CASE REPORT

Haemophilic pseudotumour in two parts of the maxilla: case report

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Haemophilic pseudotumour is a rare disease that occurs most often in femur, tibia, ilium or pelvic bone of a patient with haemophilia. Thus far, there have been only 31 reported cases in jaw bones and paranasal sinuses. Among them, the mandible is a more common site than the maxilla or paranasal sinuses. Here, we report a case of haemophilic pseudotumour in two parts of the maxilla. Contrast-enhanced CT showed an expansive and thinly corticated lesion with fluid attenuation at the left anterior maxilla which seemed like a post-operative maxillary cyst, ameloblastoma or odontogenic cyst. In addition, the thickened left palatal process of the maxilla seemed like fibrous dysplasia or intraosseous vascular malformation. Since haemophilic pseudotumour is not pathognomonic in radiological findings, when a patient who suffered from haemophilia or had taken anticoagulating agents has jaw lesion, haemophilic pseudotumour should be included in a differential diagnosis.

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Introduction

A haemophilic pseudotumour is a rare disease that occurs in a patient who suffers from either haemophilia A or haemophilia B.¹ Haemophilia A and haemophilia B are X-linked recessive disorders caused by insufficient Factor VIII and Factor IX, respectively.² A lack of these coagulating factors causes continuous haemorrhage and haemorrhage-related diseases.²

Thus far, no aetiology has been established for haemophilic pseudotumours, but some researchers have reported that bone destruction might be caused by the secondary pressure of haemorrhage in the subperiosteal or soft tissue and be followed by reactive new bone formation.^{3,4}

Several classifications for these lesions have been proposed; the lesions have been categorized into Types

I, II and III and referred to accordingly in the literature.^{5,6} Type I occurs in the soft tissue, Type II in the subperiosteal region and Type III in the bone tissue; a combination of Types II and III has also been reported. Gilbert⁷ proposed a new classification of these lesions into two groups: one included proximal lesions such as lesions in the femur and pelvis, and the other consisted of distal lesions such as lesions on the hands and feet. Of these groups, proximal haemophilic pseudotumours are known to be more common.

Haemophilic pseudotumours are mainly found in the femur, pelvic bone and tibia but may also, rarely, develop in the jaw bone. Most researchers have agreed that the mandible is a more prominent site of haemophilic pseudotumours than the maxilla and paranasal sinus.

Because of the possibility of extensive haemorrhage, biopsy or aspiration of the lesion is not recommended.

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The common radiographic finding for haemophilic pseudotumours is an expansive radiolucent osteolytic lesion.⁸ However, various features associated with the type and size of the lesion can be found. On the basis of radiographic findings, a haemophilic pseudotumour can be misdiagnosed as a giant cell tumour, aneurysmal bone cyst, osteosarcoma, ameloblastoma, myxoma or other lesion.^{8,9}

The aim of this report was to present a case of a haemophilic pseudotumour that occurred in two parts of the maxilla. An expansive and thinly corticated lesion with fluid attenuation at the left anterior maxilla mimicked a post-operative maxillary cyst, ameloblastoma or odontogenic cyst radiologically. In addition, the thickened left palatal process of the maxilla seemed like fibrous dysplasia or intraosseous vascular malformation.

Case report

A 53-year-old male patient visited Seoul National University Dental Hospital (SNUDH), Seoul, Korea, with complaints of pain and swelling in the left facial area. The patient had a history of trauma in the maxillary area about 20 years earlier, and he had an uncertain memory of operation in that region. Furthermore, he had been aware that he had haemophilia since his childhood. He had undergone surgery in both knees due to an intraosseous haemangioma about 30 years previously and a reoperation in the left knee due to a recurrent lesion 4 years earlier. During the medical evaluations for his second operation, the

patient had been referred to the Department of Hematology-Oncology of Seoul National University Hospital, Seoul, Korea for an investigation of the state of his haemophilia. The blood test had shown isolated activated partial thromboplastin time prolongation of 50.7 s (normal range: 29–45 s) and 4% Factor IX, which indicated moderate haemophilia B (0–1% severe, 1–5% moderate and 5–30% mild).

Meanwhile, he had visited Seoul National University Dental Hospital several times for routine dental checkups in the past 3 years; he had visited the Department of Prosthodontics for maxillary partial denture repair and the Department of Periodontics for scaling and curettage. There were not any teeth other than the right maxillary first molar, and left maxillary first and second molars. Before every invasive treatment, Factor IX formulation was infused intravenously to ascend Factor IX level to approximately 30% to control excessive haemorrhage.

