

Unilateral coronoid hyperplasia associated with early childhood facial trauma: a case presentation

Précis

A case presentation of coronoid hyperplasia presenting in adolescence. An association with early childhood trauma to the chin and subsequent development of the condition is proposed.

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Introduction

Although a rarely encountered clinical entity, hyperplasia of the coronoid process is a condition that dental practitioners should consider when encountering patients with limited mandibular movements. The condition was first described by von Langenbeck in 1853,¹ though currently the actual incidence is unknown.² Coronoid hyperplasia may occur bilaterally or unilaterally, and is defined as an increase in the coronoid process, resulting from an abnormal bony elongation of histologically normal bone.³ The main clinical feature is a progressive, painless limitation of mouth opening, widely accepted to be due to an impingement of the elongated process on the posterior aspect of the zygomatic arch.⁴ Unilateral cases are more common than bilateral ones, and a number of theories have been postulated as to the pathogenesis.⁵ These include developmental causes, inflammatory reactions, endocrine influences, neoplasia, and traumatic events.⁶ Within the available literature, a number of authors have outlined an association of coronoid hyperplasia with direct trauma to the zygomatic complex.⁷ The following report describes an unusual case of unilateral coronoid hyperplasia presenting in early childhood, which we suggest may be related to an incident of indirect trauma.

Case report

A seven-year-old girl was referred to the oral and maxillofacial department due to an

apparent facial asymmetry and restricted mouth opening. On examination, she was comfortable and healthy. Furthermore, she was not at this time experiencing any particular functional difficulty. Clinical examination revealed deviation to the right hand side on mandibular opening, with an interincisal distance of 15mm. The limitation in mouth opening was painless and there was no associated dysfunction of the temporomandibular joints. A history of trauma at age three years was reported and clinically she had a chin scar.

Orthopantomography showed an elongation of the right coronoid process. No evidence of condylar pathology was observed (Figure 1). Subsequently, a CT scan with appropriate three-dimensional reconstruction demonstrated gross enlargement of the right coronoid, with impingement upon the inner aspect of the zygomatic bone (Figures 2 and 3).

Due to the young age of the patient at initial presentation and the lack of relative functional problems, immediate surgical intervention was not undertaken. On further assessment following new referral at 11 years old, the patient was now more aware of her facial appearance and was concerned with the obvious asymmetry on opening. She was also distressed regarding her restricted mouth opening, recorded clinically at 15mm. Further radiographic examination showed no excessive change in the enlarged coronoid process (Figure 4).

Under general anaesthesia, a right coronoidectomy was performed via an

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FIGURE 1: OPG at aged seven showing no signs of condylar pathology.

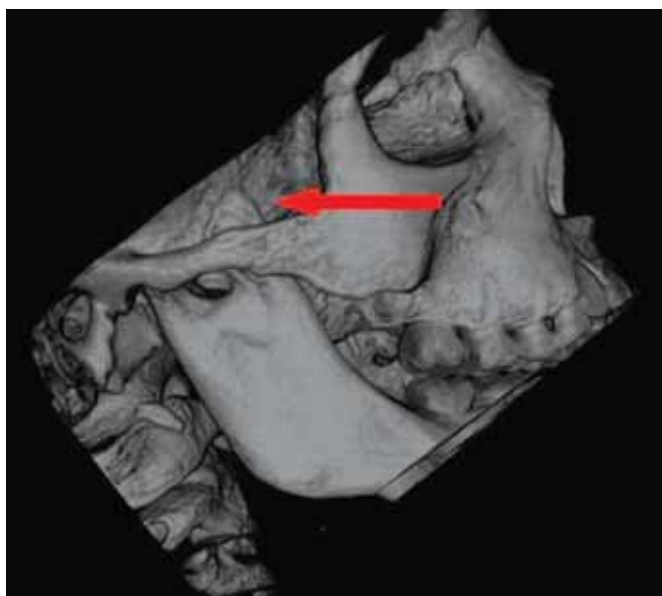


FIGURE 2: 3D scan showing hyperplasia of the coronoid process.

