Dental Traumatology

Dental Traumatology 2009; **25**: 626–630; doi: 10.1111/j.1600-9657.2009.00830.x

Unilateral coronoid hyperplasia following trauma: a case report

CASE REPORT

Syed Iqbal[1], Abdul Latif Abdul Hamid[1], Kathiravan Purmal[2]

[1]Hospital Seremban, Jalan Rasah, Seremban, Malaysia; [2]Department of General Dentistry and Oral & Maxillofacial Imaging, Dental Faculty, University Malaya, Kuala Lumpur, Malaysia

Correspondence to: Kathiravan Purmal, Department of General Dentistry and Oral & Maxillofacial Imaging, Dental Faculty, University Malaya, 50603 Kuala Lumpur, Malaysia
Tel.: +60 3 79674555
Fax: +60 3 79674575
e-mail: drkathi@um.edu.my

Accepted 8 August, 2009

Abstract – This case describes surgical correction of unilateral coronoid hyperplasia in a 13-year-old boy due to trauma. Treatment included coronoidectomy by intraoral approach after the diagnosis was confirmed. Computed tomography scan and occipitomental view radiograph were utilized for investigation. The resected coronoid process was sent for histopathological study. The histopathological examination revealed decalcified sections showing osteochondroid hyperplasia at the expanded end. On clinical and radiographic review at 2 years postoperation, the patient was well, completely symptom free and had improved mouth opening. This case report demonstrates the management of sports injury related unilateral coronoid hyperplasia. Emphasis was also placed on postoperational rehabilitation to prevent any relapse.

Coronoid hyperplasia can be defined as an abnormal bony elongation of a histologically normal coronoid process (1). The main clinical finding is a progressive limitation of mouth opening. According to McLoughlin et al. (1) it was first described in 1899 by Jacob. Coronoid hyperplasia is often confused with other entities like osteoma, exostoses, osteochondroma and temporomandibular joint disorders (2).

This condition may occur as a unilateral or bilateral hyperplasia although clear-cut difference between them is obscure. This is because a unilateral coronoid hyperplasia may often have some abnormalities on the contra lateral side (1). Hence a continuous spectrum exists between them rather than being two separate conditions. This condition is thought to be of developmental in origin with an excessive degree of growth occurring in the coronoid process of the mandible, which enlarges to impinge on the posterior aspect of the zygomatic bone to restrict mouth opening (3). No facial asymmetry is noted in most of bilateral cases but may be present in unilateral coronoid enlargements (4).

Coronoid hyperplasia is multiracial with cases occurring in Caucasian, Negroes and Asians. It largely affects male with male:female ratio of 5:1 and with bilateral:unilateral ratio of 4.7:1 (1). The youngest patient reported with coronoid hyperplasia was a neonate (5).

Many postulations have been forwarded for the pathogenesis of coronoid hyperplasia like developmental causes (6), activity of the temporalis muscle (7), trauma (8, 9), endocrine influences (10), association with syndromes like trismus-pseudocamptodactyly (11) and nevoid basal cell carcinoma (12). There is no case report on sports injury causing coronoid hyperplasia in the literature so far. The current report describes a case of unilateral coronoid hyperplasia which was caused by sports injury and treated with coronoidectomy.

Case report

A 13-year-old Malay boy was referred to our clinic with a complaint of pain and swelling at the left malar region. The patient had a sports related injury, whereby a ball hit him below the left zygomatic bone 5 years ago. Ever since then, the patient had noted the gradual development of deformity and pain on opening the mouth (Fig. 1a–c). On examination, he was comfortable and healthy. Facial asymmetry was noted with swelling on the left malar region (Fig. 1b). The swelling was marked on mouth opening as a 'joint like' movement was observed. Mouth opening was noted to be about 30 mm with no complaints of obvious restriction in opening. The mandible appeared to be deviated to the left side on opening of the mouth. On palpation the swelling was bony hard and there was mild tenderness over the left zygoma (malar) and zygomatic arch region. There were no complaints over both the temporomandibular joints. Intra-orally the maxilla and mandible was firm with no occlusal derangement. Mild tenderness was noted over the left upper buccal sulcus although the mucosa was normal.

