Mandibular Condyle Osteochondroma. Review of the Literature and Report of a Misdiagnosed Case

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Temporomandibular disorders can usually be diagnosed on the basis of a thorough history and a comprehensive examination of the patient. Additional diagnostic tests, such as imaging of the temporomandibular joint (TMJ) area, are mandatory and must be flawless in case of atypical findings. The aim of this report is to illustrate pitfalls in clinical reasoning and in imaging procedures in the diagnosis of temporomandibular pain and dysfunction. A case report of a patient with osteocartilaginous exostosis of the mandibular condyle, which was erroneously diagnosed and treated as an internal derangement of the TMJ for half a year, is presented.
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Temporomandibular disorders (TMD) is a collective term embracing a number of musculoskeletal problems that involve the masticatory musculature, the temporomandibular joint (TMJ), and associated structures, or both.[1] TMD is characterized by pain and/or restricted range of mandibular motion. Temporomandibular joint sounds often exist. These symptoms are not diagnostic if occurring solely, because many disorders and diseases show the same signs and symptoms as TMD. For example, restricted range of motion of the mandible can also result from inflammation, neoplasm, and/or soft tissue disorders. Therefore, pain and dysfunction in the orofacial region represent a diagnostic challenge. One of the lesions that may mimic TMD is osteochondroma in the region of the jaws.

Osteochondromas occur singly or as a part of an autosomal dominant syndrome known as osteochondromatosis.[2] Radiologically and histologically, multiple osteochondromatosis and solitary osteochondromas cannot be distinguished from each other.[3] This distinction is clinically relevant because multiple osteochondromatosis has a higher risk of sarcomatous transformation (11%) than that of the solitary osteochondroma (1%).[4] Another clinical feature is the location of the lesion: *(1)* the axial skeleton, more often seen in childhood with the potential to regress at adolescence,[3] and *(2)* the facial skeleton. Because of its potential to regress, the true neoplastic type of osteochondroma can be recognized only in adulthood. Although osteochondroma is one of the most common bony lesions, it is extremely rare in the craniofacial area.[5–7] Indeed, previous epidemiologic data suggest that the large majority of solitary osteochondromas occur at the distal metaphysis of the femur and proximal metaphysis of the tibia, whereas only 0.6% of osteochondroma occur in the craniofacial region.[8]

When present, the tumor is most often reported to affect the coronoid process, although other sites are described, such as the posterior maxilla, maxillary sinus, mandibular symphysis, and zygomatic arch. Osteochondroma of the mandibular condyle is extremely rare.[2,9]

The pathogenesis of osteochondroma is still under debate.[2,3,10] It is suggested that herniation of cartilaginous precursor cells as a result of defects in the epiphysial periosteal cuff allows the eventual formation of such lesions.[11] The periosteum may have the potential to form chondroblasts and osteoblasts and give rise to osteochondroma.[9] Evidence for a genetic etiology has also been reported.[3,10] In the most accepted view, the pathogenesis is a metaplastic change of the periosteum and/or the osteochondral layer in the coronoid process or the mandibular condyle, leading to production of cartilage that subsequently ossifies. The tumor is also thought to develop in and from the tendinous attachment of muscles. However, reports of osteochondromas near the lateral pole of the mandibular condyle[2,9] throw doubt on this explanation. Histologically, osteochondroma has been described as a cartilage-capped lesion, with endochondral ossification deeper in the tumor. The cartilage is often hyaline. The mean age of discovery of osteochondromas of the mandibular condyle is reported to be the fourth decade of life of the patient, and there is a female preponderance. The radiographic appearance of osteochondroma of the mandibular condyle can be pathognomonic, with the tumor appearing as an irregularly shaped, mixed density, expansile lesion.[12]

The following report illustrates a case that was erroneously treated as an internal derangement of the TMJ for a half year, until a correct diagnosis was made. The aim of this report is to illustrate pitfalls in clinical reasoning and imaging procedures in the diagnosis of TMJ pain and dysfunction.