When the patient visited SNUDH again for plaque control, he complained about the toothache in the left upper teeth in addition to swelling and tenderness on the left facial and infraorbital area. The patient was referred to the Department of Oral and Maxillofacial Surgery of SNUDH. Physical examination revealed that there was tenderness on pressure with no local fever in the left facial area. Intraorally, there was no specific clinical finding related to the symptoms. However, in the intraoral image of 3 years ago, there were periapical rarefactions and periodontitis on the left maxillary first and second molars (Figure 1a). The patient was clinically diagnosed with a cellulitis with abscess in the left maxillary area, and then

Figure 1 (a) A periapical radiograph shows periapical rarefaction and periodontitis on the left maxillary first and second molars. (b) A cropped panoramic radiograph shows the elevation of the left hard plate and floor of the nasal cavity (double headed arrows). The left medial border of maxillary sinus is displaced in the direction of the nasal cavity (arrows). The oblique line in the left maxillary sinus is also observed (arrowheads).

intraoral irrigation and drain insertion were performed under local anaesthesia. The swelling had subsided with daily dressing and antibiotic agent administration, and 4 days later, the pain was relieved. However, there was no pus discharge, redness or local heat caused by bacterial infection, and at the 1-month follow-up visit, the swelling remained with no pain in the area.

For further examination, panoramic radiography and contrast-enhanced CT were performed. The panoramic radiograph showed the elevation of the left hard plate and the floor of the nasal cavity (Figure 1b). On contrast-enhanced CT, an expansive and thinly corticated lesion with fluid attenuation was observed in the anteromedial portion of the left maxillary sinus. The lesion had a lobulated margin and pushed the infraorbital rim upwards. The lesion compressed the nasolacrimal duct posteriorly (Figure 2a,b). Considering these radiographic findings and the patient's history, the preliminary diagnosis included a post-operative maxillary cyst, ameloblastoma and

odontogenic cysts. In addition, the left palatal process of the maxilla was thicker than that on the right side with slightly altered trabecular bone pattern, and the nasal floor was located higher on the coronal CT image (Figure 2a,c). These radiographic findings seemed like a fibrous dysplasia or intraosseous vascular malformation.

The level of Factor IX had to be elevated to 100% before surgery to prevent extensive haemorrhage during surgery and be maintained at 50% after surgery for at least 7 days. The patient, therefore, underwent enucleation of the lesion with replacement of Factor IX to 100%.

Histopathological findings revealed that the lesion was composed of two parts: an intraosseous lesion and a soft-tissue lesion. In the soft-tissue lesion, there was haemorrhage surrounded by a fibrous capsule under the mucosal epithelium of the maxillary sinus. In the intraosseous lesion, the foci of the haemorrhage were scattered within the fibrous connective tissue.

Figure 2 (a) Coronal contrast-enhanced CT scan with bone window setting demonstrates different heights of both nasal floors (double-headed arrows). The lobulated and thinly corticated lesion pushes the lateral border of the nasal cavity (arrows). (b) Axial contrast-enhanced CT scan shows the expansive lesion with fluid attenuation in the anteromedial wall of the left maxillary sinus. The lesion compresses the nasolacrimal duct posteriorly (arrow). These radiographic findings mimic a post-operative maxillary cyst, ameloblastoma or odontogenic cyst such as keratocystic odontogenic tumour. (c) On the axial contrast-enhanced CT scan with bone window setting, another part of the lesion with slight altered trabecular bone pattern is observed on the palatal process of the maxilla. The nasopalatine duct is dislocated to the right side (arrow). The lesion mimics fibrous dysplasia or intraosseous vascular malformation. (d) On the sagittal contrast-enhanced CT scan with bone window setting, the cystic lesion A is adjacent to the intraosseous lesion B, and P is the pterygoid plate. (e) Axial contrast-enhanced CT scan with bone window setting demonstrates the septum in the left maxillary sinus which appears on the panoramic radiograph as an oblique line (arrowheads).

Figure 3 Photomicrograph of a histopathological slide. (a) Soft-tissue lesion. Under the mucosal epithelium of the maxillary sinus (arrow), haemorrhage surrounded by a fibrous capsule was observed [haematoxylin and eosin (H&E) stain, $\times 100$]. (b) Intraosseous lesion. Between the bone tissues, the foci of the haemorrhage were scattered within the fibrous connective tissue (H&E stain, $\times 40$).

