

A CLINICAL CASE OF SPONTANEOUS MANDIBULAR REGROWTH AFTER SEGMENTARY RESECTION AND FOLLOW-UP OF INFERIOR ALVEOLAR AND CERVICO-FACIAL NERVES RECOVERY

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Abstract: The authors present a case of mandibular complex odontoma with adjacent aneurysmal cysts with clinical findings easily confounding with ameloblastoma in which a particular surgical approach was performed. The young age of the patient and the well confined tumour border allowed preservation of the inferior alveolar nerve, which had an intralesional course, by neuroraphy after tumour resection. The postoperative paresis of the marginal branch of facial nerve was monitored and revealed an incomplete nerve recovery after the second surgery done for removing the Ti reconstruction plate.

INTRODUCTION

Odontomas are tumours of odontogenic origin (1) which combine both epithelial and mesenchymal traits and represent about 22% of the odontogenic tumours found in the jaws.(2) Odontomas are considered hamartomatous lesions (3,1) (developmental anomalies) resulting from completely differentiated epithelial and mesenchymal cells that give rise to ameloblasts and odontoblasts.(3) Odontomas are mostly intrabony lesions, though in very few cases they may spontaneously erupt into the oral cavity.(3) Odontomas are often found in conjunction with permanent dentition and very rarely associated with the teething and most of them are discovered during the second decade of life.(4) Pathological changes, such as impaction, malpositioning, aplasia, malformation and devitalization of adjacent teeth are associated with 70% of odontomas. In general, they grow asymptotically and are detected incidentally by routine radiographs.(5)

Two histological types of odontomas have been described: compound and complex.(6)

A complex odontoma is a hamartomatous lesion or malformation of odontogenic origin in which both epithelial and mesenchymal cells exhibit complete differentiation (all the dental tissues are represented (7) – but occurring in a more or less irregular pattern).(8) The compound one consists in a different size teeth crowding mass inside the maxillary bone. On radiographic images, the complex odontoma looks like a calcified, apparently amorphous mass, with a tooth-like radiodensity.(1) When a subjacent impacted tooth is present, there has to be decided if it can be orthodontically pulled-out on the dental arch.(1)

CASE REPORT

Patient, P.G., male, 13 years old, was referred to the Oral and Maxillofacial Surgery Department of Emergency Military Hospital Sibiu by a dentist who noticed a presumable odontogenic tumour in the right mandible, on a routine OPT.

The patient was asymptomatic, except for the lack on the dental arch of permanent molar 4.7., which normally comes out at the age of 12.

Due to the undistinguishable mandibular canal on the OPT in the tumour site, a cone-beam CT was done in order to

visualize the inferior alveolar nerve relation to the tumour.

Figure no. 1*. The OPT of patient, P.G., 13 years old, at first consult in Nov. 2014: a mixed radiolucence-radiopaque image in the right mandibular angle with well defined polycyclic contour, in relation with the partial mineralized crown of 4.7., suggesting an odontoameloblastoma (with permission of Professor Dr. Viorel Ibric Cioranu, Dr. Radu Ioan Neacșu)



Figure no. 2*. Preoperative CT (Nov. 2014): Inferior alveolar nerve enters the tumour mass and becomes unperceivable



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The CT showed the mixed structure of the tumour with alternating radiolucencies and radioopacities. The inferior alveolar nerve relationship to the tumour was not clearly identifiable even on the CT: the nerve course is lost in the tumour mass and comes again clear distally in the 4.6. region. The 3D CT highlights the tumour bone effraction in the mandible angle region. The radiologic findings suggest an odontogenic tumour, possibly an ameloblastoma or a related histological form. Due to the extensive growth of the tumour in the mandibular angle and also to the highly recurrence rate of the ameloblastic tumours, the therapeutic first aim option was segmentary resection of the right mandibular angle.

Actually, considering such an odontogenic tumour does not infiltrate but only pushes out the soft tissues, i.e. the inferior alveolar nerve, it was analyzed the possibility of preserving the nerve when performing the bone resection. Therefore, the surgical approach was planned to use a piezoelectric device for the bone section proximally and distally to the tumour, hence sparing the inferior alveolar nerve. After dividing the nerve at the exit from the tumour, its proximal end was going to be pulled out of the tumour and sutured to the distal end.

