Clin Neuroradiol (2019) 29:787–789
https://doi.org/10.1007/s00062-019-00778-0

CORRESPONDENCE

Coronoid Process Hyperplasia

A Rare Disorder Masquerading as Temporomandibular Joint Disease

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Received: 21 June 2018 / Accepted: 12 March 2019 / Published online: 15 April 2019

Introduction

Bilateral coronoid process hyperplasia is a rare condition causing painless, progressive reduction in mouth opening, referred to as trismus. The elongated coronoid process impinges on the medial or temporal surface of the zygomatic arch limiting mandibular motion that can severely impact oral hygiene and quality of life. Given its insidious onset, a missed or delayed diagnosis can occur, even when plain radiography is utilized [1]. The author reports a case of coronoid hyperplasia in a young child that was initially misdiagnosed with a temporomandibular joint (TMJ) disorder. The discussion emphasizes the role of computed tomography (CT) in initial diagnosis and surgical planning of this rare entity. Although rare, this diagnosis should be familiar to radiologists specializing in maxillofacial imaging.

Case Report

A 4-year-old male presented to his dentist for routine dental cleaning. The patient demonstrated decreased mouth opening with a hard stop at 15 mm incisal opening distance. The patient's mother reported limited mouth opening since birth without a history of trauma, perinatal/childhood disease, or family history of trismus. The patient was given a presumptive diagnosis of TMJ disorder and referred to oral maxillofacial surgery.

Physical examination by the oral surgeon also demonstrated a maximum incisal opening of 15 mm although clinical suspicion for TMJ disorder was deemed less likely secondary to lack of pain on palpation and hard stop on mouth opening. Initial axial CT imaging revealed abnormal elongation of the coronoid process extending above the level of the zygomatic arches without evidence of mandibular hypoplasia or other facial malformations. Based on the radiologic findings, coronoid process hyperplasia was diagnosed with operative plan for bilateral coronoidectomy and repeat perioperative CT with the patient under anesthesia with 2-D and 3-D multiplanar reformations to aid in surgical planning (Fig. 1a–c). Postoperative incisal opening distance improved to 25 mm.

The oral range of motion 5 years postoperatively returned to its preoperative level with a maximum incisal opening of 15 mm. Repeat CT with 3-D reconstructions to confirm recurrence and prepare for repeat intervention revealed recurrent and progressive coronoid hyperplasia (Fig. 1d). The coronoid process measured 2.7 cm on the left, and 2 cm on the right in craniocaudal dimension. Initial surgical failure was attributed to continued facial growth secondary to the patient's young age at presentation and inability to actively engage in postoperative physical therapy. Ultimately, repeat surgical intervention was delayed until the patient could fully engage in postoperative rehabilitation.

Discussion

Coronoid hyperplasia as a cause of limited mouth opening remains relatively unknown and often misdiagnosed. First referenced as Jacob's disease, the hyperplastic coronoid process is described as forming a pseudocartilaginous joint with the zygomatic process [2, 3]. Histologically, the elongated coronoid process is formed of normal bone closely associated with the zygomatic bone [4]. Proposed etiologies include mandibular hypomobility or hypoplasia, temporalis muscle disorder, TMJ dysfunction and genetic inheritance. Occurring in both unilateral and bilateral forms, the unilateral form is often associated with trauma or other