Case Report

History

A 40-year-old man presented with clicking of the right TMJ, pain in the joint area, restricted range of motion, and a facial asymmetry (Fig 1). The patient reported the onset of TMJ clicking about 7 to 8 years before; moreover, he reported a facial trauma (vehicle accident) 5 years before. Since this accident he noticed a slowly increasing asymmetry of the face with a chin deviation to the left side, accompanied by a progressive limitation of mandibular movements that had become worse during the last 8 months. The main complaint of the patient was pain in the right TMJ during closing movements. After opening wide, he could hardly close his mouth; he was able to bring his teeth into contact only after specific maneuvering that included manual assistance. This movement was accompanied by audible crepitation in the right TMJ area.

Before presenting to our department, the patient had been referred to 2 other dental clinics. In the first clinic, the patient was diagnosed as having an internal derangement of the right TMJ; this diagnosis was based upon the history, clinical examination, and radiographic evaluation with panoramic and tomographic radiography of the TMJ. He was treated with a mandibular stabilization splint that was worn 24 hours a day over a 5-month period. As the patient did not benefit from this splint, he consulted a second dental clinic. In the second clinic, a specialist diagnosed a Class III malocclusion and suggested orthodontic treatment followed by orthognathic surgery. This diagnosis was made on the basis of new clinical records and on the basis of the radiographs obtained in the first dental clinic. The suggested treatment was not accepted by the patient. Six months after the first diagnosis was made, the patient presented to our department.

Inspection and Clinical Examination

Clinical examination showed a marked facial asymmetry. The mandible deviated to the left side in intercuspal position (Fig 1). Previous photographs of the patient from 10 years earlier did not show the asymmetry. In occlusion, a deviation of the chin to the left side of approximately 9 mm and an asymmetric prognathism was evident. Inspection revealed a unilateral posterior crossbite on the left side. The active range of motion on jaw opening was 35 mm; passive range of motion on opening was 40 mm. At maximal jaw opening the deviation of the mandible to the left side was less pronounced, with the dental midlines almost in line. The movement pattern was irregular and not well coordinated. The range of motion in protrusion was almost 2 mm. The lateral excursions were asymmetric and the ranges of motion in left lateral movement and right lateral movement were 4 and 7 mm, respectively. All mandibular movements provoked pain in the right TMJ. Both condyles were not tender to palpation and the morphology of the lateral poles of both condyles was normal. During jaw opening, translation of the right condyle was practically absent. Manually assisted translation of the right TMJ was hardly possible

