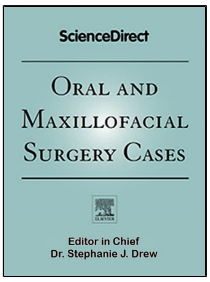
# Accepted Manuscript




Mandibular coronoid process tumor resembling a mandibular condyle: a case report

Cimino Roberta, Piombino Pasquale, De Riu Giacomo, Mascolo Massimo, Vollaro Stefano, Valletta Rosa, Michelotti Ambrosina, Califano Luigi

| | |
|---|---|
| PII: | S2214-5419(18)30082-8 |
| DOI: | 10.1016/j.omsc.2018.10.007 |
| Reference: | OMSC 85 |
| To appear in: | *Oral and Maxillofacial Surgery Cases* |
| Received Date: | 02 July 2018 |
| Accepted Date: | 24 October 2018 |

Please cite this article as: Cimino Roberta, Piombino Pasquale, De Riu Giacomo, Mascolo Massimo, Vollaro Stefano, Valletta Rosa, Michelotti Ambrosina, Califano Luigi, Mandibular coronoid process tumor resembling a mandibular condyle: a case report, *Oral and Maxillofacial Surgery Cases* (2018), doi: 10.1016/j.omsc.2018.10.007

# Mandibular coronoid process tumor resembling a mandibular condyle: a case report.

*Cimino Roberta[1], Piombino Pasquale[2], De Riu Giacomo[3], Mascolo Massimo[4], Vollaro Stefano[5], Valletta Rosa[6], Michelotti Ambrosina[7], Califano Luigi[8].*

1 Roberta Cimino, DDS,

Assistant Professor,

Section of Orthodontics andGnathology Department Neuroscience, Reproductive Sciences and Oral Sciences, University of Naples Federico II, Via Pansini 5, 80131 Naples, Italy. rocimino@unina.it

2 Pasquale Piombino. MD, PhD, FEBOMFS

Assistant Professor

ENT Department, University of Campania "Luigi Vanvitelli", Via Pansini 5, 80131, Naples, Italy. pasquale.piombino@unicampania.it

3 Giacomo De Riu, MD, PhD, FEBOMFS

Assistant Professor

Operative Unit of Maxillofacial Surgery, University of Sassari, Viale San Pietro 43B, 07100 Sassari, Italy.gderiu@uniss.it

4 Massimo Mascolo, MD, PhD,

Clinical consultant,

Department of Advanced Biomedical Sciences, Pathology Section, University of Naples Federico II, Naples, Italy. mmascol@gmail.com

5 Stefano Vollaro , MD,

Visiting Professor,

Section of Orthodontics andGnathology Department Neuroscience, Reproductive Sciences and Oral Sciences, University of Naples Federico II, Via Pansini 5, 80131 Naples, Italy. stefano.vollaro@unina.it

6 Rosa Valletta, DDS,

Associate Professor,

Section of Orthodontics andGnathology Department Neuroscience, Reproductive Sciences and Oral Sciences, University of Naples Federico II, Via Pansini 5, 80131 Naples, Italy. valletta@unina.it

7 Ambrosina Michelotti, DDS,

Associate Professor,

Section of Orthodontics andGnathology Department Neuroscience, Reproductive Sciences and Oral Sciences, University of Naples Federico II, Via Pansini 5, 80131 Naples, Italy. michelo@unina.it

8 Luigi Califano, MD,

Full Professor,

Department Neuroscience, Reproductive Sciences and Oral Sciences, University of Naples Federico II, Via Pansini 5, 80131 Naples, Italy. califano@unina.it

***Correspondence to:***

Cimino Roberta, DDS, Assistant Professor, Department Neuroscience, Reproductive Sciences and Oral Sciences, University of Naples Federico II, Via Pansini 5, 80131 Naples, Italy

Tel:+393393026678, +390817464980; email: rocimino@unina.it

***Abstract***

Abnormal elongation of mandibular coronoid process, often defined as coronoid hyperplasia, is a rare condition, which is frequently associated with limited mouth opening. In some cases, the enlarged coronoid pushes the zygoma forward causing facial asymmetry. This case report describes a 16-year-old boy whose chief complaint was a progressive difficulty and deviation in mouth opening, together with a deformity appearing at maximum opening at the zygomatic area. The diagnosis was Unilateral Accessory Mandibular Condyle at coronoid process, without reduction of the mouth opening capacity. A coronoidectomy was carried out by means of piezoelectric surgery, instead of a coronoidotomy which is usually performed in these cases, due to a suspect of ramus neoplasm.

***Keywords***

Coronoid hyperplasia, Accessory Condyle, Temporomandibular Disorder, Piezoelectric surgery, Adolescent

***Introduction***

Accessory Mandibular Condyle is a rare anomaly that sometimes arises from the mandibular coronoid process, much like a coronoid process hyperplasia (Siddikaand Ferdousi, 2016). This condition usually causes slow and progressive reduction of mouth opening (Gibbson, 1995; Fabie et al, 2002) because of the impingement of the enlarged coronoid processes against the medial surfaces of the body of the zygomatic bones. It generally occurs at puberty and continues over the years. It can be associated with facial asymmetry.

Etiology includes temporalis muscle hyperactivity, trauma and neoplasia, but in many cases there is no clearly identifiable pathogenesis. Some authors suggested that the coronoid reactive elongation depends on the increased pull of the temporalis muscle (Sarnat and Engle, 1951; Isberg et al, 1987), or that bony hypertrophy of the coronoid process and mandibular angle could be the result of a prolonged and significant contraction of the masticatory muscles (Murakami et al, 2000). Unilateral Coronoid Hyperplasia can also be the consequence of a trauma or a pathologic condition such as a neoplasm (eg. Ostechondroma) (Tucker et al, 1984; Sawada et al, 2015). A pseudo-joint between the coronoid process and the zygomatic arch due to an osteochondroma was first described by Jacob in 1899 (Ajila et al, 2012). The differential diagnosis of coronoid hyperplasia includes all causes of ankylosis and pseudo ankylosis of the temporomandibular joint, in which an overgrowth of the coronoid process causes a pseudo-joint formation with the zygomatic arch. (Losa-Munoz et al,2014; Sawada et al, 2015; Dandryial et al, 2015).

