A case report of a 15-month history of a facial swelling

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INTRODUCTION

A 15-month history of facial and neck swelling is presented and dilemmas of diagnosis and treatment are mentioned together with a brief discussion of the condition. Involvement of many dental and medical specialties is also documented.

CASE REPORT

January 2012

Early in January 2012, a young lady, just one week short of her 16th birthday, was seen in the maxillofacial unit at Westmorland General Hospital. She arrived in an ambulance attended by an orthodontic practitioner, who was concerned about bleeding from the buccal mucosa in the lower-right quadrant. The young lady in question had recently returned from a holiday in America, where she had developed a sore throat, some lumps in her neck on both sides, and a swelling of the right cheek and right mandibular buccal mucosa.

As she was currently undergoing treatment from an orthodontist, she visited the practitioner on her return to Britain, who noticed the fluctuant swelling, took an X-ray of the lower-right first molar and second premolar teeth and noticed that there was a slight widening of the lamina dura on these X-rays around the second premolar tooth. This particular tooth was not responsive to ethyl chloride spray and was very slightly mobile. A diagnosis of periapical infection was made, for which amoxicillin and metronidazole had already been prescribed by her own dentist, but in order to obtain drainage the orthodontist incised the area. This resulted in a very profuse amount of buccal bleeding, which continued for some while, and hence the journey into the maxillofacial unit.

A full medical history was taken, which revealed no history of any note at all, no previous bleeding problems (indeed the patient had had four teeth extracted some two years previously for orthodontic reasons with no abnormal haemorrhage). The patient took no medicines routinely, did not smoke and had never had synthetic hormones or ever been pregnant. She was normally fit and healthy other than the recent development of a sore throat.

On examination, it was noted that there was a 1 cm incision site in the buccal sulcus apical to the first molar and premolar teeth and it appeared that there was arterial bleeding from the right mental artery. Under local anaesthetic, the bleeding was stopped by bipolar diathermy overlaid with Surgicel® and enclosed with buccal sutures. The bleeding stopped around two hours after the original incision had been made.

Routine haematological investigations did not reveal any abnormality.

The patient was reviewed again one week later. At that appointment, it was determined that she had been positive for

infectious mononucleosis. The right cheek was still swollen but was decreased in size compared with the week before. She still had a sore throat and also a painful lump on the back of the head on the right side. The clinician was informed that the patient had had a rash for the last three days after being prescribed amoxicillin for the tonsillitis. The intra-oral wound had healed well and there was no paraesthesia of the right mental area. Lymph nodes were still palpable on the right and left sides of the neck but less than 1 cm in size and there were also right posterior auricular nodes palpable. A diagnosis of infectious mononucleosis was made: she was advised to keep up her hydration and to use paracetamol for pain. A further appointment was given for eight weeks' time to check that the right cheek swelling had subsided.

February 2012

Four weeks later, the swelling of her right cheek had returned, and also she reported that she had discomfort from her lower-right first molar tooth and intermittent paraesthesia of her right mental area. On examination, it was noted (by a different clinician) that there was a little bit of swelling of the right cheek evident. A review appointment was made for one month later.

March 2012

At this appointment, in mid-March, the patient was noted to have some swelling of the right cheek and palpable bilateral lymph nodes of her neck. She complained of an intermittent sore throat. It was suspected that the swelling in the cheek was a resolving haematoma and consequently an ultrasound was arranged, further blood tests were ordered and a magnetic resonance imaging (MRI) scan to check for possible reactive lymph nodes was arranged. A review appointment was given for two months later.

May 2012

The next visit, on 1 May 2012, was to a third clinician. The swelling in the right cheek had increased in size and spread to the midline of the mandible. It was noted that her dental practitioner had started her on some metronidazole for this and also because there was an overlying flap of gingival tissue (an operculum) over the distal part of the lower-right second molar tooth. The intra-oral swelling extended from the first molar tooth in the lower-right quadrant to the lateral incisor tooth. There was no suspicion of infection or systemic upset. At this visit, a rotational tomogram (OPG) was taken (see figure 1) on which the clinician noticed an altered appearance of the mandible on the right side just extending over the midline towards the left. An ultrasound fine needle aspiration (FNA) of the swelling was arranged at the one-stop head-and-neck clinic the following day. The report of the MRI scan was available which suggested a possible vascular malformation haemangioma on the right side of the mandible.

The following day, the patient presented to the one-stop head-and-neck clinic with a fluctuant swelling of the right lower cheek of her face in the submandibular area, and there was also swelling of the buccal mucosa, an operculum was visible

Figure 1 OPG

overlying the distal part of the second molar tooth. The swelling felt fluid and pulsatile to palpation and the ultrasound showed a large right-sided vascular soft tissue mass associated with the right of her face. FNA was, consequently, not undertaken (see figures 2 and 3).