New bone formation was observed adjacent to the lesion. Numerous histiocytes suggestive of haemosiderin-laden macrophages were found throughout the lesion (Figure 3). The patient was confirmed to have a haemophilic pseudotumour on the basis of these histopathological findings. After 3 months, the patient reported no specific discomfort (Figure 4).

Discussion

In this report, we briefly summarized our case and the previous 24 studies,^{1,3-6,8-26} which included 32 cases, on haemophilic pseudotumours of the jaw. Table 1 shows the summary of studies conducted between 1968 and 2014, including the present case. Among these cases, 8 cases of lesions occurred in the maxilla and 4 cases of lesions in the paranasal sinus. The remaining 20 cases were of lesions located in the mandible.

The incidence of haemophilic pseudotumours is known to be 1–2% of severe haemophilia cases,²⁷ and Xue *et al*²⁵ reported that the incidence of haemophilic pseudotumours in Chinese patients was 1.12% (14 patients among 1248 patients with haemophilia).

The age range of haemophilic pseudotumour patients varied from 6 months to 53 years. The previous oldest patient was 34 years old,⁶ and in our case, the patient was 53 years old. Thus, we confirmed that a haemophilic pseudotumour could occur at any age.

The severity of haemophilia also had a diverse range from mild to severe. There were two reported cases of patients who were not suffering from haemophilia. One of them took anticoagulant agents due to a cardiac disorder,²⁰ and the other patient had no medical history.²¹

Among patients with mild to severe haemophilia, many had a history of haemorrhage-related disease in different sites such as the hand, tibia, elbow joint, foot, radius and knee.^{5,10,11,17,18,22,23} Our patient also had a history of haemangioma on the knee.

The proportion of patients who had a history of trauma was 37.5% (12 patients of 32), which revealed that trauma was not a necessary and sufficient factor but an aetiological factor. Haemophilic pseudotumours can

be caused not only by traumatic falls and strikes but also by mumps,¹² incisional biopsies¹⁶ and extractions.¹⁷

On the basis of radiographs, a haemophilic pseudotumour could be often misdiagnosed as a benign myxoma, giant cell tumour, aneurysmal bone cyst, osteosarcoma, ameloblastoma or similar lesion^{8,9} because this disease was not pathognomonic.

In the previous 31 cases, the imaging feature was usually an expansive radiolucent lesion. Other patterns of imaging features including an extensive fine trabecular bone pattern,¹² spicules perpendicular to a lesion,¹³ fluid–fluid level in an osteolytic lesion¹⁹ and periosteal strut from the cortex into a soft-tissue mass,^{4,8} only new bone formation without an osteolytic portion⁶ and flecks of calcification inside the lesion^{1,22} have also been described in the previous reports.

These various radiographic findings can be explained by the three types of lesions mentioned above.^{5,6} Type I lesions are difficult to find in plain radiographs since they occur in the soft tissue. In Type II lesions, repeated haemorrhage forms an encapsulated area in the subperiosteal region; secondarily, these lesions may resorb the adjacent bone with periosteal bone formation. Most expansive radiolucent lesions, including our case of the anterior maxilla, could be explained by this mechanism. This cyst-like portion in our patient was mentioned that it seemed like a post-operative maxillary cyst. To diagnose a post-operative maxillary cyst, the patient's history of the maxillary surgical intervention is essential.^{28,29} Although there was no evidence of a surgery in CT imaging, our patient had uncertain history of

Figure 4 Post-operative panoramic radiograph shows no pathological change at the operative site.

Table 1 List of the reported cases including our case of a haemophilic pseudotumour of the jaw