An occipitomental (15°) radiograph view was taken. This revealed an obvious elongation of the left coronoid process and an enlargement of the tip of the coronoid process (Fig. 2a). Axial and coronal sections of the computed tomography (CT) scan revealed that the left zygomatic bone appeared prominent and there was bowing of the arch antero-posteriorly (Fig. 2b–d). The left coronoid process was elongated and enlarged

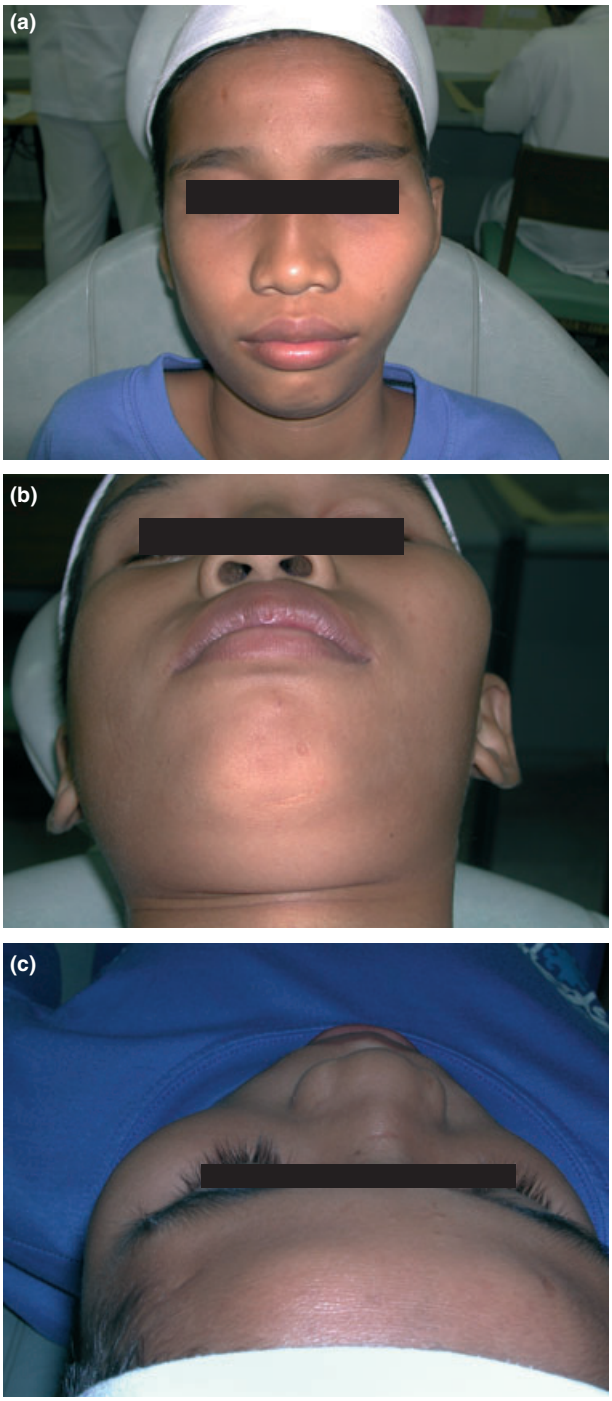

Fig. 1. (a) Preoperative frontal view showing facial asymmetry; (b) preoperative view showing prominent left zygoma; (c) preoperative view from the top showing swelling on the left zygoma region.

resembling a condyle. There was no fusion between the elongated coronoid process and the zygomatic arch (Fig. 2d). Both TMJ were symmetrical and normal. There was no radiographic evidence of presence of any neoplastic growth in the coronoid process (Fig. 2a–d).