The surgical intervention was done in Dec. 2014 under general anesthesia, using a cervical approach. The incision was placed 3 cm under the mandibular body, then the marginal mandibular branch of the facial nerve was identified. The pterygo-masseterin belt was divided, then the periosteum and muscular insertions on both sides of mandibular angle and body anteriorly to the mental foramen, were dissected.

Figure no. 3*. Cervical approach for tumour resection: it can be seen the thinning of the external cortical plate by the tumour in the right mandibular angle and body



The 4.6. molar was extracted, in order to perform the osteotomy of the mandibular body. The osteotomy of the mandibular ramus (proximal site) and body (distal site) was performed using a piezoelectric device with 1 cm safety margin to the tumour, sparing the inferior alveolar nerve. The nerve was then exposed on a 1,5 cm length at the both osteotomy sites. Before nerve sectioning, neuroraphy axial guiding sutures through the perinerve were done. The inferior alveolar nerve was divided at the distal osteotomy site between the guiding sutures, then the bony resection piece was unmolded off the proximal end of the nerve and submitted to the histopathologist.

After the segmental bony resection mandibular reconstruction was performed using an angled Ti plate fixed by osteosynthesis screws to the ramus and body. Then the inferior alveolar nerve neuroraphy was performed with 3 points perineural sutures.

Figure no. 4*. During the mandibular body osteotomy, the inferior alveolar nerve was identified



Figure no. 5*. The inferior alveolar nerve was also identified at the mandibular ramus osteotomy site



Figure no. 6*. The mandibular resection specimen – lateral view – without the inferior alveolar nerve that was preserved by pulling it out of the tumour and suturing to the distal end.



Figure no. 7*. The bony resection specimen – internal view



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Figure no. 8*. Immediate mandibular reconstruction with angled Ti plate: the inferior alveolar nerve ends are held together for neuroraphy



Figure no. 9*. Intraoperative view after Ti plate reconstruction and neuroraphy



The immediate postoperative healing was uneventful, with no complications. The patient presented for a 6 month period the right inferior lip and chin anesthesia and also the paresis of the mandibular marginal branch of the facial nerve which was initially assessed as VI-th degree on House-Brackmann adapted scale. Both inferior alveolar nerve territory anesthesia and cervicofacial paresis regressed without neurotrop treatment; after 6 months, the cervicofacial paresis was reassessed as III-rd degree.

Figure no. 10*. Clinical aspect 7 month after the first surgical procedure: II-nd degree paresis of the right mandibular marginal facial branch on House- Brackmann adapted scale

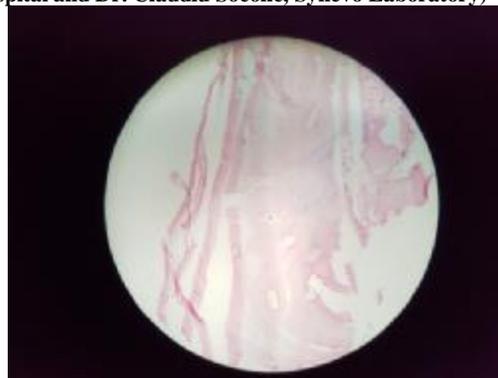


The initial histological examination of the specimen pointed the diagnosis of osteoma, which came into disagreement with the clinical and imagery findings (lack of molar 4.7. on dental arch, radiographic image of the tumour which comprises

tooth like structures). We asked for re-examination of the resection specimen and the histological diagnosis was rectified to complex odontoma.

Nevertheless, for an accurate diagnosis, considering the radiographic image very conclusive for an ameloblastoma, we also asked for a immunohistochemical examination, which confirmed the diagnosis of complex odontoma and associated secondary aneurysmal bony cysts. The IHC tests showed: CK 19-positive in limited epithelial odontogenic membranal areas; it does not reveal odontogenic epithelium on the inner surface of the bony cysts; CALRET-negative in the odontogenic epithelium (Dr. Claudiu Socoliuc, Synevo Laboratory)

Figure no. 11*. Histopathological view of the specimen (Dr.Ioan Sorin Zaharie, Sibiu Clinical County Emergency Hospital and Dr. Claudiu Socolic, Synevo Laboratory)



Histological sections show cortical and spongy bone tissue including a well defined odontogenic formation bordered by a thin vascular connective capsule. There can be seen papillary vascular –connective invaginations with a dental papilla appearance, which are bordered by odontoblasts; displazic dentin, cement and enamel. Inside the tumour and in the adjacent bony tissue there can be seen cystic interwired spaces with a sero-hematic content and without epithelial bordering. (Dr. Claudiu Socolic, Synevo Laboratory)

At the clinical reassessment 6 months postoperatively, one could also notice the already described neurosensitive features, a light asymmetry of the inferior third of the face due to the right mandibular angle proeminency and the left deviation ~ 4 mm of the inferior medial line.