Figure 2 Photograph of the patient showing the submandibular and right-sided swelling

Under local anaesthetic, a small biopsy of this operculum was taken and not surprisingly the area bled quite profusely. This was stopped with Surgicel® and suturing, and the patient was prescribed tranexamic acid for oral use as a tablet being dissolved in water and held over the area before being

Figure 3 This shows swelling in the right buccal sulcus and an expansion of gingival tissue overlying the second molar tooth which was also ulcerated, due to the trauma of the upper second molar tooth onto this area. The patient was afebrile

Figure 4 This shows a reconstructed 3D picture. The report states that 'There is a large AV malformation based upon the right side of the mandible, this has recruited feeding vessels from the branches of the external carotid arteries bilaterally, drains into the anterior jugular veins bilaterally. The AV malformation is causing pressure related destruction of the adjacent mandible. There is no significant soft tissue component and no significant enlarged lymph nodes. The remainder of the neck appears within normal limits.'

swallowed so that both a local and systemic affect would occur. An urgent computerised tomography (CT) scan with contrast was performed (see figure 4).

The next day, the patient was referred to the cranio-facial unit at the Birmingham Children's Hospital.

The report of the biopsy confirmed oral squamous cell mucosa showing extensive ulceration and granulation tissue formation. The granulation tissue did show some endothelial atypia such as is commonly seen in pyogenic granulomas, but no evidence of malignancy. The diagnosis of the report was of granulation tissue/pyogenic granuloma.

The patient's mother contacted the maxillofacial unit at Royal Lancaster Infirmary (RLI) on 16 May 2012 to say that there was some oozing from the mucosa overlying the lower second molar on the right. Examination at that time showed that there was an increase in the swelling of her cheek and that the cheek was becoming more vascular and blue. The patient was very quickly reviewed in Birmingham, where a decision was made on angiography with a view to embolisation, but in the interim, she was sent back to Lancaster with a course of antibiotics.

On 21 May, the on-call maxillofacial surgeon was asked to see the patient as an emergency. When she arrived at A&E she was bleeding profusely from the mouth. Her Hb was found to be 9.4, and her BP was about 110/70.

She was taken straight up to theatre for an examination under local anaesthetic, with suction and anaesthetic facilities being available. There was an extremely brisk arterial bleed. Local anaesthetic was given; Surgicel® was inserted and sutured into position through very fragile tissue. Pressure

was also applied and the patient was kept occupied by the excellent theatre staff talking to her.

One of the possibilities that were considered at that time was extraction of the upper-right second molar tooth, which would ease the trauma and pressure to the overgrown gingivae of the lower jaw. This, indeed, had been a suggestion from Birmingham, but at this stage there had been no letter received. However, a close examination of the CT scan showed that this tooth was also likely involved in the arteriovenous malformation (AVM), and if this was done it may give rise to uncontrollable bleeding.

A second possibility was for a blow-down splint to be constructed over the lower teeth at the front, which would mean that the patient was just occluding on the splint overlying the front teeth and no trauma would be taking place at the back. The third possibility was to tie off the major vessels, but this would a) likely cause problems for Birmingham and b) there were also feeders from the left side.

The fourth possibility was an urgent transfer to Birmingham as soon as possible so that the bleeding would at least be stopped properly. Options one and three were discounted for the reasons stated.

At 8.30 the next morning, her blood pressure was 92/60, she was pale, but there was no further bleeding. The Queen Elizabeth Hospital (QEH) in Birmingham was contacted and later that morning the patient was transferred, with a maxillofacial surgeon in attendance, by blue-light ambulance to QEH.

The subsequent letter from QEH shows that, on 22 May 2012, she was taken to theatre under an intubated anaesthesia, and by a femoral approach, embolisation of the area with 7X15ml of Onyx®. The upper-right second molar tooth was also extracted, which gave rise to a massive bleed which was dealt with by vascular glue to the socket and suturing with haemorrhage control. She was then admitted to the intensive care unit (ITU), and she remained intubated for three days. She was discharged home on 27 May. An appointment was made in Birmingham for the second stage of embolisation (11 x 1.5ml of Onyx®).

June 2012

The patient was reviewed at RLI on 5 June, and it was noticed that the right cheek was still pulsatile but there had been no further bleeding. She was reviewed again a month later on 3 July, and the right side of her face had swollen, she had recently lost two plugs of glue from inside of her mouth but had had no further bleeding. Further appointments were made for review at Lancaster, as well as further plans for embolisation at Birmingham at the end of August.

