than 50% of all recurrences occur within 5 years postoperatively (11). The recurrence rate for true unicystic ameloblastoma is approximately 14% (3). Because of this, our patient is on long-term follow-up and after 18 months, no recurrence has been detected.

Autogenous bone has been shown to be superior to allogeneic bone in reconstructing segments of bone; however, because of limitations such as the inadequate supply and donor-site morbidity, allografts were developed as an alternative (18-19). There are various types of allografts available, including freeze-dried bone allografts (FDBA) and demineralized freeze-dried bone allografts (DFDBA). DFDBA is the best-studied and most widely used allograft material because of its availability, safety and purported osteoinductive and osteoconductive properties (19). Experimental animal studies have shown that DFDBA has osteogenic potential (19-21). Clokie et. al. (20) reported that utilization of a demineralized bone matrix putty appeared to allow for complete closure of critical size calvarial defects in New Zeland white rabbits with viable new bone at 12 weeks. In our case, the large bone defect filled with DFDBA resulted in succesful ossification of the cystic cavity.

Preoperative radiographies have important role in treatment planning. The multicurved outline of the mandible is difficult to image with standart roentgenographic modalities such as panorex display, however, the panorex lacks depth representation and three-dimensionality. Preoperative CT scan is imperative in enabling the surgeon to make the important decision of whether to curette and freeze or to resect. CT's capability to simultaneously display bone and soft tissues has significantly advanced the diagnosis of maxillofacial neoplasms (22). Cohen et al. (23) advocated the use of preoperative CT scans in 1985. Both bone and soft tissue windows are helpful in evaluating cortical thinning, perforation, and soft tissue involvement (12). Determination of the extent of such a lesion is significantly facilitated with the three-dimensional display (22). 3D CT is advantageous in that it allows simulation of surgery and various determinations of skeletal measurements (24). 3D images are of particular value to the clinician, who may be uncomfortable analysing multiple two dimensional axial or coronal CT images. The ability to view an area of interest in three dimensions allows both clinician and radiologist to examine subtle points of anatomy in various perspectives and assess spatial relationships (7). Three dimensional reconstruction of the mandible can be used in planning both ablative and reconstructive procedures of the mandible and other maxillofacial structures, thus optimizing presurgical planning (22). Therefore, we used 3D CT for preoperative surgical planning and postoperative follow-up period. In conclusion, unicystic ameloblastoma was best evaluated using 3D CT reconstructions for diagnosis, surgical planning and postoperative examination purposes. Besides, 3D CT reconstructions provided best view of the healing of the bony defect filled with DFDBA.

REFERENCES

1. Choudhury M, Dhar S, Bajaj P: Primary diagnosis of ameloblastoma by fine-needle aspiration:a report of two cases. Diagn Cytopathol Dec 23:414, 2000
2. Günhan Ö: Oral ve Maksillofasiyal Patoloji, Atlas Kitapçılık, Birinci Baskı, Ankara, 2001,p.42
3. Neville BW, Damm DO, White DK: Color Atlas of Clinical Oral Pathology. Second Edition, USA,Williams & Wilkins, 1999, p. 400
4. Cawson RA, Binnie WH, Speight PM, Barrett AW, Wright JM.: Lucas's Pathology of Tumors of the Oral Tissues. Fifth Edition, London, Churchill Livingstone, 1999, p. 25