The diagnosis can be made with panoramic x-ray imaging, which is a widely available screening modality, or via 3D computed tomography (Izumi et al, 2005; Akan and Mehreliyeva, 2006; Peck et al, 2014)).

As Mulder reported in a systematic review there is no significant incidence and prevalence regarding Coronoid Process Hyperplasia (CPH) (Mulder et al, 2012). The mean age of patients affected by CPH is 23 years and the age distribution has a peak between 15 to 19 years. Usually there is a time frame of approximately 7 years between disease onset and diagnosis. The condition can be uni or bilateral, with the bilateral form occurring 4.1 times more frequently than the unilateral one, and with a male–to-female ratio of 3.3 to 1. (McLoughin et al, 1995, Mulder et al, 2012).

Several surgical approaches have been proposed such as extraoral/intraoral coronoidectomy along with a histopathological examination in order to determine the nature of the specimen. As Mulder reported in his review the therapeutic approach was a coronoidectomy in 84% of the cases (Mulder et al, 2012).

This is a report of a case with non-neoplastic accessory mandibular condyle located at the coronoid process without limitation of mouth opening, treated with transoral piezoelectric surgery.

***Case report***

A 16-year old boy was referred to the Section of Temporomandibular Disorders and Orofacial Pain of the University of Naples Federico II, complaining about a progressive difficulty and deviation in mouth opening, with a slight face deformity appearing during maximum opening at the zygomatic area.

Two years prior to his first examination, the patient had a facial trauma and referred for a progressive development of a mandibular deviation during mouth opening. The mandibular range of motion was as follows: active mouth opening 60mm, passive opening 62 mm, right lateral movement 11 mm, left lateral movement 13 mm, protrusion 6 mm. The patient did not report any masticatory muscle pain, nor TMJ pain or facial pain, both spontaneous and at palpation. The opening pattern showed an uncorrected deviation of about 2 mm towards the left side (fig.1). A slight facial asymmetry, with swelling at the right zygomatic region, appeared at maximum opening.

During all mandibular movements, a clicking noise was reported by the patient and recorded during clinical examination on the right side, and a small bump in the right malar zone was observed during opening.Intraoral examination showed an Angle class I occlusion. The patient sometimes complained of low back pain. During clinical examination, an overall body hyperlaxity was evident with a Beighton index of 9. There was no facial or trigeminal nerve involvement and no history of TMJ open locks or dislocations.

Panoramic x-ray, prescribed during a previous consultation one month earlier (fig.2), revealed a quite normal shape of the right coronoid process, and normal-appearing condyles located in the glenoid fossa. In order to better evaluate the morphology of the coronoid process, a coronal and axial computed tomography (CT) was requested. CT confirmed a large bony overgrowth arising from the right coronoid process. This atypical right coronoid process was projected laterally from the mandibular ramus with an angle of about 30° (fig.3,4). This part of the bone was morphologically similar to a condylar head, without any impingement with the zygomatic arch. In open mouth position, a hemifacial soft tissue displacement was observed, due to the right coronoid process atypical growth. This deformity was not observed in closed mouth position.

The patient and his parents were informed about the diagnosis and walked through the surgical treatment protocol, including the excision of the accessory condyle.

*Surgical procedure*

Based on clinical and radiographic images, a provisional diagnosis of Coronoid Hyperplasia was made. Treatment protocol included resection of the accessory bone with a piezoelectric device.

The patient was placed in a supine position under general anesthesia with naso-tracheal intubation. After infiltration of local anesthetic and vasoconstrictor in the upper right hemimandibular vestibular fornix region, a cold

knife incision of the mucosa anterior to the ramus was performed. The periosteum was then elevated from the anterior border of the ramus to the outer surface of the middle part of the ramus, upward to the lingual and lateral surface of the coronoid process, 1 cm above the sigmoid notch.

Temporalis muscle attachment was partly preserved. A retractor was inserted from the outer side of the ramus and a malleable retractor was placed lingually to protect the soft tissues and the inferior neurovascular bundle when cutting the coronoid process.

Piezoelectric surgery was performed with an osteotomy from the sigmoid notch to the anterior border of the ramus, parallel to the inferior border of the mandible. The removed bony specimen appeared very similar to a condylar head and was covered by a white and smooth surface (fig.5,6). Extraoral pressure dressing was placed over the skin above the ramus to prevent severe swelling after surgery. Histology revealed a non-neoplastic fibrous perichondrium covering a hyaline cartilage cap in continuity with the periosteum of the underlying bone (FIG.9a,b). A diagnosis of osteocartilaginous exostosis was made.

*Post-surgical management*

No significant differences in the patient aspect were highlighted before and after surgery. In the early postoperative period the patient's maximum mouth opening (MMO) was reduced. Home exercises were therefore prescribed after surgery (Michelotti et al, 2005; Durham J et al, 2016).

After 1 month of exercises, MMO gradually improved from 27mm to 38mm. After 6 months, the active mouth opening was 51mm and the passive one 53 mm, without any difficulty in opening/closing pattern.

A control CT scan was performed four months after surgery and no osteo-muscular alterations were detected. Histology revealed an osteocartilaginous exostosis (fig.7a,b), in agreement with the clinical diagnosis of coronoid hyperplasia, like in Jacob's disease.

***Discussion***

Mandibular development abnormalities, like CPH and accessory condyle, are rare disorders of uncertain etiology that can occur in both unilateral and bilateral forms. Although CPH has been considered an uncommon condition, its actual incidence is expected to be higher than that reported in the past, taking into account the existence of many unreported and/or undiagnosed cases. Most common sign, in unilateral or bilateral cases, is a facial asymmetry with a deformation of the zygomatic arch, accompanied by a slow and progressive reduction in mouth opening with no reported pain (McLoughin et al, 1995, Mulder et al, 2012).

