Abstract. In the current case, a 31-year-old patient who presented with a painful unilateral malocclusion and an unclear mass in the region of the right temporo-mandibular joint (TMJ) is reported. The malocclusion had been noticed by the patient a few months earlier. Chewing on the right side had become severely impaired and painful. The patient had no history of trauma to the TMJ. Plain radiographs and computed-tomographic scans depicted an enlarged and deformed mandibular condyle. A condylectomy was performed. The histological investigation of the specimen revealed an osteochondroma. The tumour did not express insulin-like growth factor-1 receptor. Five years following the surgical intervention, there has been no local recurrence and dental occlusion was re-established, without further treatment.

Osteochondroma of the mandibular condyle is an exophytic, cartilage-covered lesion arising from the cortex of the bone (1, 2). This localisation is rare for osteochondromas. Osteochondromas constitute approximately 20-50% of all benign tumours and 10-15% of all bone tumours (3). The synonymously used denomination ‘osteocartilagenous exostosis’ refers to the entity as an entirely benign, hamartous lesion (2). Indeed, current concepts of bone tumours regard osteochondroma as a developmental lesion rather than a true neoplasm (3). Osteochondroma only rarely arises in the maxillofacial region (4) and has a preference for the mandibular coronoid process (5, 6). Osteochondroma of the mandibular condyle is rather rare (7, 8). Fewer than 70 cases have been described in the English literature, to date (9). Differential diagnosis from condylar osteochondroma is condylar hyperplasia (9). Expression of the insulin-like growth factor-1 receptor was identified in condylar hyperplasia (10). The aim of this report is to add further radiological and morphological findings regarding knowledge on this rare entity.

Case Report

Patient. The 31-year-old patient was referred to our outpatient clinic for diagnosis and treatment of facial asymmetry, due to deviation of the mandible to the left side, painfully impaired mobility of the right temporomandibular joint (TMJ), and visible swelling in the right pre-auricular region. On admission, the otherwise healthy patient was unable to occlude the teeth on the right side (Figure 1A) and had a cross-bite on the left. Mandibular movement to the right side was severely restricted. There was no history of trauma.

Radiology. An osseous tumour of the right mandibular condyle was depicted on panoramic radiographs (Figure 2A). Tomograms of this region following maximum mouth opening and closure depicted the impaired mobility of the extensive osseous tumour (Figure 2B). On computed-tomographic scans, a partially exophytic mass was depicted, with irregular internal radiopacities (Figure 3B). Three-dimensional reconstructed CT images of the osseous surfaces revealed a spherical to egg-shaped osseous mass of the right condylar head, extending to the anterior and lateral side (Figure 3A-3C).

Therapy. Under general anaesthesia the right TMJ was exposed via a pre-auricular approach. The deformed condyle was identified and was completely resected with the cutting line at the interface of the clumpy lesion to the base of the
condyle above the incisura semilunaris. Intraoperatively, occlusion of the teeth of the right side was achieved (Figure 1B). The articular capsule was sutured and the soft tissues were replaced in anatomical layers. Healing was uneventful. During the recovery period, a malocclusion persisted for several months. This malocclusion was definitely less than the one observed in the preoperative situation and obviously due to swelling, oedema and protracted adaptation to the neo-articulation (Figure 2C). After about six months the teeth of the right side regained complete contact, identical to the intraoperative situs. Mouth opening is restricted to a distance between the incisors of about 35 mm, with no functional impairment of chewing (Figure 1C). Five years after surgery, there is no sign of local recurrence (Figure 2D).

**Histology.** An osseous specimen, with marginal cartilaginous covering, 3 cm, in diameter maximum, was prepared for histological investigation. In routinely processed slices in basal portions of the lesion, osteons with adjacent trabeculae were seen. Inside the medullary space, haematoipoietic cells were present. On the surface of the lesion, opposite to the basal region, an irregularly arranged hyaline cartilage was fixed to the bone. In some areas, remnants of cartilage were located inside the spongious bone (Figure 3D). The specimen was negative for anti-insulin-like growth factor-1 receptor antibody (polyclonal, rabbit, clone c20, sc1713, dilution 1:50; Santa Cruz Biotechnology, CA, USA).

**Discussion**

Osteochondroma of the mandibular condyle is a rare phenomenon (8). Literature on the specific subject is mainly in the form of reports, based on single case descriptions. Only a few reports deal with more than one case but cases are usually small in number (9).