Case number	Authors	Year of study	Age (years)	Sex	Site	Severity of haemophilia
1	Lazarovits and Greim ¹⁰	1968	11	M	Mandible	Haemophilia A
2	Stoneman and Beierl ¹¹	1975	13	M	Mandible	Haemophilia A
3	Mulkey ¹²	1977	8	M	Mandible	Severe classic haemophilia
4	Mulkey ¹²	1977	7	M	Mandible	Severe classic haemophilia
5	Mulkey ¹²	1977	16	M	Mandible	Severe classic haemophilia
6	Marquez et al ¹³	1982	13	M	Maxilla	Moderate haemophilia A
7	Martinez-Lage et al ¹⁴	1983	10	M	Mandible	Haemophilia A
8	Brook et al ¹⁵	1985	16 months	M	Mandible	Haemophilia A
9	Bryan et al ¹⁶	1990	2.5	M	Mandible	Haemophilia A
10	de Sousa et al ¹⁷	1995	11	M	Maxilla	Severe haemophilia A
11	de Sousa et al ¹⁷	1995	11	M	Mandible	Mild haemophilia A
12	Zheng and Zheng ¹⁸	1997	26	M	Maxilla	Mild haemophilia A
13	Kale et al ¹⁹	2001	17	M	Mandible	Haemophilia A
14	Gupta et al ⁵	2001	20	M	Paranasal sinus	Haemophilia A
15	Stevenson and Keast ²⁰	2002	75	M	Paranasal sinus	Warfarin (mechanical aortic valve replacement and chronic atrial fibrillation)
16	Steele et al ²¹	2004	6 months	M	Paranasal sinus	No past medical history
17	Rey et al ²²	2007	7	M	Mandible	Mild haemophilia A
18	Rey et al ²²	2007	14	M	Mandible	Mild haemophilia A
19	Rey et al ²²	2007	12	M	Mandible	Mild haemophilia A
20	Lima et al ²³	2008	12	M	Maxilla	Mild haemophilia A
21	Shi et al ⁴	2009	9	M	Mandible	Mild haemophilia A
22	Lee et al ⁶	2009	34	M	Mandible	Moderate haemophilia B
23	Lv et al ²⁴	2011	9	M	Mandible	Mild haemophilia A
24	Srinivasan et al ⁸	2011	7	M	Mandible	Moderate haemophilia B
25	Yoshitake et al ³	2011	5	M	Mandible	Severe haemophilia A
26	Cox et al ⁹	2011	2	M	Mandible	Moderate haemophilia B
27	Xue et al ²⁵	2011	24	M	Maxillary sinus	Moderate haemophilia A
28	Yang et al ¹	2012	1	M	Maxilla	Moderate haemophilia B
29	Yang et al ¹	2012	2	M	Maxilla	Moderate haemophilia A
30	Yang et al ¹	2012	2	M	Maxilla	Moderate haemophilia A
31	Thronsdon et al ²⁶	2012	14	M	Mandible	Severe haemophilia A
32	Our case	2014	53	M	Maxilla (two parts)	Moderate haemophilia B

M, male.

operation of the maxilla. For this reason, post-operative maxillary cyst could not be excluded from the preliminary differential diagnoses. Type III intraosseous lesion can cause bone expansion. According to a report by Lee et al,⁶ bone expansion could be caused by intraosseous haemorrhage due to trauma, and the next healing stage caused by the treatment of haemophilia might lead to new bone formation. After a long interval, the lesion showed only new bone formation. Since our 53-year-old patient had sufficient interval time, the lesion in the palatine process of the maxilla might show the image features of fibrous dysplasia or intraosseous vascular malformation. Although MRI examination was not conducted in our case, MRI might be helpful in the diagnosis of a haemophilic pseudotumour because MRI could enable distinguishing among the various blood clot states.

Pain can occur depending on the size, site of the lesion and existence of secondary infection. Many other patients whose symptoms lasted for more than a month experienced no pain. On the basis of this observation, we could conclude that if the lesion would grow slowly, the lesion could not cause pain due to secondary pressure because the bone remodelling process would also proceed simultaneously. In our patient, pain was relieved after drainage; thus, the pain was probably caused by the cyst-like lesion of the anterior maxilla.

Numerous attempts have been made to treat a haemophilic pseudotumour of the jaw by using treatment modalities such as radiotherapy,^{5,10,20,21} cryoprecipitation,¹¹ curettage,^{3,12,17,26} enucleation,^{14,23} replacement therapy,^{1,3,4,9,15,19,22,25,26} immobilization,¹⁹ ethanol embolization²⁴ and resection.^{4,13} The treatment plan is chosen on a case-by-case basis, and most physicians have accepted that replacement therapy that provides the coagulation factor is a fundamental component of treatment and another appropriate therapy with replacement therapy is effective.^{9,23}

In conclusion, our case of a patient with haemophilic pseudotumour required a differential diagnosis with post-operative maxillary cysts, ameloblastoma, odontogenic cysts and fibrous dysplasia which were relatively common diseases compared with haemophilic pseudotumours. Although the preliminary diagnosis of a haemophilic pseudotumour is usually assumed by the medical history of haemophilia, previous reports showed haemophilic pseudotumour could occur in a patient who has no medical history although low percentage. Therefore, if differential diagnosis from other common mimicking diseases was possible with MR or contrast-enhanced CT through further many studies, it could be helpful to include haemophilic pseudotumour in preliminary diagnosis list.