In the case here reported the patient was concerned about progressive deviation during the mouth opening pattern, with a slight face deformity at the zygomatic area, and without ankylosis between the ramus and the arch, neither any reduction of mouth opening.

CPH can be easily diagnosed if the patient presents a limited mouth opening (Izumi et al, 2005; Mazzetto and Hotta, 2007; Tavassol et al,2012; Mulder et al, 2012). Indeed, the CPH diagnostic criteria according to the expanded taxonomy for temporomandibular disorders (Peck et al, 2014), include a progressive enlargement of the coronoid process that impedes mandibular opening due to the zygomatic process mechanical obstacle. The patient usually reports a history of jaw opening limitation. During clinical examination there should be a reduction of active and passive maximum jaw opening, imaging (panoramic x-rays or CT/CBCT) should show an elongated coronoid process which comes into close contact with the posterior aspect of the zygomatic process of the maxilla during opening.

The novelty of this case is that the patient did not present any reduction in mouth opening and the diagnosis was not made at an early stage. Thus in the patient previous panoramic x-ray, a quite normal shape of the right coronoid process was evident, although it was not clearly represented.

The very special shape of coronoid process, as shown in the CT images, was defined as accessory condyle (Siddika & Ferdousi, 2016), since it was covered with cartilage, as reported by the histopathological report. The cartilaginous component was a typically condylar characteristic, which does not normally belong to the coronoid process.

The histological report could be consistent with a Jacob's disease (Akan and Meherliyeva N, 2006; Sreeramanemi et al, 2011;Ajila et al, 2011). The coronoid shape, his relationship with zygomatic arch and the absence of a joint with the arch, and the coronoid appearance to a mandibular condyle, like a duplication of a portion of the facial skeleton (Peacock et al, 2001) induced us to name this clinical condition as an accessory condyle on coronoid process.

Hyperplastic coronoid process without impingement to the zygomatic bone could be followed through a slight elongation (Izumi 2005), but causal relationship of the morphological changes is uncertain. In this case it could be speculated that the previous trauma lead to the unexpected elongation and atypical shape of the coronoid process.

The aim of a coronoidectomy, which removes the coronoid process completely, is to relieve any interference between an elongated coronoid process and the zygoma, or an articulation between them, as well as to prevent the upper displaced coronoid process from interfering with the movement of the mandible by contacting the upper part of the ramus. However, coronoidectomy needs wider tissue reflection, which causes more tissue damage and creates more scarring after surgery, and which may cause trismus to occur again. It is usually difficult to perform coronoidectomy if articulation of the coronoid process and zygoma exists. Some surgeons approach this by extraoral approach to ensure complete coronoidectomy combined with myotomy, fasciotomy, and zygomatic osteotomy.

Coronoidotomy was conventionally used (Gerbino et al, 1997) for CPH treatment with good results, frequently creating a gap by upward displacement of the coronoid process after coronoidotomy, so as to prevent reunion of the two bone segments, and avoid probable relapse. The gap coronoidotomy can also be applied (Chen MC et al, 2011) when separation between coronoid process and the zygoma is difficult, or when complete removal of the prominent deformed coronoid process is very difficult.

In the present case the coronoid process was spontaneously elongated with a curve that allowed it to exceed the zygomatic arch, thus avoiding the impaction during mouth opening.

A piezoelectric surgery, was adopted, proceeding to a coronoidectomy, instead of the coronoidotomy, that was normally used in these cases, due to a suspect of ramus neoplasm (Vercellotti, 2004; Beziat et al, 2007; Robiony et al, 2007; Gonzales and Mareque, 2008; Vercellotti et al, 2005; Landes et al 2008a, 2008b, D'Amato et al, 2014; Chiarini et al, 2014)

***Conclusion***

An accessory condyle originating from the coronoid process is the atypical clinical condition of the presented case, in which the patient did not report any significant facial deformity or limitation in mouth opening.

This condition is quite different with respect to the majority of coronoid process hyperplasia cases reported in the literature. These are normally characterized by a marked opening mouth limitation and a clear lateral deviation of the mandible during its opening movement.

A coronoidectomy was performed to remove the pathological process with the histological diagnosis of osseocartilaginous exostosis and no recurrence has occurred. .

***References***


- Ajila V, Hegde S, Gopakumar R, Shubas Babu G. Imaging and Histopathological Features of Jacob's Disease: A Case Study. Head and Neck Pathol 2012,6:51–53.
- Akan H, Mehreliyeva N. The value of three-dimensional computed tomography in diagnosis and management of Jacob's disease. Dentomaxillofac Radiol 2006,35:55-9.
- Beziat JL, Bera JC, Lavandier B, Gleizal A. Ultrasonic osteotomy as a new technique in craniomaxillofacial surgery. Int J Oral Maxillofac Surg 2007,36:493–500.
- Chen MC, Chen CM, Ho CM, Huang IY. Gap coronoidotomy for management of coronoid process hyperplasia of the mandible. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2011,112:e1-e4.
- Chiarini L, Albanese M, Anesi A, Galzignato PF, Mortellaro C, Nocini P, et al. Surgical treatment of unilateral condylar hyperplasia with piezosurgery. J Craniofac Surg 2014,25:808–10.
- Dandriyal G, Giri KY, Pant S, Alam S, Joshi A. Giant Osteochondroma of the Coronoid Process. J. Maxillofac Oral Surg 2015,14(1):S412–S416.
- Durham J, AlBaghdad M, Baad-Hansen L, Breckons M, Goulet JP, Lobbezoo F, List T, Michelotti A, Nixdorf DR, Peck CC, Raphael K, Schiffman E, Steele JG, story W, Orbach R. Self-management programmes in temporomandibular disorders: results from an international Delphi process JOR 2016 43; 929–936.
- D'Amato S, Sgaramella N, Vanore L, Piombino P, Orabona GD, Santagata M. Piezoelectric bone surgery in the treatment of an osteoma associated with an impacted inferior third molar: a case report. Clinical Cases in Mineral and Bone Metabolism2014,11(1), 73–76.
- Fabie L, Boutault F, Gas C, Paoli JR. Neonatal bilateral idiopathic hyperplasia of the coronoid processes: case report. J Oral Maxillofac Surg 2002,60:459–62.
- Gerbino G, Bianchi D, Bernardi M, Berrone S. Hyperplasia of the mandibular coronoid process: long-term follow-up after Coronoidotomy. J of Cranio-Maxfac Surg 1997,25:169-173.
- Gibbons AJ. Case report: computed tomography in the investigation of bilateral mandibular coronoid hyperplasia. Br J Radiol 1995,68:531–3.
- Gonzalez Lagunas J, Mareque J. Piezosurgery its role in TMJ surgery. Italian J Oral Maxillofac Surg 2008,19(17):119–21.