Based on the clinical and radiographic findings a diagnosis of left unilateral coronoid hyperplasia was made. The patient and his parents were told about the condition and they agreed to surgical removal of the hyperplastic bone. A decision of intra-oral approach was taken as mouth opening was not a problem and to avoid any external scars.

After nasal intubation, a standard sagittal split osteotomy incision was placed on mucosa overlying the mandible. The incision was carried on the anterior aspect of the ramus and the tissues were retracted up the coronoid process. A forked retractor was used on the anterior aspect of the ramus and pulled upwards. As this was done some of the temporalis muscle attachments were detached until the sigmoid notch was identified. The coronoid was then held with a heavy bone holding forceps. An oscillating electric saw was used to cut the coronoid towards the sigmoid notch. Once the coronoid had been separated, it was pulled down with the forceps and remainder of the temporalis attachments were stripped. The hyperplastic bone was then delivered intra-orally. Bleeding was controlled and closure was done with vicryl 3/O sutures. The sectioned coronoid process resembled a condylar head and had a pearly white structure resembling cartilage. The size was 3 cm × 1.5 cm (Fig. 3a, b). The histopathological examination revealed decalcified sections showing osteochondroid hyperplasia at the expanded end. There was no evidence of malignancy. Vigorous mouth opening exercises were commenced immediately on the third postsurgical day. He was discharged and followed up regularly at our clinic.

At the 2 years follow up, the patient did not have any complaints of pain or discomfort when opening the mouth. His mouth opening was about 3.5 cm. The face was symmetrical (Fig. 4a). Occipito mental radiograph was taken to confirm there is no regrowth of the condylar process (Fig. 4b).

Discussion

Coronoid hyperplasia is usually associated with progressive and painless limitation of mouth opening (13). Lateral and forward movements of the mandible may be restricted due to the obstruction of the enlarged coronoid process against the zygomatic arch. This could be more evident on maximum mouth opening.

However in the unilateral case the above problems may not be so marked. Other clinical signs like a mobile lump above zygomatic arch, facial asymmetry and pain on opening the mouth may be present (13) as was the case in our patient.

Hall et al. (14) and Rowe (10) had postulated that a unilateral coronoid hyperplasia differs from a bilateral coronoid hyperplasia as its bone displays neoplastic growth or cartilaginous changes. However this notion was completely disputed because McLoughin et al. (1) showed that unilateral and bilateral coronoid hyperplastic bone to be of histologically normal bone. Lyon & Sarnat (7) reported that bone compression at the zygoma–coronoid contact induces formation of chondrocytes. This explains the cartilaginous changes of the coronoid hyperplastic tip. Therefore these changes cannot be accepted as neoplastic growth. This was the situation with our patient.