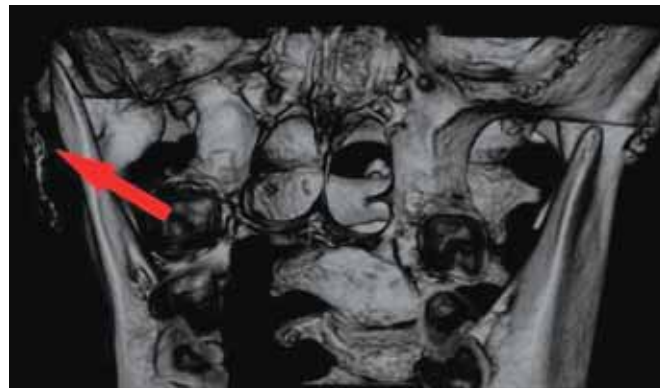


FIGURE 3: 3D CT scan demonstrating proximity of enlarged coronoid process to the inner aspect of the zygomatic arch.

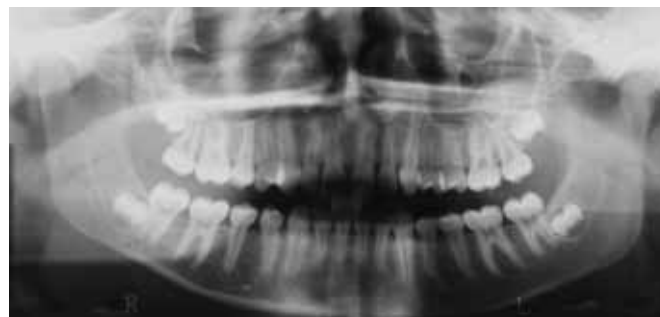


FIGURE 4: OPG at aged 11.

intraoral approach. A standard sagittal split incision was made along the anterior border of the mandibular ramus, and the tissues reflected to expose the top of the coronoid process. Blunt dissection was used to detach the temporalis muscle and a horizontal osteotomy was made from the sigmoid notch to the anterior border of the ascending ramus. The entire right coronoid process was then removed, bleeding controlled, and the surgical site closed with resorbable sutures. Opening improved to 30mm at operation. Postoperative recovery was uneventful and mouth-opening exercises were commenced. The bone appeared histologically normal.

Discussion

Restricted mouth opening caused by coronoid hyperplasia is rare, and so is often overlooked or misdiagnosed.¹ Limitation in mandibular movements logically draws the clinician towards the temporomandibular joint; nonetheless, pathoses of the coronoid process anterior to the joint may well be the origin of the problem. Despite several case reports within the literature, the aetiology of

coronoid hyperplasia remains under discussion. The mean age of presentation is 25 years.³ Unilateral coronoid enlargement is seen more frequently than bilateral cases,⁸ and it had been suggested that the former differs in distinction to the bilateral condition, as its bone displays neoplastic growth or cartilaginous changes.⁹ This theory was disputed by the work of McLoughlin *et al.*, who provided evidence of histologically normal bone in both unilateral and bilateral cases of coronoid hyperplasia.³ Numerous causal factors have been proposed in the development of the condition. Hyperactivity of the temporalis muscle causing reactive elongation of the coronoid process has been hypothesised,¹⁰ as has an association with long-standing temporomandibular joint dysfunction. The latter theory was put forward by Isberg *et al.*, who explained that hyperplasia of the coronoid process may result from internal joint dysfunction due to an increased pull of the temporalis muscle without adequate counterbalance from the condylar region.¹¹ Facial trauma has been implicated within the literature, with particular reference to zygomatic arch injury.⁷

Although plain radiographs are useful in illustrating enlargement of the coronoid process, three-dimensional reconstructions from computerised tomography are by far the superior aids to diagnosis, as the exact shape and relation to surrounding bones can be clearly established.¹² Treatment by an intraoral coronoidectomy is generally accepted to be the preferred surgical approach,¹³ with adequate postoperative physiotherapy key to the success of management.¹⁴ This case outlines an unusual presentation of coronoid hyperplasia, with an early age of onset and a history of trauma to the chin region some years earlier. We assume that there may indeed be a causal relationship between the events. It is likely the trauma caused a fracture of the coronoid process with temporalis muscle pulling the fracture ends apart, acting in a similar fashion as distraction osteogenesis. Uninterrupted development of the unaffected side leads to lengthening and shifting of the mandibular midline to the affected side.¹⁵

Conclusion

Despite its relatively rare occurrence, coronoid hyperplasia should not be ignored in the differential diagnoses of patients presenting with reduced mandibular opening. Dental clinicians should be aware of the possibility of coronoid pathoses when encountering patients with painless restriction in mouth opening. If suspected, referral to a specialist practitioner is appropriate. With the routine availability of computerised tomography to assist clinical examination, coronoid hyperplasia is an easily detectable condition and should not go undiagnosed.¹⁶

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