- Isberg A, Isacsson G, Nah KS. Mandibular coronoid process locking: a prospective study of frequency and association with internal derangement of the temporomandibular joint. Oral Surg Oral Med Oral Pathol 1987,63:275-279.
- Izumi M, Isobe M, Toyama M, Ariji Y, Gotoh M, Naitoh M, et al. Computed tomographic features of bilateral coronoid process hyperplasia with special emphasis on patients without interference between the process and the zygomatic bone. Oral Surg Oral Med Oral Pathol 2005,99:93-100.
- Landes CA, Stübinger S, Ballon A, Sader R: Piezo-osteotomy in orthognathic surgery versus conventional saw and chisel osteotomy. Oral Maxillofac Surg 2008a,12:139-147.
- Landes CA, Stübinger S, Rieger J, Williger B, Ha TKL, Sader R: Critical evaluation of piezo-electric osteotomy in orthognathic surgery: operative technique, blood loss, time requirement, nerve and vessel integrity. J Oral Maxillofac Surg 2008b,66:657-674.
- Losa-Muñoz PM, Burgueño-García M, González-Martín-Moro J, Sánchez-Burgos R. Osteochondroma of Coronoid Process: A Rare Etiology of Jacob Disease.Craniomaxillofacial Trauma and Reconstruction 2014,(7)4:306-309.
- Mazzetto MO, Hotta TH. Hypertrophy of the Mandibular Coronoid Process and Structural Alterations of the Condyles Associated with Limited Buccal Opening: Case Report. Braz Dent J 2007,18(2):171-174.
- Mcloughlin PM, Hopper C, Bowley NB. Hyperplasia of the mandibular coronoid process: an analysis of 31 cases and a review of the literature. J Oral Maxillofac Surg 1995,53:250–5.
- Michelotti A, De Wijer A, Steenks M, Farella M. Home-exercise regimes for the management of non-specific temporomandibular disorders. JOR 2005 32; 779–785.
- Mulder CH, Kalaykova SI, Gortzak RA Th. Coronoid process hyperplasia: a systematic review of the literature from 1995. Int. J Oral Maxillofac. Surg 2012,41:1483-1489.
- Murakami K, Yokoe Y, Yasuda S, Tsuboi Y, Iizuka T. Prolonged mandibular hypomobility patient with a “square mandible” configuration with coronoid process and angle hyperplasia. Cranio 2000,18:113-119.
- Peck CC, Goulet JP, Lobbezoo F, Schiffman EL, Alstergren P, Anderson GC, DeLeeuw R, Jensen R, Michelotti A, Orbach R, Petersson A, List T. Expanding the taxonomy of the diagnostic criteria fortemporomandibular disorders. JOR 2014, 41; 2-23.
- Peacock ZS, Resnick CM, Faquin WC, Kaban LB. Accessory Mandibular Condyle at the Coronoid Process. J Craniofac Surg 2011;22(6):2168-2171.
- Ramakant Dandriyal • Kolli Yada Giri • Swati Pant • Sarwar Alam • Ankur Joshi Giant Osteochondroma of the Coronoid Process.J. Maxillofac. Oral Surg 2015,14(1):412–416.

- Robiony M, Polini F, Costa F, Sembronio S, Zerman N, Politi M: Endoscopically assisted intraoral vertical ramus osteotomy and piezo-electric surgery in mandibular prognathism. J Oral Maxillofac Surg 2007,65:2119e2124.
- Sarnat BG, Engel MB. A serial study of mandibular growth after removal of the condyle in the Macaca rhesus monkey. Plast Recostruc Surg 1951;7:364-380.
- Sawada K, Schulze D, Matsumoto K, Hirai S, Hashimoto K, Honda K. Osteochondroma of the coronoid process of the mandible. Journal of Oral Science 2015,57(4):389-392.
- Siddika A, Ferdousi AM. Case Report of a True Accessory Mandibular Condyle - an Exceptionally Rare Abnormality Delta Med Col J 2016,4(1):45–50.
- SreeramanemiSK, Chakravarthi PS, Prasad LK, Satish PR, Beeram RK. Jacob's disease: report of a rare case and literature review. Int J Oral Maxillofac Surg 2011;40(7):753-757.
- Tavassol F, Spalthoff S, Essig H, Bredt M, Gellrich NC, Kokemuller H. Elongated coronoid process: CT-based quantitative analysis of the coronoid process and review of literature. Int J Oral Maxillofac Surg 2012,41:331–338.
- Tucker MR, Guilford WB, Howard CW. Coronoid process hyperplasia causing restricted opening and facial asymmetry. Oral Surg Oral Med Oral Pathol 1984,58:130–2.
- Vercellotti T, Nevins ML, Kim DM, Nevins M, Wada K, Schenk RK, et al. Osseous response following resective therapy with piezosurgery. Int J Periodontics Restorative Dent 2005,25(4):543–9.
- Vercellotti T. Technological characteristics and clinical indications of piezoelectric bone surgery. Minerva Stomatol 2004,53:207–14.