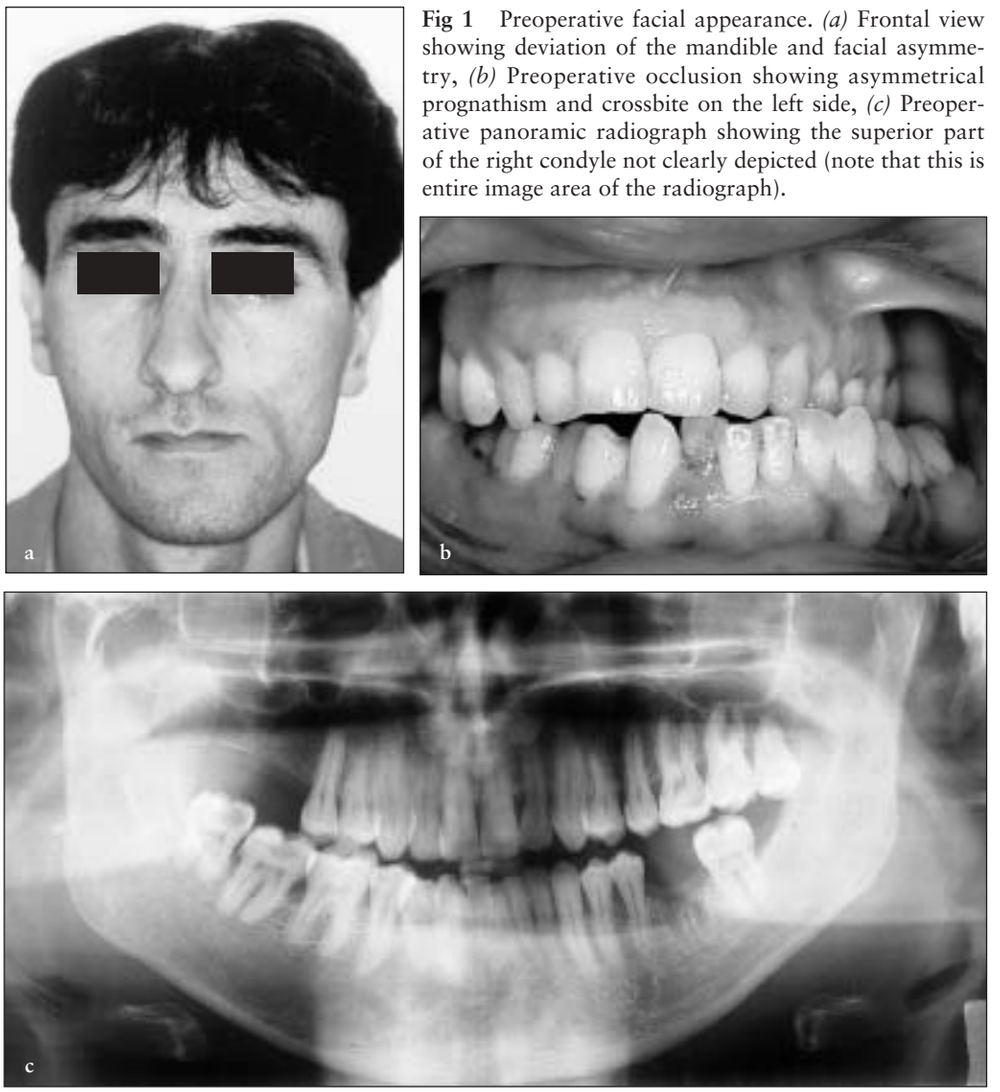

Fig 1 Preoperative facial appearance. *(a)* Frontal view showing deviation of the mandible and facial asymmetry, *(b)* Preoperative occlusion showing asymmetrical prognathism and crossbite on the left side, *(c)* Preoperative panoramic radiograph showing the superior part of the right condyle not clearly depicted (note that this is entire image area of the radiograph).

and provoked moderate pain. The right masseter muscle was tender to palpation.

The intraoral inspection showed that 6 teeth were missing (18, 17, 16, 36, 38, 48). Both maxillary and mandibular incisors showed signs of moderate attrition (limited to enamel). Occlusal contact in intercuspal position was limited to the left side (teeth 26, 27, 37).

In the panoramic radiograph, available from the first consultation, the superior part of the right condyle was not well depicted (Fig 1). On TMJ tomograms, available from the first consultation, the right condyle seemed flattened anteriorly. On close examination, a radiopaque structure anterior and superior to the condyle could be seen (Fig 2). Computed tomography (CT) scans, requested in addition to the previously made radiographs to evaluate its extension, showed an irregularly shaped structure medially and superiorly, extending from the right condyle to the base of the skull (Fig 3). Intracranial extension of the lesion was not evident. In sagittal view of the closed mouth position, the condyle was positioned anteriorly out of the glenoid fossa at the summit of the eminence. In the open-mouth radiograph, this position had hardly changed. The neuroradiologist reported the structure to be consistent with an osteochondroma or condylar hyperplasia.

Management

The treatment called for was the surgical approach. Under general anesthesia, a condylectomy was performed. After a preauricular incision was made, the right condyle and the extending structure were