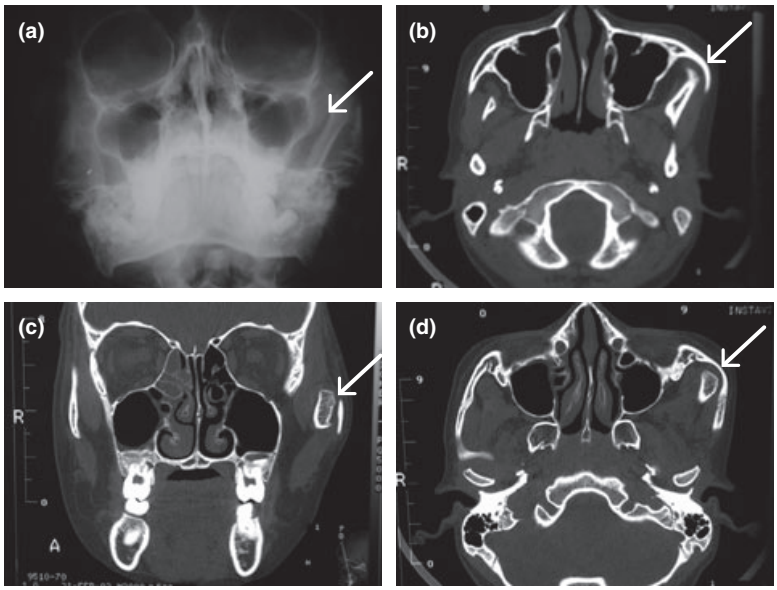

Fig. 2. (a) Occipitomental view 15° showing elongated left coronoid process (arrow); (b) axial view of the CT scan showing the enlarged coronoid process (arrow); (c) coronal view of the CT scan showing the coronoid process (arrow); (d) higher axial view of the CT scan showing there is no fusion between the coronoid process and the zygomatic arch (arrow).

The pathogenesis of coronoid process hyperplasia is not well understood. Studies have indicated that there is a significant relationship between temporomandibular joint (TMJ) disorders and coronoid hyperplasia. Isberg et al. (15) noted eight patients with coronoid hyperplasia, four of which had long standing internal derangement of the temporomandibular joint. They went on to explain that dysfunction of the joint leads to an increased pull or activity of the temporalis muscle without a counter balance from the condylar region. This resulted in an increase in the size of the coronoid process. A similar argument was put forward by Sarnat & Engel (16). In our case both the TMJ was normal under CT scan examination. Moreover, the temporalis activity theory was disputed by Hall et al. (14). They reported that electro-myographic examination in patients with coronoid hyperplasia to be normal. Similar electromyographic examinations by Gerbino et al. (17) confirmed normal activity of the muscle. On the other hand, Shira & Lister (18) suggested that there could be a developmental abnormality in which the cartilaginous growth centres in the coronoid process persist causing continued growth and hyperplasia. Smyth & Wake (3) reported an association of coronoid hyperplasia with tissue trauma following the removal of a patient's adenoids. This was concurrent with the timing of limitation mouth opening. They postulated that the procedure could have torn the tendinous insertion of the temporalis muscle on the coronoid. Subsequent intramuscular haematoma and its organization can lead to formation of bone around the coronoid. There are a few reports of coronoid hyperplasia associated with facial trauma (4, 8, 9, 13, 19), but none on sports related injury. In the present case, there was a history of facial trauma following a sports injury that occurred some 5 years prior to his presentation. Although the management of this sports related injury is similar to unilateral condylar hyperplasia due to trauma, we hope the clinicians will be aware sports injury can cause such problems.

Plain radiographs appear to be the initial step in the diagnosis of coronoid hyperplasia, with orthopantomography (OPT) and occipitomental (OM) view being the favourites. OM view is useful in demonstrating the relationship between the coronoid and the zygoma while OPT is able to clearly show the coronoid enlargement. However radiographs are not able to reveal the spatial relationship between the coronoid enlargement and the zygoma, let alone the changes in the malar bone. Kubota et al. (20) suggested the use of Levandoski analysis from the OPT views to determine the presence of coronoid hyperplasia.

With the advent of computerized tomography (CT), better and more accurate means of accessing coronoid hyperplasia has been achieved (21). An axial and coronal CT imaging in the position at the level of coronoid, zygomatic arch and the upper third molar gives the most information (22). By detecting this proximity or impingement other possibilities of restriction of mouth opening can be eliminated. A three dimensional CT scan can even be used to determine the surgical approach (2, 23).

The treatment for coronoid hyperplasia, which presents as a mechanical problem, is mainly surgical. The surgical approach could be either intraoral or extraoral. An intraoral approach for coronoidectomy has been the preferred mode in about 90% of reported cases (1). Gerbino et al. (17) have reported success in intraoral coronoidotomy even though an earlier report by Allan et al. (24) disagreed with this procedure due to rapid fusion of the sectioned coronoid and the ramus. Hayter & Robertson (25) advocated and described the extraoral approach via a coronal flap for bilateral cases and a modified Al-Kayat & Bramley (26) incision for the unilateral cases. There was also a report of endoscopically assisted removal of unilateral coronoid hyperplasia approached via a hair-bearing temporal skin incision (27). This would avoid the external scar and possible trauma to the facial nerve in the extraoral approach. We used the intraoral approach because mouth opening was