Figure legend

FIG.1 patients light deviation in opening pattern

FIG.2 first visit panoramic x-ray

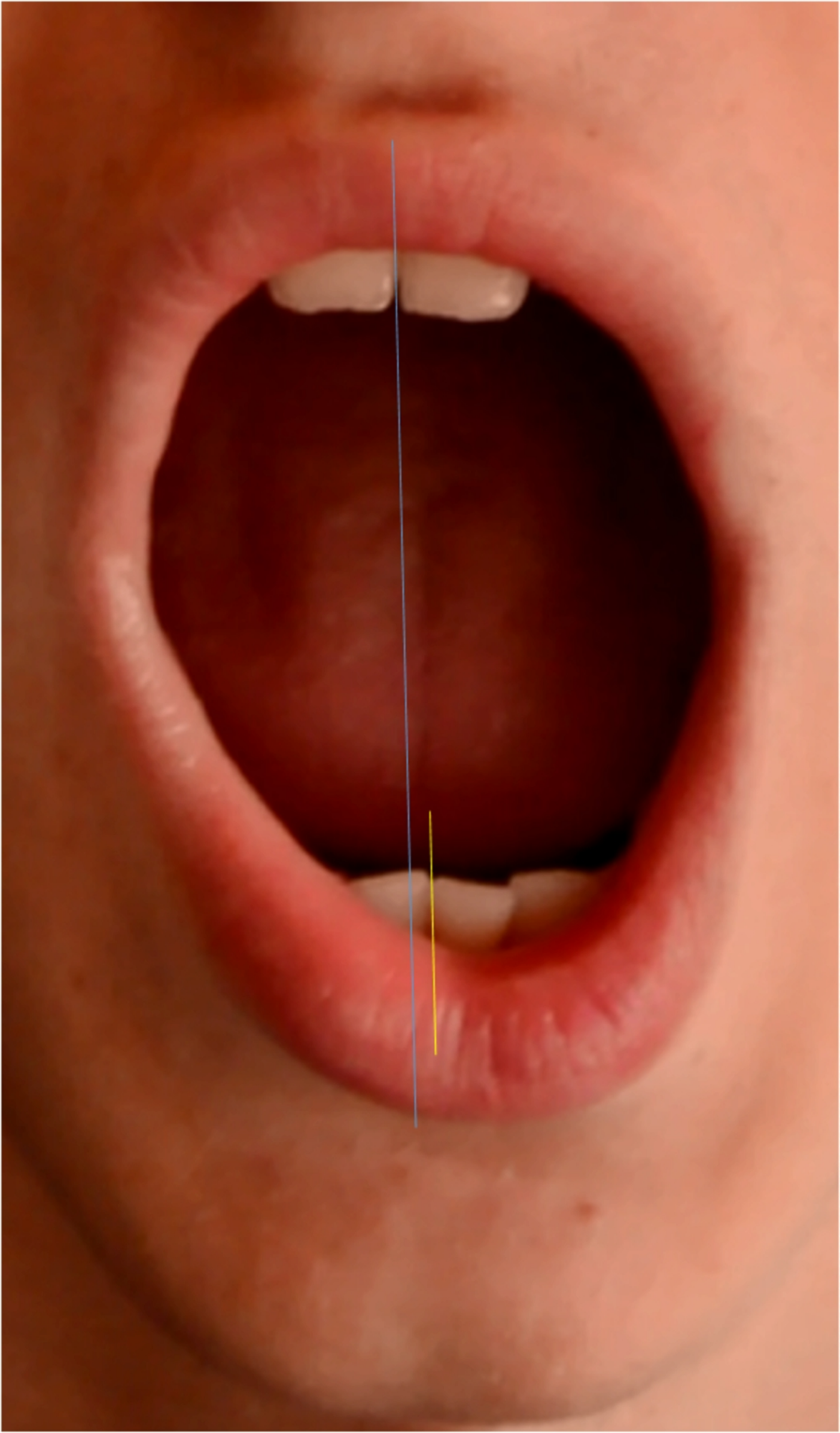
FIG.3 3D VR Image frontal open