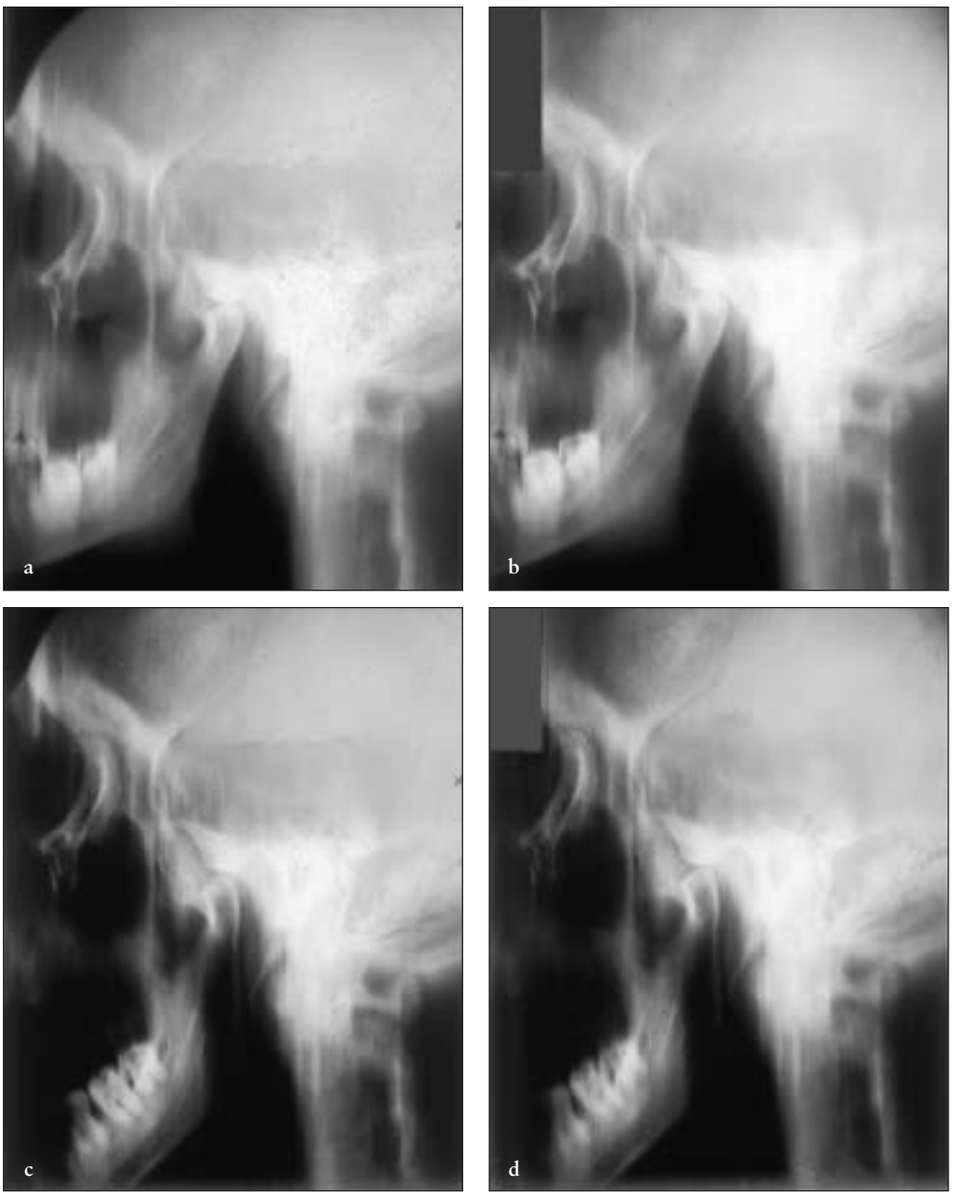

Fig 2 Tomograms of the right closed (a,b) and opened (c,d) mouth position. Note the radiopaque area surrounding the condyle and the position of the condyle in closed mouth position.

excised. The bony end of the condylar neck was shaved. The surgical specimen was mushroom-shaped, consisting of 3 parts, the greater measuring about 3 cm on its main axis. The final diagnosis, based on the histologic report, was an osteocartilaginous exostosis.

One month after the surgical procedure, the patient could open his mouth 47 mm; there was a slight deviation of the mandible to the right side due to the condylectomy. Lateral movements of the mandible were 8 mm to the left side and 11 mm to the right side. Intraoral fixation using elastics guided the maxillomandibular relation in the first 6 months. On follow-up 1 year later, the patient reported no significant problems concerning function or esthetics of the reconstructed area. The panoramic radiograph showed remodeling of the resection stump, serving as a condyle (Fig 4).