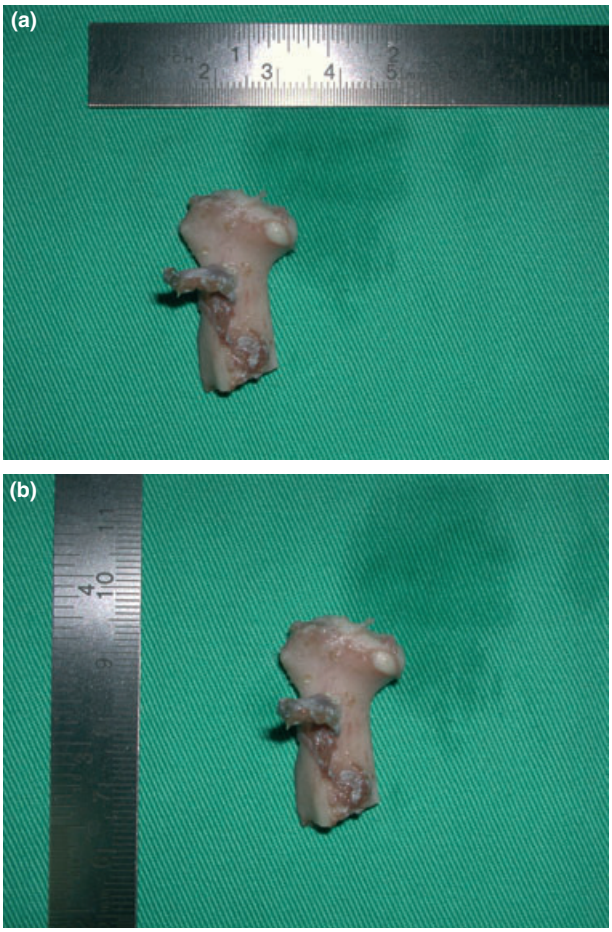

Fig. 3. (a) The sectioned coronoid process; (b) sectioned coronoid process showing whitish cartilaginous like covering at the tip.

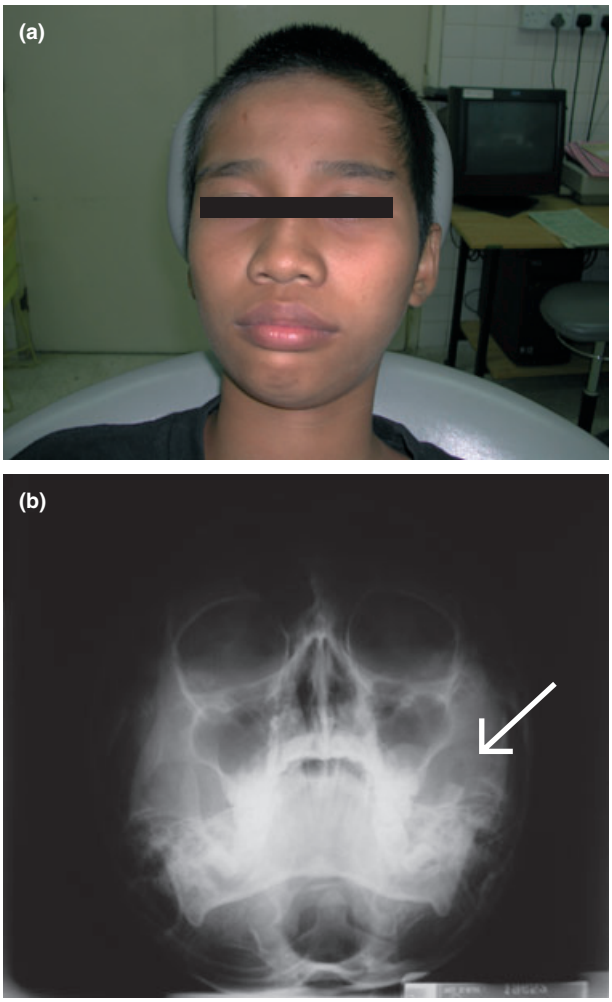

Fig. 4. (a) Follow up after 2 years, face is symmetrical; (b) occipitomental view showing the postoperative view after 2 years. Arrow shows the resected coronoid process without any signs of regrowth.

not limited and we could gain good access to the surgical site.