August–November 2012

At the end of August 2012, further embolisation with Onyx® took place and in September the patient was once again reviewed at RLI. A comment was made that she was still swollen but comfortable. The patient was reviewed in Birmingham by a consultant plastic surgeon, a consultant interventional radiologist and a consultant maxillofacial surgeon in November 2012. It was stated that at that time there was an increased swelling of the lower half of the right side of the face with regular bleeds in the intra-oral site and that there was some pus discharging from the hyperplastic gingival tissues

which were thought to be a foreign body reaction to the Onyx®. The lower molar teeth on the right side were slightly mobile but were not tender. Following this, the patient was reviewed at RLI and we were informed that she was to have a course of bleomycin, with the intention of reducing the size of the lesion prior to any surgery.

March 2013

By March 2013, there was clinical suspicion of an increasing lesion. Clinical examination on that day shows the vascular malformation to have increased in size and there was a 2 x 2cm hard mass in the swelling which was painful to the touch. All three distal right molar teeth had increased in mobility and the soft buccal mass in the sulcus had extended forward and was quite friable.

Further ultrasound and CT angiogram was undertaken at RLI on 21 March. The comments on this CT scan and in comparison with the one taken in May 2012 stated that: 'There is considerable artefact of the injected sclerosant labelled agent. However, the main feeding vessels on the right to the AVM are still present, there is increased swelling over the right side of the face and an increased recruitment of vessels around the right angle of mandible and the main feeding vessel from the left is still patent.'

The patient has recently been transferred as an emergency to Birmingham, where she was immediately intubated and spent the next few days on ITU. The cause of the swelling was an abscess due to the Onyx® material used for the sclerosis.

DISCUSSION

The brief points of this case highlight quite a few problems in initially obtaining a diagnosis. The patient had a sore throat and was tested for glandular fever. There was a swelling in the soft tissue buccal to the apical area of a tooth and the radiograph showed what most people would think to be possible signs of infection, hence treatment by incision and antibiotics was not unreasonably started. Following that, matters settled down somewhat and the young lady was kept under review. The side comment is on the report of the radiograph taken on 1 May 2012, which is reported as 'normal' by radiology, but which a clinician in the maxillofacial department noted to be abnormal.

AVM is a fast flow anomaly (because it involves abnormal arteries) composed of abnormal connections between arteries and veins. AVM's cause pain, skin breakdown, overgrowth of tissue, and even congestive heart failure. AVM's can expand, often requiring embolisation, in which an inert material is injected into the vessels to block them, followed by surgical treatment. Some years ago, surgical treatment was the first choice, but this seems to have led to many more problems and now generally it is agreed that sclerosis is ideal.

In this case, care has had to be taken with embolisation to prevent the blocking agent getting into areas such as the main or ophthalmic arteries, where it is certainly not required and could cause obvious serious problems. Onyx® is a mixture of ethylene vinyl alcohol copolymer dissolved in dimethyl sulfoxide and micronised tantalum powder is suspended in liquid polymer so that it will show up on X-ray.

The case highlights many of the difficulties that are encountered in a general hospital maxillofacial department

that has to deal with a rare and very complex condition when the tertiary specialist centre is over 100 miles away. We are grateful to our colleagues in Birmingham and our local

radiology department for helping us to see the case through to a stable and satisfactory situation.

Maxillofacial laboratory manager gives keynote lecture at Polish Orthodontic Congress

Paul Mallett, who is the maxillofacial laboratory manager based at Royal Lancaster Infirmary, was invited to give a series of lectures throughout a three-day orthodontic congress held in Olsztyn, Poland (17-19 May, 2013). Paul's principal lecture focused on 'thermo-formable orthodontic appliances'. Multiple techniques were presented explaining manufacturing technique, materials, and their application in orthodontic practice. The majority of the techniques reflect work that is carried out within the maxillofacial laboratory.

Lukasz Grodzinski, who is vice chairman of the Polish Orthodontic Technicians Association (PTTO), invited Paul to present at their annual congress. Paul is also currently the chairman of the Orthodontic Technicians Association (UK). His lecture was very well received. Paul doesn't speak many words in the Polish language; however, the PTTO's vice chairman also served as his personal translator and guide. This involved simultaneous translation of lectures so that the non-English speaking delegates understood the lectures.

Paul and Lukasz review the lectures, which contained 198 Powerpoint slide images

Paul, first left, accepts his speaker's gift from PTTO president, chair and vice chair

Speaking at the Association's annual general meeting, he also developed a closer relationship between European groups, with a proposed reciprocal relationship where Polish delegates would be invited to attend UK orthodontic conferences. He also went on to encourage the Polish delegates to attend the forthcoming International Orthodontic Congress, to be held in London in 2015.

Paul was so impressed by the Polish Association that he now has membership of the PTTO and intends to attend future congresses in Poland. At the end of his lectures, Paul was presented with a book about Poland, signed by all of the PTTO committee members.