Discussion

Clinical Diagnosis

According to the classification of the American Academy of Orofacial Pain (AAOP), the term TMD

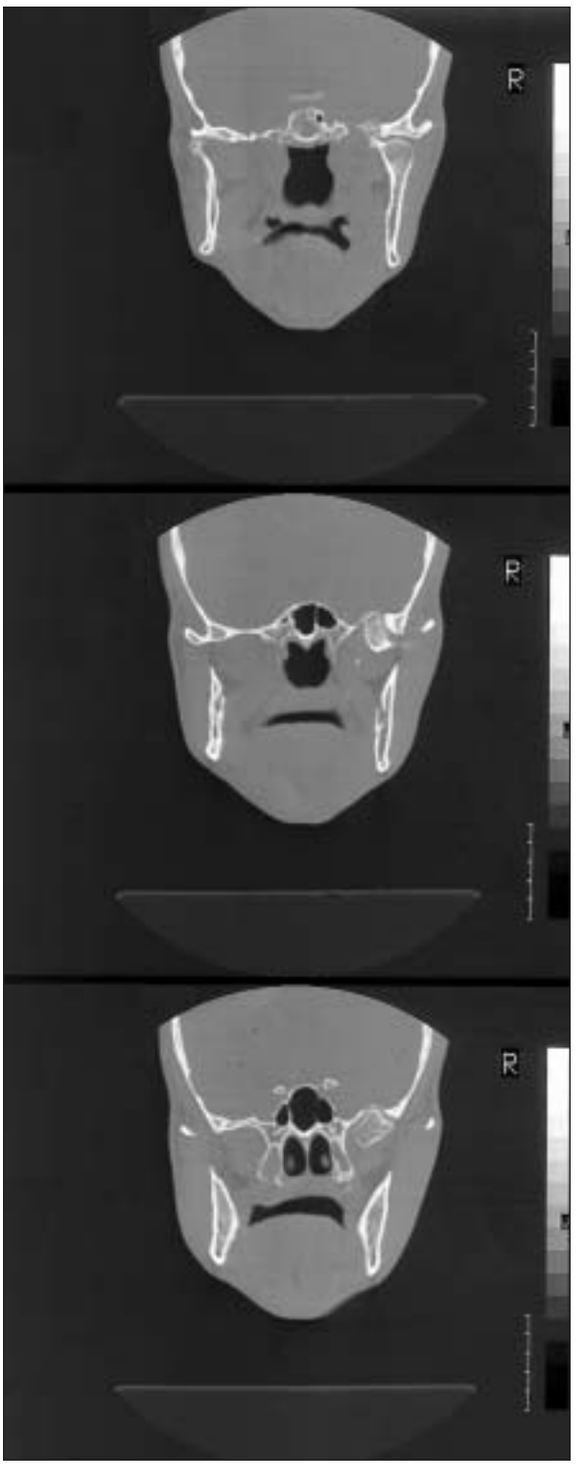

Fig 3 Coronal projection of CT scan demonstrating the irregular shaped structure extending from the right condyle medially and superiorly.

refers to TMJ articular disorders and masticatory muscle disorders, category 11.7 and 11.8, respectively.[1] TMJ articular disorders include congenital or developmental disorders, disc derangement disorders, dislocation, inflammatory conditions, arthritides, ankylosis, and fracture. The research diagnostic criteria (RDC/TMD) Axis I refer to clinical TMD conditions,[13] and 3 groups are distinguished (after ruling out "less common" muscle and joint conditions): *(1)* muscle disorders, *(2)* disc displacements, and *(3)* arthralgia, arthritis, and arthrosis. The authors use the term TMD in this case report to reflect the clinical TMD conditions mentioned in the RDC/TMD, realizing the broader meaning mentioned in the AAOP guidelines. This case report illustrates that adopting the narrower view of TMD at the beginning of the diagnostic process can lead to false conclusions. Only an accurate interpretation of all signs and symptoms by means of a thorough history and clinical examination in combination with an open mind to either confirm or rule out the less common conditions, allows a correct diagnosis and treatment.

Benign tumors such as osteochondroma, condylar hyperplasia, or osteocartilaginous exostosis are described regularly in adult patients who present symptoms similar to TMD.[9,14] In the general population, tumors and growth or developmental disorders are far less prevalent than clinical TMD conditions. Therefore, the odds to evaluate a patient for TMD are much higher than for a tumor mimicking TMD. However, throughout the diagnostic process the clinician has to keep an open mind for other diseases, especially when signs and symptoms occur that are noncharacteristic for TMD.