5. Cawson RA, Odell EW: Essentials of Oral Pathology and Oral Medicine, In: Odontogenic tumours and tumour-like lesions of the jaws. Sixth Edition, London, Churchill Livingstone, 1998, p.117
6. Gozdog AR, Lone JM, Glaser D, Farster RA: Alternatives to autogenous bone graft: efficacy and indications. J The American Academy of Orthopaedic Surgeons 3: 1, 1995
7. Schellhas KP, El Deeb M, Wilkes CH, Check RK, Larsen JW, Heithoff KB, Fritts HM: Three-dimensional computed tomography in maxillofacial surgical planning. Arch Otolaryngol Head Neck Surg 114: 438, 1988
8. Shafer WG, Hine MK, Levy BM: A Textbook of Oral Pathology, In: Disturbances of development and growth cysts and tumors of odontogenic origin. Fourth Edition, Philadelphia, Sounders Company, 1983, p.276
9. Ackermann GL, Altini M, Shear M: The unicystic ameloblastoma: a clinicopathological study of 57 cases. J Oral Pathol 17:541, 1988
10. Martins RH, Andrade SJ, Rapoport A, Rosa MP: Histopathologic features and management of ameloblastoma: study of 20 cases. Sao Paulo Med J Jul1; 117: 171, 1999
11. Reichart PA, Philipsen HP, Sonner S: Ameloblastoma: Biological profile of 3677 cases. Eur J Cancer B Oral Oncol 2:86, 1995
12. Sampson DE, Pogrel MA: Management of mandibular ameloblastoma: the clinical basis for a treatment algorithm. J Oral Maxillofac Surg. Sep; 57: 1074, 1999
13. Christos CG, Yoo MC: Ameloblastoma: clinical features and management of 21 cases. Ethiop Med J 38: 247, 2000
14. Gardner DG: Critique of the 1995 review by Reichart et al. of the biologic profile of 3677 ameloblastomas. Oral Oncol 35: 443, 1999
15. Kim SG, Jang HS: Ameloblastoma: a clinical, radiographic and histopathologic analysis of 71 cases, Oral Surg Oral Med Oral Pathol Oral Radiol Endod Jun 91: 649, 2001
16. Gardner DG, Corio RL: The relationship of plexiform unicystic ameloblastoma to conventional ameloblastoma. Oral Surg Oral Med Oral Pathol. 56: 54, 1983
17. Olation AA, Adekeye EO: Unicystic ameloblastoma of the mandible: a long term follow-up. J Oral Maxillofac Surg. 55: 345, 1997
17. Goldberg VM, Stevenson S: The biology of bone grafts. Seminars in Artroplasty. 4: 58, 1993
18. Rosenberg E, DipDent H, Rose LF: Biologic and clinical consideration for autografts and allografts in periodontal regeneration therapy. Dental Clinics of North America. 42: 467, 1998
19. Nasr HF, Aichelmann Reidy ME, Yukna RA: Bone and bone substitutes. Periodontology J 19:74, 2000
20. Clokie CM, Moghadom H, Jackson MT, Sandor GK: Closure of critical sized defect with allogenic and alloplastic bone substitutes. J Craniofac Surg 13: 111, 2002
21. Sayan NB, Akal ÜK, Pektaş ZÖ, Şahin M, Acar E: demineralize kemik grefti ve collagrafttm kemik greft materyali ile yapılan bir klinik ve densitometrik çalışma. Diş Hekimliği Bilimleri Dergisi, 8: 134,2002
22. Zinreich SJ, et al: CT for cranial facial and laryngeal surgery. Laryngoscope 98: 1212, 1998
23. Cohen MA, Hertzanu T, Mendelsohn DB: Computed tomography in the diagnosis and treatment of mandibular ameloblastomas: report of cases. J Oral Maxillofac Surg. 796, 1985
24. Ono I, et al: Three-dimensional analysis of craniofacial bones using three-dimensional computer tomography. J Cranio Maxillofac Surg 49, 1992

Geliş Tarihi:12.06.2003

Yazışma Adresi: Dr. Nergiz TAŞKAYA-YILMAZ
OndokuzMayısÜniversitesi
Diş Hekimliği Fakültesi
Ağız, Diş, Çene Hastalıkları ve Cerrahisi AD,
SAMSUN
nergizy@omu.edu.tr

¶This case report was presented in XVIth Congress Europan Association for Cranio-Maxillofacial Surgery 3-7 September 2002, GERMANY.