References

- Yang BT, Wang YZ, Wang XY, Wang ZC. Imaging features of paediatric haemophilic pseudotumour of the maxillary bone: report of three cases and review of the literature. *Br J Radiol* 2012; **85**: 1107–11. doi: <http://dx.doi.org/10.1259/bjr/12938443>
- Hoyer LW. Hemophilia A. *N Engl J Med* 1994; **330**: 38–47. doi: <http://dx.doi.org/10.1056/NEJM199401063300108>
- Yoshitake Y, Nakayama H, Takamune Y, Yasunaga M, Hiraki A, Shinohara M. Haemophilic pseudotumour of the mandible in a 5-year-old patient. *Int J Oral Maxillofac Surg* 2011; **40**: 120–3. doi: <http://dx.doi.org/10.1016/j.ijom.2010.05.013>
- Shi H, Wang S, Wang P, Yu Q. Haemophilic pseudotumour of the mandible. *Dentomaxillofac Radiol* 2009; **38**: 182–4. doi: <http://dx.doi.org/10.1259/dmfr/28176102>
- Gupta S, Mohapatra BB, Ghai S, Seith A, Kashyap R, Sharma R, et al. Haemophilic pseudotumour of the paranasal sinuses: management with radiotherapy and factor replacement therapy. *Haemophilia* 2001; **7**: 595–9. doi: <http://dx.doi.org/10.1046/j.1365-2516.2001.00566.x>
- Lee DW, Jee YJ, Ryu DM, Kwon YD. Changes in a hemophilic pseudotumor of the mandible that developed over a 20-year period. *J Oral Maxillofac Surg* 2009; **67**: 1516–20. doi: <http://dx.doi.org/10.1016/j.joms.2008.08.023>
- Gilbert MS. Characterizing the hemophilic pseudotumor. *Ann N Y Acad Sci* 1975; **240**: 311–15. doi: <http://dx.doi.org/10.1111/j.1749-6632.1975.tb53365.x>
- Srinivasan K, Gadodia A, Bhalla AS, Choudhury AR, Bhutia O, Gupta A, et al. Magnetic resonance imaging of mandibular hemophilic pseudotumor associated with factor IX deficiency: report of case with review of literature. *J Oral Maxillofac Surg* 2011; **69**: 1683–90. doi: <http://dx.doi.org/10.1016/j.joms.2010.07.005>
- Cox DP, Solar A, Huang J, Chigurupati R. Pseudotumor of the mandible as first presentation of hemophilia in a 2-year-old male: a case report and review of jaw pseudotumors of hemophilia. *Head Neck Pathol* 2011; **5**: 226–32. doi: <http://dx.doi.org/10.1007/s12105-011-0267-x>
- Lazarovits P, Griem ML. Radiotherapy of hemophilia pseudotumors. *Radiology* 1968; **91**: 1026–7. doi: <http://dx.doi.org/10.1148/91.5.1026>
- Stoneman DW, Beierl CD. Pseudotumor of hemophilia in the mandible. Report of a case. *Oral Surg Oral Med Oral Pathol* 1975; **40**: 811–15. doi: [http://dx.doi.org/10.1016/0030-4220\(75\)90453-3](http://dx.doi.org/10.1016/0030-4220(75)90453-3)
- Mulkey TF. Hemophilic pseudotumors of the mandible. *J Oral Surg* 1977; **35**: 561–8.
- Marquez JL, Vinageras E, Dorantes S, Nussbaumer C, Flores A, Perez Rolon G. Hemophilic pseudotumor of the inferior maxilla. Report of a case. *Oral Surg Oral Med Oral Pathol* 1982; **53**: 347–50. doi: [http://dx.doi.org/10.1016/0030-4220\(82\)90433-9](http://dx.doi.org/10.1016/0030-4220(82)90433-9)
- Martinez-Lage JL, Sanchez H, Garcia JS, Lorenzo F. A pseudotumour of the mandible in a haemophiliac patient. A case report. *J Maxillofac Surg* 1983; **11**: 171–3. doi: [http://dx.doi.org/10.1016/S0301-0503\(83\)80041-1](http://dx.doi.org/10.1016/S0301-0503(83)80041-1)