The timing of the surgery is always a case for arguments. Rivas (28) recommends surgery is delayed until growth has ceased in order to avoid the possibility of a second operation at a later date to correct any recurrent deformity. We proceeded to operate on this 13-year-old boy because the coronoid process may enlarge causing further disfigurement and there was pain on opening the mouth.

Postoperative relapse is a cause for concern. McLoughin et al. (1) suggested that this could be due to a haematoma formation and subsequent fibrosis around the surgical site or the persistence of the causes that initially attributed to the coronoid hyperplasia. Bisphosphonates has been used successfully in restricting bone regrowth over hyperplastic site. This is due to its ability in suppressing the levels of alkaline phosphatase, reduction of bone turnover and inhibitory mitotic activity of osteoclasts (3).

However, postoperative success also lies on physiotherapy (29, 30). Gerbino et al. (17) strongly emphasized on dynamic physiotherapy immediately after surgery and continued for 12 months. Totsuka & Fukuda (31) promoted the use of short wave diathermy, massage and stretching exercise of the masticatory muscle. In the present case, mouth opening exercise was initiated with the use of wooden spatulas from the third postsurgical day. At the 2 years follow up the patient's mouth opening was noted to be improved and there was no signs of regrowth of the coronoid process (Fig. 4b) although there have been reports of regrowth and surgically induced fibrosis in the literature (32). Therefore a longer follow-up would be necessary for this patient.

Conclusion

This case report highlights one of the reasons for facial asymmetry. The clinician should be aware that unilateral condylar hyperplasia can cause facial asymmetry with or without limitation in mouth opening. Sports related injury has been put forward as one of the aetiology for this condition. Proper investigations, i.e. OM view radiographs and CT scans are essential for the diagnosis

and treatment planning. Postoperative rehabilitation should be an integral part of the treatment.

Acknowledgement

We would like to express our gratitude to the Director of Oral Health Division, Ministry of Health, Malaysia for allowing us to use the records of this patient for publication.