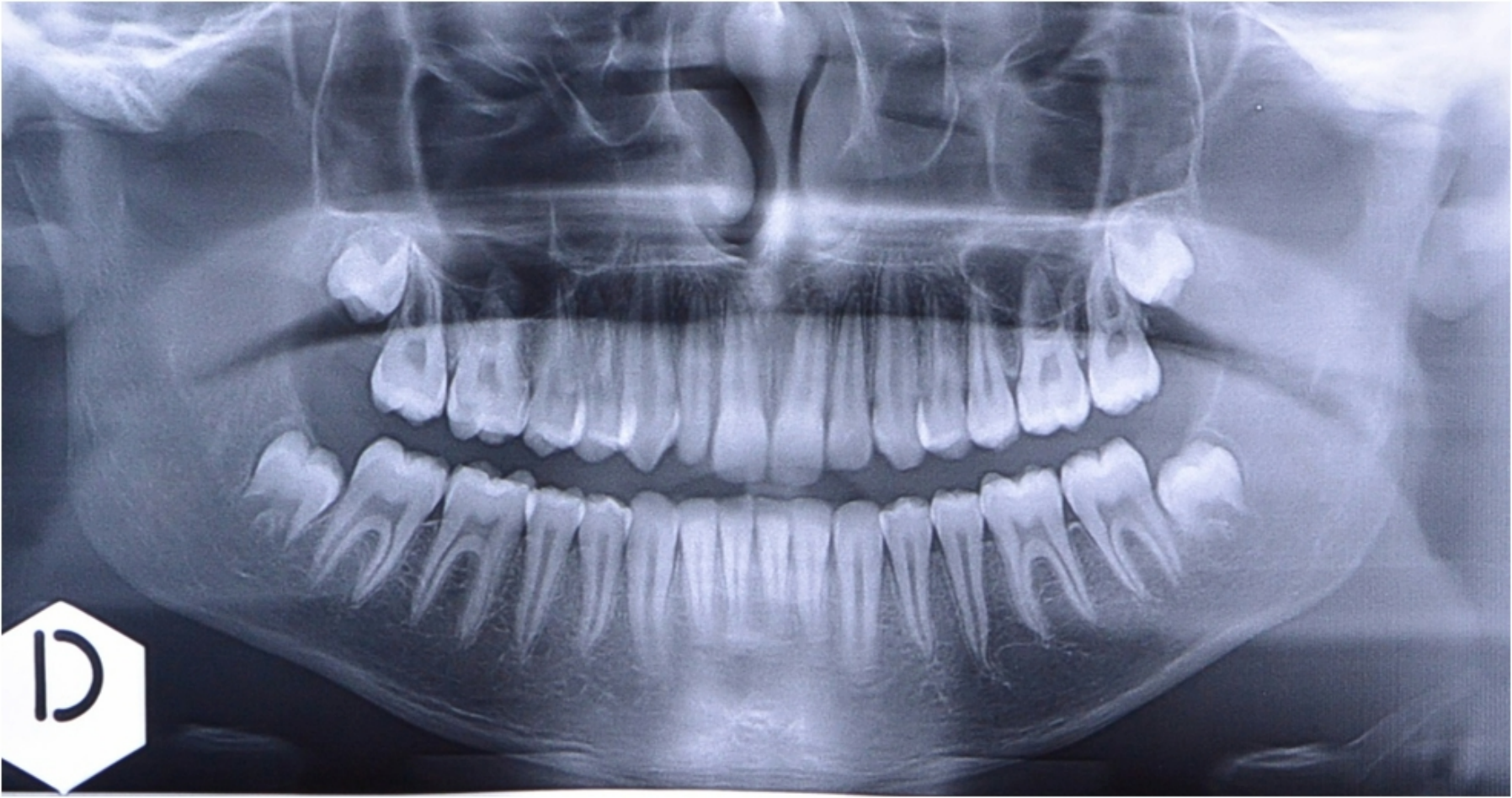
FIG.4 3D VR Image submental open

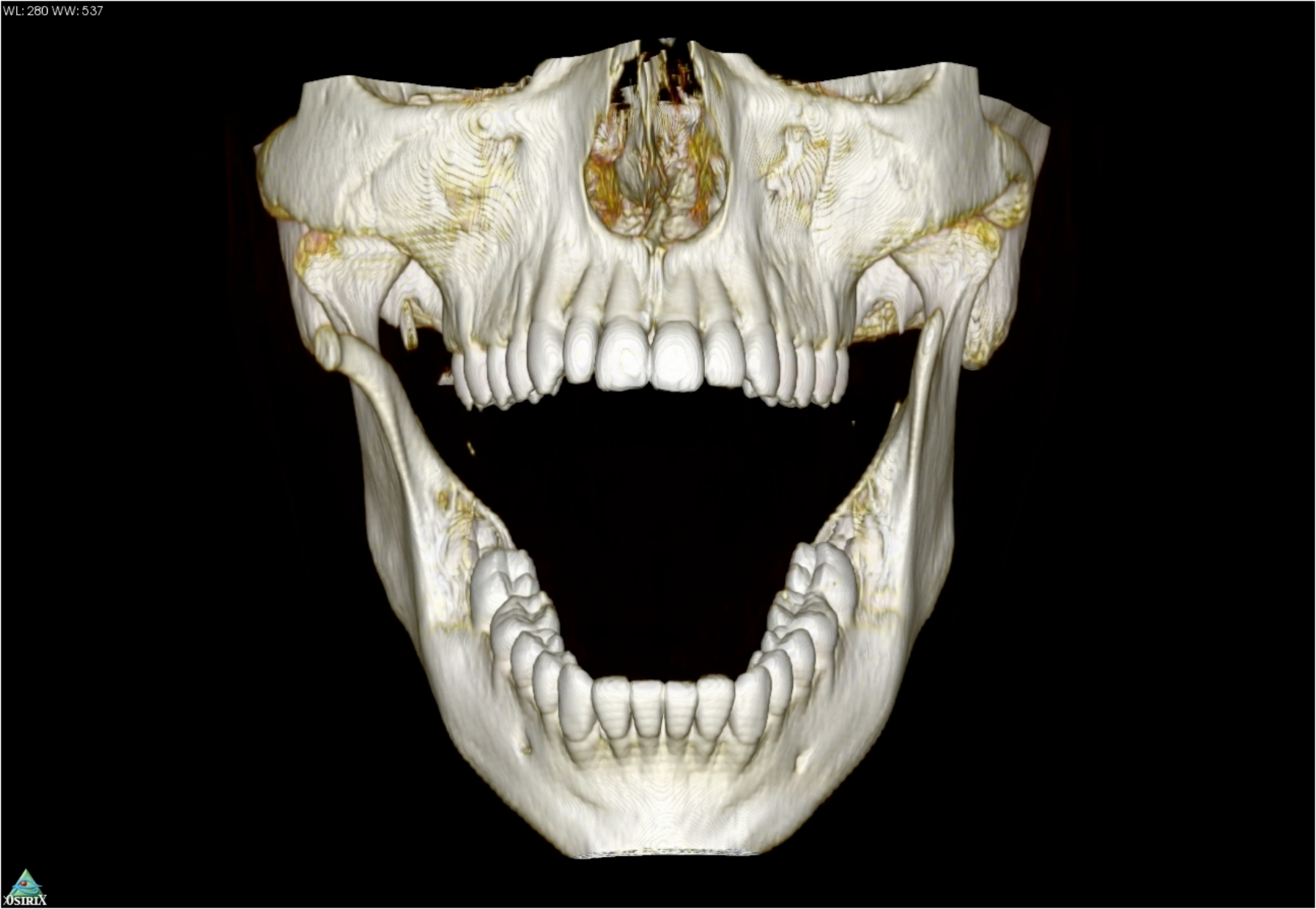
FIG.5 condylar accessory head at coronoid process removing