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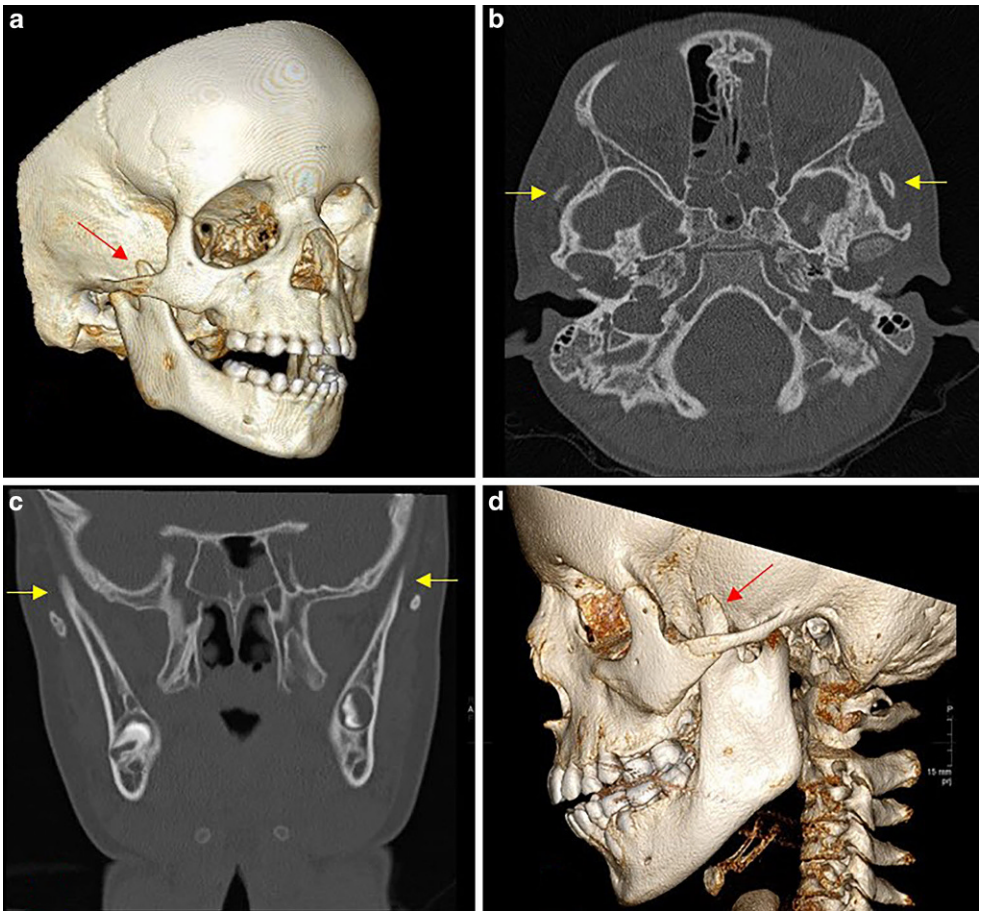

Fig. 1 Volume-rendered 3-D image from the patient's preoperative CT shows the tip of the right coronoid process (*red arrow*) extending above the level of the mandibular condyles (**a**). Axial (**b**) and coronal (**c**) Computed Tomography images show the bilateral coronoid processes extending above the level of the zygomatic arches (*yellow arrows*).The patient's mandibular range of motion returned to preoperative levels 5 years postoperatively necessitating repeat CT. A volume-rendered 3D image demonstrates progressive elongation of coronoid processes projecting superior to the zygomatic arch (*red arrow*) (**d**)

pathological conditions [5, 6]. Given an often progressive and insidious onset, misdiagnosis of TMJ dysfunction often occurs initially. A considerable period between symptom onset and definitive diagnosis is described [7, 8]. In a systematic meta-review, the mean age of clinical presentation was 23 years with 7 years of mean symptom duration prior to appropriate treatment [4]. The case presented is unusual in the young age of presentation.

A coronoid/condyle ratio of <1.0 has been proposed as the definition of normal anatomy [9]. Although initial detection may occur with panoramic radiograph, CT best delineates the morphology and extent of involvement. Protocols include axial and sagittal CT reformats in both the open and closed positions with multiplanar reformats and 3D rendering acting as a fundamental surgical planning tools for oral surgeons. Postoperatively, magnetic resonance imaging (MRI) can be considered to assess for soft tissue complications, specifically fibrotic reaction of the temporalis muscle at the coronoidectomy site. The TMJ disc is usually morphologically normal although internal derangement may be present [10].

The advantages of CT over MRI include rapid image acquisition and improved spatial resolution with 3D image reconstruction. Imaging goals are to aid in preoperative planning while conforming to as low as reasonably achievable (ALARA) radiation exposure principles [11]. Whereas the lowest possible radiation dose is desired, effort should be made to obtain high quality images that prevent repeat examinations. This patient required a second CT under general anesthesia immediately prior to surgery as initial low dose imaging was subject to motion artifacts and lacked the desired multiplanar reformation (MPR) and 3D reconstructions. Although the lack of MRI can be considered a limitation of this case study, soft tissue injury and associated TMJ dysfunction were not suspected thus eliminating the necessity for MRI.

After diagnosis, successful treatment includes coronoidectomy with postoperative physical therapy [12]. Relapse is most likely the result of persistent underlying causes of hypertrophy, postoperative fibrosis, and/or an inadequate physiotherapy program [13]. Although further surgery may be required to treat limited opening, prognosis of coronoid hyperplasia shows satisfactory, long-term results with adequate treatment [14].

As a rare, often painless, non-neoplastic condition, coronoid hyperplasia has significant potential for underreporting by both patients and clinicians. Restricted mouth opening can limit eating, yawning, routine dental care, and could

even be life-threatening if intubation is needed. Fortunately, these morbidities have the potential to be corrected with appropriate diagnosis and treatment. As such, radiologists who specialize in maxillofacial imaging are crucial to developing the appropriate diagnosis and surgical planning of this uncommon but treatable entity.

Conflict of interest E. Schneble declares that she has no competing interests.

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