Osteochondroma is predominantly composed of bone tissue and is usually found in long bones (1). The pathomechanism of this lesion is related to a progressive and pathological enchondral ossification. The facial skeleton is predominantly composed of desmal ossified bones. This is one explanation why osteochondromas are rarely found in the facial skeleton (1, 11). Very rarely, manifestations of osteochondroma in the maxillofacial region are of the skull base, maxilla, mandibular ramus, corpus and the symphyseal region (12). The coronoid process of the mandible is of cartilaginous origin and osteochondromas arising in this part of the bone have been regarded as true neoplasms (11). It was proposed that osteochondromas arise from the tendinous attachment of the lateral pterygoid muscle (8). This theory is based on the observation that the tumours (or lesions) in the majority of cases arise from the anterior and medial surface of the condylar process, the site of pterygoid muscle attachment (13). This hypothesis on the pathogenesis of condylar osteochondroma argues in favour of analogous findings in long bones, where osteochondroma develops at the site of tendinous attachments (8). However, in the presented case the lesion extended to both lateral and anterior sides, a finding that supports the hypothesis of a neoplastic origin of the lesion (14).

**Differential diagnosis of osteochondroma from unilateral condylar hyperplasia is necessary (9). In condylar hyperplasia, a regularly shaped but extended condyle is seen on radiographs. Differences in the length of the condylar neck compared to the unaffected side are also regular findings in unilateral hyperplasia. Furthermore, the bone and cartilage have normal patterns of structure and proliferation. On the other hand, osteochondroma often exhibits spherical projections that originate from the margins of the condylar head (Figure 3A). On CT, the osteochondroma is depicted as a growth from a morphologically normal condylar neck, while in unilateral hyperplasia, the whole process is enlarged (15). In the majority of cases, the tumour is well-demarcated on panoramic radiographs as an increase of the condylar head. However, the distinction between tumour and condyle may be hampered due to the combination of cartilage and bone resulting in alternate radiopacities and radiotranslucencies of the lesion (Figures 3B and 3C).

Osteochondroma of the mandibular condyle should be considered in the differential diagnosis of osseous space-occupying lesions of the TMJ. Panoramic radiographs are a valuable screening modality which detect the lesions, but do not depict its extension (9). Functional tomography reveals the joint movement in the sagittal plane (Figure 2B). CT is highly recommended in cases of suspected osteochondroma (15) and allows a precise description of the lesion's border and internal structure (Figure 3B).

Our findings are in accordance with reports from Wolford et al. (16) and Holmlund et al. (17) arguing in favour of condylectomy and conservative treatment for the disease. These authors also described a restitution of the occlusion in the follow-up and no local tumour recurrence after condylectomy, recontouring of the condylar stump, maintaining the articular disc and capsule (16, 17).

Schoen et al. (13) proposed an intraoral approach to resect a condylar osteochondroma. The authors argue against a preauricular approach due to the impaired visibility of the medial aspect of the condylar head, the predominant site of the lesion’s origin. According to these authors, a further disadvantage of the current technique is the necessity to open the articular capsule (13). Indeed, a medial component of the osteochondroma was registered in 52% of cases, sometimes hardly defined on plain radiographs (9). This clinical finding is a reference to the insistence of adequate CT in osteochondroma treatment planning (15). Schoen et al.’s report describes a successful resection of the osteochondroma, but the absence of the lesion is depicted in one plane only, a panoramic radiograph (13). This does not
Figure 1. A: Malocclusion of the jaws at the time of admission. Teeth that are distal to the central incisors cannot come into contact. B: Resection specimen of about 3 cm in diameter. C: Regained occlusion after condylectomy.

Figure 2. A: Panoramic radiograph of the right mandibular condyle depicting an enlarged and deformed condylar head that does not fit into the temporal fossa. B: Zonarc™ radiograph of the right mandibular condyle at maximum mouth opening. The condyle is partly retained inside the fossa and is closely interfaced with the maxillary tubercle. C: Panoramic radiograph of the same region immediately after condylar head resection. There is some space left between the articulating surface of the residual neck and the temporal fossa. D: Panoramic radiograph 5 years after surgery shows the newly formed articular process.

Figure 3. A: Computed-tomography based three-dimensional reconstruction of the skull showing the right temporomandibular joint (TMJ). The osseous lesion has markedly enlarged the condylar head and is growing out of the temporal fossa laterally and anteriorly. B: Axial slice of a computed tomogram of the right TMJ region. The anterior side of the body is to the top. The osseous mass exhibits an inhomogeneous radiodensity (B), growing out in continuity from the condyle and is in part pedunculated (C). D: Toluidine-blue staining of the resection specimen demonstrates the thick cartilaginous layer.
allow the conclusion of complete lesion removal (15). In the present case, the growth of the tumour was not oriented to the centre of the skull base. The lateral approach allowed the inspection of the site of the lesion prior to its resection and the outlining of the new articular process. Opening of the capsule proved to cause no harm to healing and postoperative function, as also reported by others (16, 17).

A tentative diagnosis of osteochondroma rather than hyperplasia can usually be made on routine radiographs (9). However, in both entities, the condylar region exhibits a disproportional growth. The regulation of osseous growth in condylar hyperplasia has been related to insulin-like growth factor-1 receptor expression (10). We were not able to identify this receptor in condylar osteochondroma. However, it remains speculative to negate a role for this factor and its receptor in this disease, based on a single case investigation.

**References**

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