The following aspects were atypical for TMD and should have prompted the clinician to suspect another pathology:

- Progressive asymmetry during the past years, indicative for a growth and development anomaly
- Difficulty in the closing pattern after opening wide, accompanied by loud crepitation; the patient needed manual assistance to close his mouth and to bring the teeth into contact during the whole closing movement (this phenomenon differs from a locked open mandible or posterior disc displacement)
- Progressive limitation of mouth opening, without the specific history and findings, consistent with an internal derangement

Indeed, the progressive asymmetry of the mandible is not consistent with an internal derangement of the TMJ. When the occlusion is changing after growth, condylar pathology must

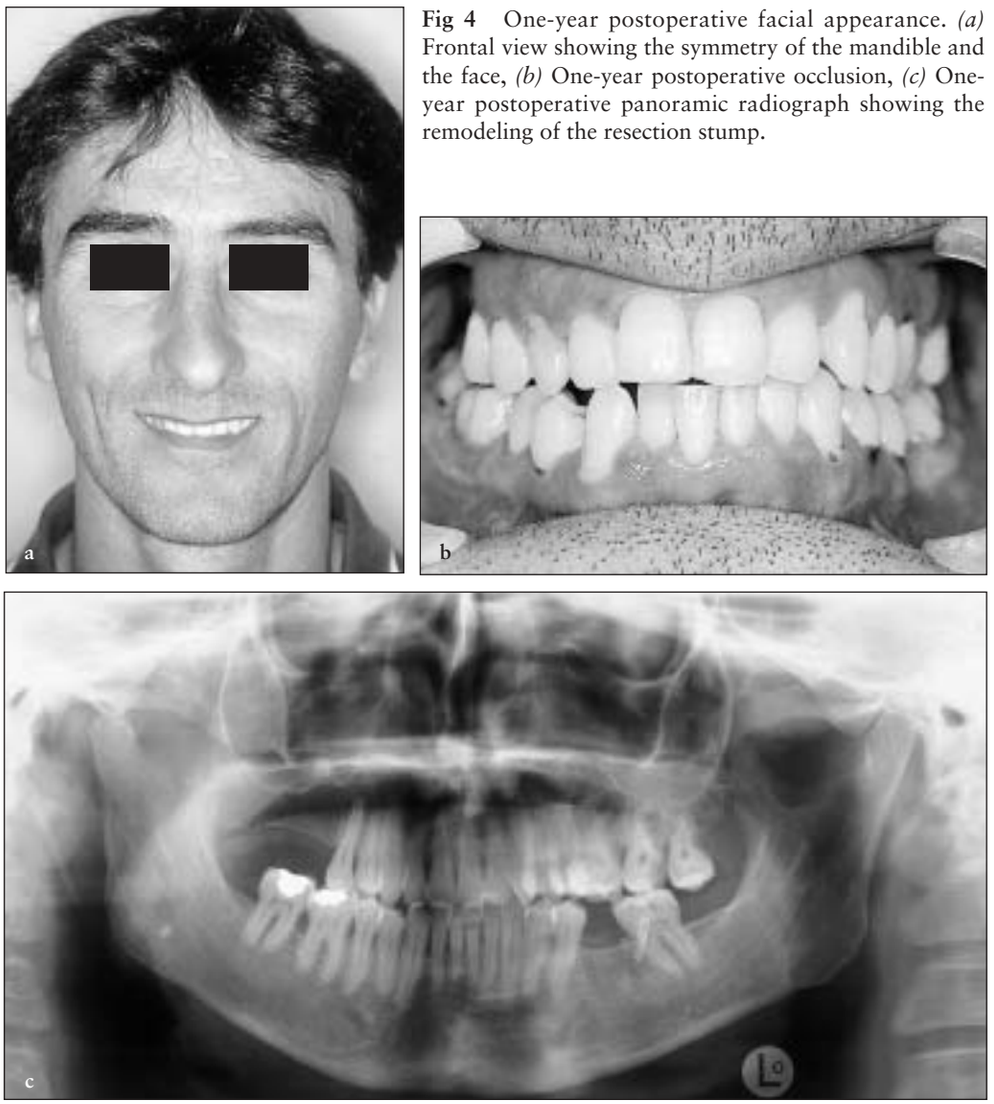

Fig 4 One-year postoperative facial appearance. *(a)* Frontal view showing the symmetry of the mandible and the face, *(b)* One-year postoperative occlusion, *(c)* One-year postoperative panoramic radiograph showing the remodeling of the resection stump.

be considered. With the exception of idiopathic condylysis, in the absence of systemic diseases or severe osteoarthrosis, condylar resorption is generally not expected.