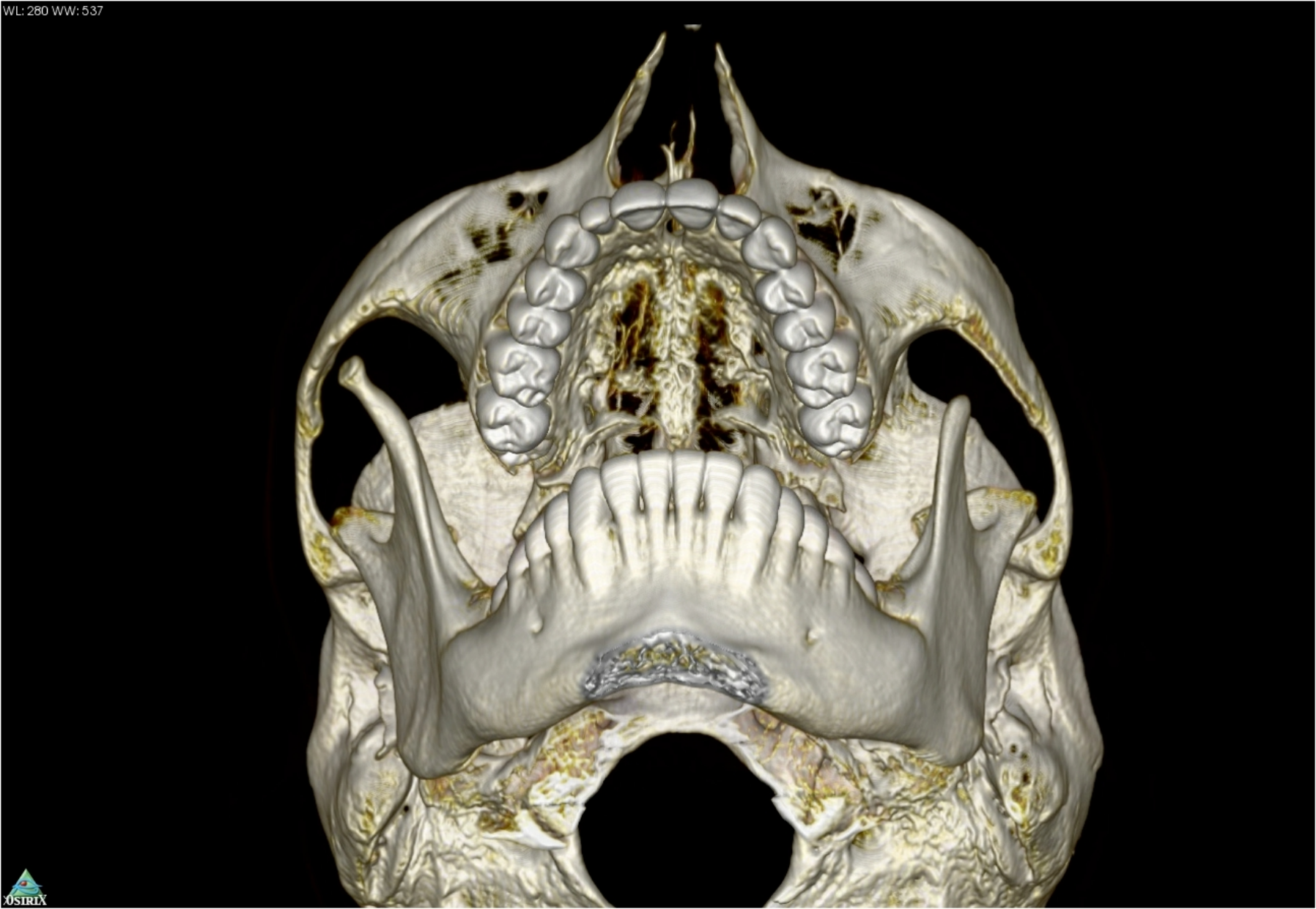
FIG.6 bony specimen

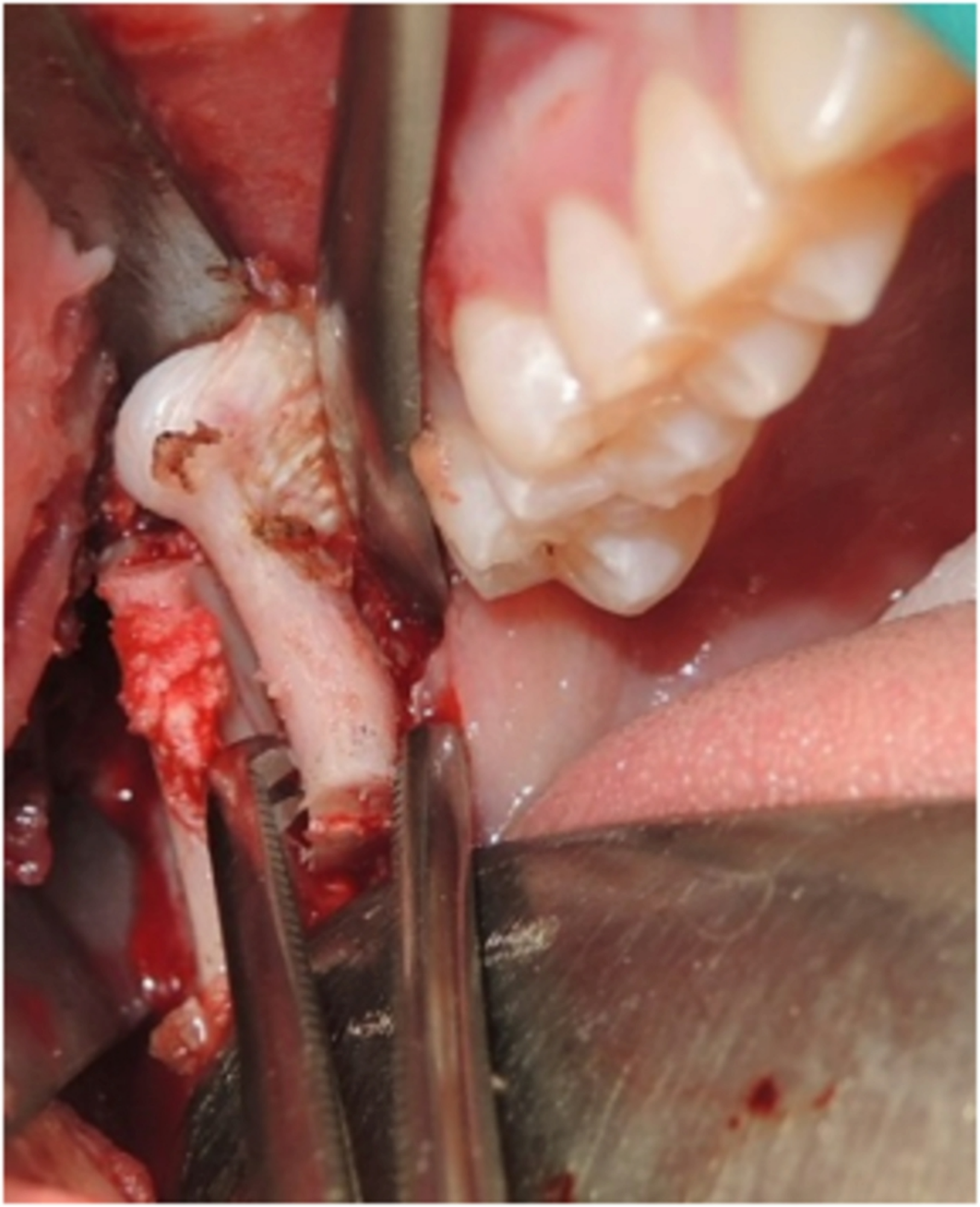
FIG.7a,b Histological appearance of the lesion (low and high power magnification): the perichondrium covers the cartilage cap that merges into the periosteum of the underlying bone (a: haematoxylin and eosin stain, original magnification x 40; b: haematoxylin and eosin stain, original magnification x80)