Figure no. 12*. Clinical frontal view 6 month after the first surgery: light facial asymmetry due to right mandibular angle proeminency



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Figure no. 13*. Clinical right lateral view 6 month after the first surgery: postoperative scar after segmentary resection of right mandibular angle and primary reconstruction with Ti plate



Figure no. 14*. Intraoral aspect 6 month after the first surgery: then could be observed the superior lateral incisors anodontia and the postoperative leftwards mandibular deviation ~ 4 mm



The patient was radiologically re-examined at 4 and 6 month after the first surgery. The consecutive OPT-s revealed the spontaneous progressive mandibular regrowth through the preserved periosteal tunnel along and above the rigid stabilization Ti plate. The newly regenerated bone finally completely filled the mandibular gap.

Figure no. 15*. OPT – Feb. 2015 at 4 months after the first surgery. There can be seen the bony-like radioopaque structure in the tumour resection site and the newly formed mandibular canal



Figure no. 16*. OPT – May 2015 at 6 month after the first surgery reveals the normal structure increasing radioopacity in the tumour resection site with normal bone structure on whole width of the mandibular angle and body



Considering the obvious mandibular regrowth on radiological exams, in order to allow the future correction of the slight facial asymmetry, a second surgery was planned for removing the Ti plate.

The second surgery approach was mixed oral and cervical and revealed the newly regenerated bone in the segmental resection site.

Figure no. 17*. Intraoperative view: spontaneously regenerated bone in the right mandibular angle region



Figure no. 18*. Intraoperative view (the second surgery June 2015): right mental nerve foramen



After the second surgery, the patient presented the worsening of the mandibular marginal branch paresis probably due to its elongation during the operation. This time the paresis showed a longer recovering time. The paresis of this facial branch was assessed as VI-th degree 1 year after the second surgery.

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DISCUSSIONS

This case implied particular difficulties regarding diagnosis, conservatory surgical approach, spectacular spontaneous bone regeneration and partial motor and sensitive nerve recovery. The clinical and imagistic findings (OPT, CT) at the first consult advocated the diagnosis of ameloblastoma as a related histological entity (lack on the dental arch of molar 4.7., the “bubbles” radiological image with dental surrounding structures strongly suggesting an ameloblastic fibroodontoma). Finally, the histological examination concluded to an associated entity of complex odontoma and aneurismal bony cysts. Due to the tumour size involving the whole mandibular width, and also to the major probability of an ameloblastoma, it was chosen a radical surgical therapy consisting in mandibular segmental resection.

The mandibular segmental resections in case of ameloblastic tumours generally do not respect the inferior alveolar nerve. Yet in this case considering the young age of the patient and the soft tissue non-invasive feature of the ameloblastomas, there was performed a modified segmental mandibular resection in which the continuity of the inferior alveolar nerve was re-established. The conservative operative attitude regarding the inferior alveolar nerve was finally proved to be optimal, the complex odontoma being known to have an insignificant recurrence rate.

Preserving of the mandibular periosteal sleeve and the inferior alveolar nerve continuity, providing in the same time a good stability of the bony ends by the Ti reconstruction plate and lastly the young age of the patient were probably the key points which allowed the mandibular spontaneous regrowth along with a newly formed mandibular canal.

The good recovery without any adjuvant medication of the facial marginal mandibular branch after its first operation counted on lack of a major nerve injury and on the patient young age.

The slower evolution to an incomplete recovery of the same nerve after the second operation points out the high risks of functional sequellae of the facial nerve branches when performing a second operation in the nerve territory.

CONCLUSIONS

The surgical approach for inferior alveolar nerve repair reported in this case of mandibular segmentary resection proved to be efficient in ensuring spontaneous bone regeneration around a newly formed mandibular canal.

The surgical reentries in the territory of the facial nerve branches have a high risk of irreversible nerve injury.

**Casuistry of Professor Dr. Viorel Ibric Cioranu, Dr. Radu Ioan Neacșu - Sibiu Military Emergency Hospital.*

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