A Class III malocclusion with side deviation of the mandible rarely develops in an adult. A late-onset prognathism, especially when presenting with an asymmetry, is much more suggestive of condylar changes mediated by hormones, tumor, or inflammation. If within 6 to 12 weeks of treatment the signs and symptoms do not change, the diagnosis needs to be re-evaluated, and the need for an additional examination or test established.

In addition, the patient's difficulty in closing after opening wide and in closing into intercuspal position is not consistent with either TMJ internal derangement or with myofascial pain with limited opening. For these reasons the diagnoses from the 2 previous consultations were not accepted.

In the case of noncharacteristic findings, the use of a flowchart can help the clinician to arrive at the correct diagnosis (Fig 5). History, clinical examination, and the panoramic radiograph are the methods of first choice for the diagnosis of TMD or other pathologies of the stomatognathic system. Additional tests are indicated if the symptoms are inadequately explained by the signs or findings, or if the panoramic radiograph shows conditions that need further evaluation. Thus, to make a correct diagnosis, the findings need to explain the symptoms.

The report of a vehicle accident is noteworthy, because trauma has been considered as a potential

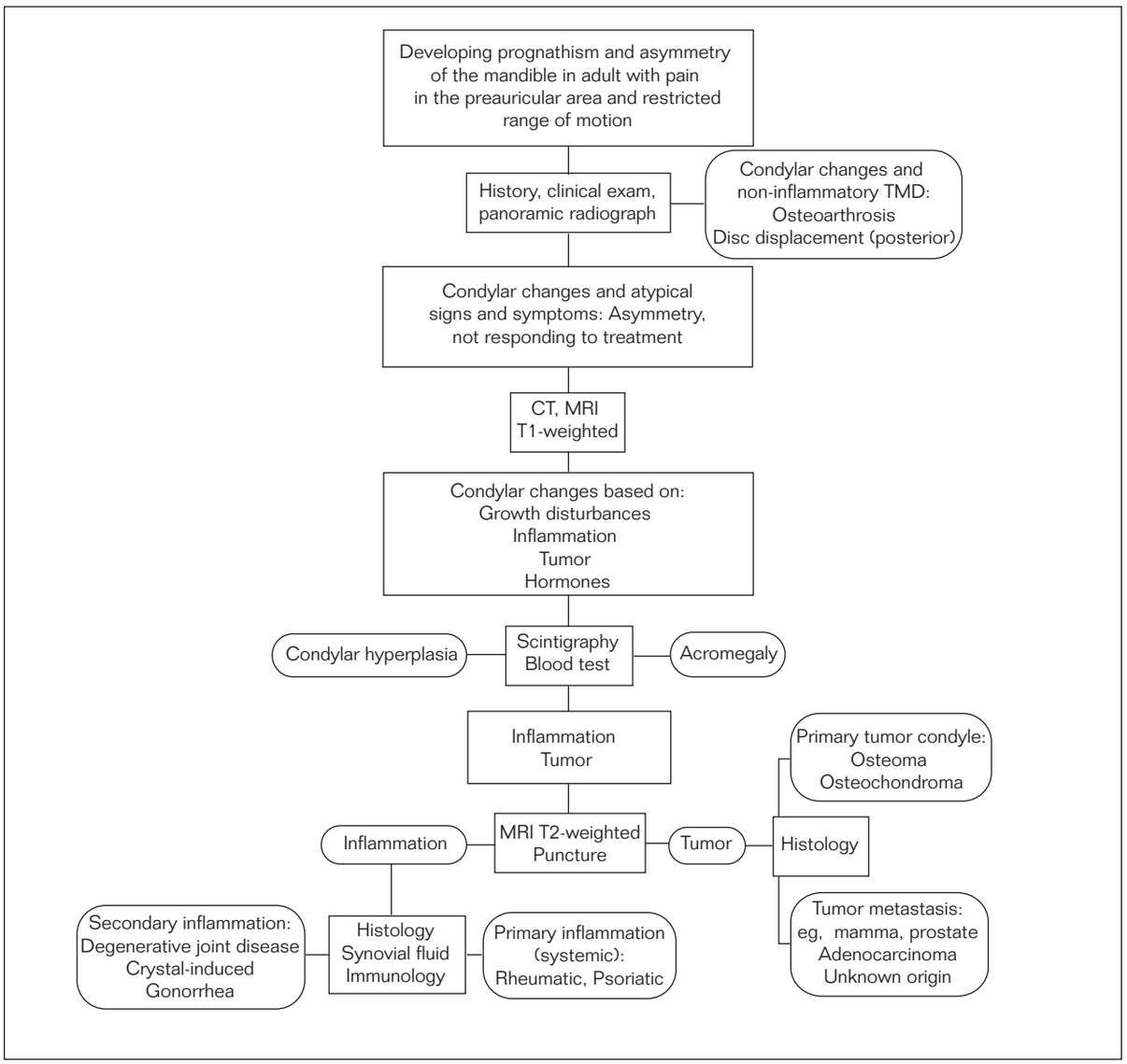

Fig 5 Flowchart of diagnostic process. The information obtained from preceding tests is used in subsequent decisions. T1-weighted images demonstrate the anatomy; T2-weighted images indicate effusion as a sign of pathology.

etiologic factor of osteochondroma. It is possible that damage of the condylar surface stimulates a reactive bone overgrowth.[11] Osteochondromas can develop spontaneously or by metaplasia of the periosteum to form cartilage that subsequently undergoes endochondral ossification.[2,6]

Imaging

In general, the working diagnosis resulting from the history must be confirmed or rejected by the findings of the clinical examination. Radiographs are necessary if one suspects a bony or soft tissue lesion. Imaging is mandatory in progressive asymmetries in order to evaluate any kind of condylar change. In this case, the preoperative panoramic radiograph was properly collimated but did not show the entire right condyle, probably because of an improper positioning of the patient, resulting in inadequate imaging of the condyle. This led to the loss of essential information, in this case the extending osteocartilaginous exostosis. Any panoramic radiograph not depicting the entire condyle and its surroundings is not acceptable for a TMD diagnosis and should be repeated. The seemingly normal appearance of the majority of the