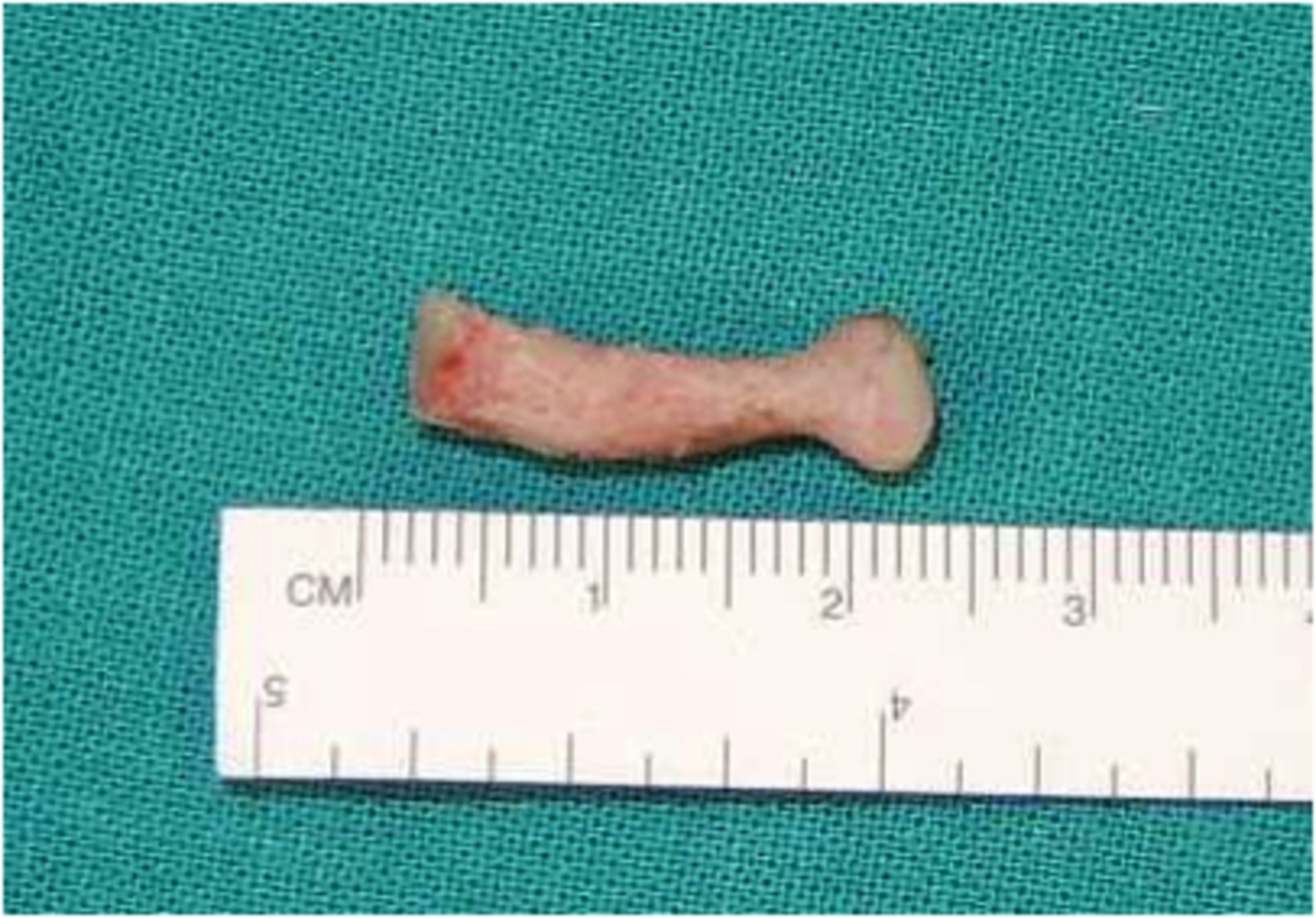





WL: 280 WW: 537
OSIRIX


WL: 280 WW: 537




CM
1
2
3
5
4