- Brook AH, Bedi R, Lui WY, Yuen PM. Haemophilic pseudotumours of the mandible: report of a case in a one-year-old child. *Br J Oral Maxillofac Surg* 1985; **23**: 47–52. doi: [http://dx.doi.org/10.1016/0266-4356\(85\)90078-6](http://dx.doi.org/10.1016/0266-4356(85)90078-6)
- Bryan GW, Leibold DG, Triplett RG. Hemophilic pseudotumor of the mandible. Report of a case. *Oral Surg Oral Med Oral Pathol* 1990; **69**: 550–3. doi: [http://dx.doi.org/10.1016/0030-4220\(90\)90232-H](http://dx.doi.org/10.1016/0030-4220(90)90232-H)
- de Sousa SO, de Piratininga J, Pinto Junior DS, de Araujo N. Hemophilic pseudotumor of the jaws: report of two cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1995; **79**: 216–19. doi: [http://dx.doi.org/10.1016/S1079-2104\(05\)80284-7](http://dx.doi.org/10.1016/S1079-2104(05)80284-7)
- Zheng K, Zheng P. Hemophilic pseudotumor involving maxilla and tibia. *Chin Med J (Engl)* 1997; **110**: 233–5.
- Kale HA, Rathod KR, Prasad SR, Madiwale CM, Sheth RJ. Mandibular haemophilic pseudotumour containing a fluid-fluid level. *Br J Radiol* 2001; **74**: 186–8. doi: <http://dx.doi.org/10.1259/bjr.74.878.740186>
- Stevenson DS, Keast AT. An unusual cause of epistaxis: a haemophilic pseudotumour in a non-haemophiliac, arising in a paranasal sinus. *J Laryngol Otol* 2002; **116**: 294–5. doi: <http://dx.doi.org/10.1258/0022215021910573>
- Steele NP, Myssiorek D, Zahtz GD, Diamond A. Pediatric hemophilic pseudotumor of the paranasal sinus. *Laryngoscope* 2004; **114**: 1761–3. doi: <http://dx.doi.org/10.1097/00005537-200410000-00016>
- Rey EA, Puia S, Bianco RP, Pinto MT. Haemophilic pseudotumour of the mandible: report of three cases. *Int J Oral Maxillofac Surg* 2007; **36**: 552–5. doi: <http://dx.doi.org/10.1016/j.ijom.2006.10.002>
- Lima GS, Robaina TF, de Queiroz Chaves Lourenço S, Dias EP. Maxillary hemophilic pseudotumor in a patient with mild hemophilia A. *J Pediatr Hematol Oncol* 2008; **30**: 605–7. doi: <http://dx.doi.org/10.1097/MPH.0b013e3181758926>
- Lv M, Fan X, Su L, Zheng J. Ethanol embolization of hemophilic pseudotumor of the mandible. *Cardiovasc Intervent Radiol* 2011; **34**: 880–2. doi: <http://dx.doi.org/10.1007/s00270-011-0098-x>
- Xue F, Sun C, Sui T, Zhang L, Jiang L, Yang R. Hemophilic pseudotumor in Chinese patients: a retrospective single-centered analysis of 14 cases. *Clin Appl Thromb Hemost* 2011; **17**: 279–82. doi: <http://dx.doi.org/10.1177/1076029610366433>
- Thronsdon RR, Baker D, Kennedy P, McDaniel K. Pseudotumor of hemophilia in the mandible of a patient with hemophilia A. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2012; **113**: 229–33. doi: <http://dx.doi.org/10.1016/j.tripleo.2011.07.040>
- Lewis TR, Webb HR, Bell JP, Beall DP, Fish JR. Hemophilic pseudotumor. *J Okla State Med Assoc* 2005; **98**: 485–7.
- Gang TI, Huh KH, Yi WJ, Lee SS, Heo MS, Choi SC. The effect of radiographic imaging modalities and the observer's experience on postoperative maxillary cyst assessment. *Imaging Sci Dent* 2014; **44**: 301–5. doi: <http://dx.doi.org/10.5624/isd.2014.44.4.301>
- Lee JH, Huh KH, Yi WJ, Heo MS, Lee SS, Choi SC. Bilateral postoperative maxillary cysts after orthognathic surgery: a case report. *Imaging Sci Dent* 2014; **44**: 321–4. doi: <http://dx.doi.org/10.5624/isd.2014.44.4.321>