contour of the right condyle may have disguised the pathology because the clinician is tempted to fill in the information lacking.

The tomographic radiographs prescribed in the other dental clinic did not allow the pathologic change to be discovered, because of the poor technique used, the limited number of slices, and the superposition of anatomical structures. To evaluate the structural change and its extension in relation to the surrounding tissues, CT scans in the relevant planes are indicated. The results of the combination of history, clinical examination, and CT scans, in this case, led to the correct diagnosis of a TMJ tumor. The diagnosis of the type of tumor was made through histology.

Management

A surgical intervention was performed because of the severe limitation of the mandibular movements as well as the potential malignant transformation of the suspected tumor. Although condylar osteochondroma has been reported to be benign, an early diagnosis is important because of the risk of sarcomatous transformation.[4,8] Moreover, more gross deformity and related sequelae may occur.[4] Regardless of the pathogenesis of the lesion, in almost all reported cases the treatment has been a radical resection of the tumor, including the condylar process.[5,6] Immediate reconstruction of the condyle should be considered if function is not adequate and the occlusion cannot be corrected by surgery (eg, osteotomy).[12]

Conclusions

In most cases, subgroups of TMD can be diagnosed only by means of the history and of a comprehensive clinical examination of the patient. Many disorders or pathologies in the head and neck can present signs and symptoms similar to those of TMD. The panoramic radiograph must be flawless in order to determine the necessity of additional imaging or tests. In the case of atypical signs and symptoms or of nonresponse to treatment within the expected healing time, re-evaluation is necessary and additional diagnostic tests, such as imaging of the TMJ area, may be mandatory. Because of the broad spectrum of disorders and pathologies that may cause orofacial pain and limitation of mandibular movements, the clinician must not think only of TMD when examining an orofacial pain patient, but must always keep in mind all those pathologies and disorders that may mimic TMD.

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