References

1. 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.
2. Hernandez-Alfaro F, Escuder O, Marco V. Joint formation between an osteochondroma of the coronoid process and the zygomatic arch (Jacob disease): report of case and review of literature. J Oral Maxillofac Surg 2000;58:227–32.
3. Smyth AG, Wake MJ. Recurrent bilateral coronoid hyperplasia: an unusual case. Br J Oral Maxillofac Surg 1994;32:100–4.
4. Alan W, Macleod G. Limitation of mandibular opening due to enlarged coronoid processes. Aust Dent J 1987;32:120–5.
5. 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.
6. York BV, Cockerham S. Bilateral hyperplasia of the coronoid processes in siblings. Oral Surg Oral Med Oral Pathol 1983;56:584–5.
7. Lyon LZ, Sarnat BG. Limited opening of the mouth caused by enlarged coronoid processes: report of case. J Am Dent Assoc 1963;67:644–50.
8. Javid B. Unilateral hyperplasia of the coronoid process of the mandible. Case report. Int J Oral Surg 1981;10:145–7.
9. Colquhoun A, Cathro I, Kumara R, Ferguson MM, Doyle TC. Bilateral coronoid hyperplasia in two brothers. Dentomaxillofac Radiol 2002;31:142–6.
10. Rowe NL. Bilateral developmental hyperplasia of the mandibular coronoid process. A report of two cases. Br J Oral Surg 1963;1:90–104.
11. Azaz B, Zeltser R, Nitzan DW. Pathoses of coronoid process as a cause of mouth-opening restrictions. Oral Surg Oral Med Oral Pathol 1994;77:579–84.
12. Leonardi R, Sorge G, Caltabino M. Bilateral hyperplasia of the mandibular coronoid processes associated with the nevoid basal cell carcinoma syndrome in an Italian boy. Br Dent J 2001;190:349–50.
13. 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.
14. Hall RE, Orbach S, Landesberg R. Bilateral hyperplasia of the mandibular coronoid processes: a report of two cases. Oral Surg Oral Med Oral Pathol 1989;67:141–5.
15. 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;63:275–9.
16. Sarnat BG, Engel MB. A serial study of mandibular growth after removal of the condyle in the Macaca rhesus monkey. Plast Reconstr Surg (1946). 1951;7:364–80.
17. Gerbino G, Bianchi SD, Bernardi M, Berrone S. Hyperplasia of the mandibular coronoid process: long-term follow-up after coronoidotomy. J Craniomaxillofac Surg 1997;25:169–73.
18. Shira RB, Lister RL. Limited mandibular movements due to enlargement of the coronoid processes. J Oral Surg (Chic) 1958;16:183–91.
19. Cooper JC, Flinch LD. Coronoid osteochondroma presenting as a corono-zygomatic ankylosis. Br Dent J 1974;123:99–101.
20. Kubota Y, Takenoshita Y, Takamori K, Kanamoto M, Shirasuna K. Levandoski panographic analysis in the diagnosis of hyperplasia of the coronoid process. Br J Oral Maxillofac Surg 1999;37:409–11.
21. Masahiro I, Yoshiko A, Kenichi K. 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 Oral Radiol Endod 2005;99:93–100.
22. Gross M, Gavish A, Calderon S, Gazit E. The coronoid process as a cause of mandibular hypomobility – case reports. J Oral Rehabil 1997;24:776–81.
23. Asaumi J, Kawai N, Honda Y, Shigehara H, Wakasa T, Kishi K. Comparison of three-dimensional computed tomography with rapid prototype models in the management of coronoid hyperplasia. Dentomaxillofac Radiol 2001;30:330–5.
24. Allan PG, Reade PC, Steidler NE. Healing following coronoidotomy in rats. Int J Oral Maxillofac Surg 1989;18:109–13.
25. Hayter JP, Robertson JM. Surgical access to bilateral coronoid hyperplasia using the bicoronal flap. Br J Oral Maxillofac Surg 1989;27:487–93.
26. Al-Kayat A, Bramley P. A modified pre-auricular approach to the temporomandibular joint and malar arch. Br J Oral Surg 1979;17:91–103.
27. Mavili E, Akyurek M, Kayikcioglu A. Endoscopically assisted removal of unilateral coronoid process hyperplasia. Ann Plast Surg 1999;42:211–6.
28. Rivas PH. Reduction of mouth opening due to mandibular coronoid process enlargement. Oral Surg 1979;47:131–4.
29. Ferro MF, Sanroman JF, Gutierrez JS, Lopez AC, Sanchez L, Perez AE. Treatment of bilateral hyperplasia of the coronoid process of the mandible. Presentation of a case and review of the literature. Med Oral Patol Oral Cir Bucal 2008;13:E595–8.
30. Gibbons AJ, Abulhoul S. Use of a Therabite appliance in the management of bilateral mandibular coronoid hyperplasia. Br J Oral Maxillofac Surg 2007;45:505–6.
31. Totsuka Y, Fukuda H. Bilateral coronoid hyperplasia. Report of two cases and review of the literature. J Craniomaxillofac Surg 1991;19:172–7.
32. Satoh K, Ohno S, Aizawa T, Imamura M, Mizutani H. Bilateral coronoid hyperplasia in an adolescent: report of a case and review of the literature. J Oral Maxillofac Surg 2006;64:334–8.