

Anesthetic management of a patient with Kimura's disease for superficial parotidectomy

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Abstract

Kimura's disease is a rare form of chronic eosinophilic inflammatory disease with vascular proliferation involving salivary gland, skin, lymph node, and kidney. Important anesthetic concerns include increased surgical bleeding due to its vascular nature, airway involvement by the mass leading to a possible difficult airway, allergic symptoms associated with high eosinophil count and elevated IgE level and nephrotic syndrome due to involvement of kidney by the inflammatory process. There is paucity of information in the literature on the anesthetic management of Kimura's disease. We describe the anesthesia technique and review the literature of such a case posted for superficial parotidectomy.

Key words: Anesthesia, Kimura's disease, parotidectomy

Introduction

Kimura's disease (KD) is a chronic inflammatory disease of unknown etiology affecting young males, most commonly Asian.^[1] The disease was first described in China by Kim and Szeto^[2] in 1937, but it was given its name and became more widely known after the systematic description by Kimura^[3] in 1948 in Japan. The disease is endemic in China and Japan, although sporadic cases have been reported elsewhere.^[1] Reports on anesthetic management in patients with KD are however rare.^[4] We report a patient with parotid enlargement for superficial parotidectomy, who was pre-operatively diagnosed to have KD. We have reviewed the literature and discussed the anesthetic management of the disease.

Case Report

A 22-year-old male patient presented with gradually progressive swelling over the right parotid region for last 7 years. He had

multiple bilateral enlarged cervical lymph nodes. History and physical examination did not reveal any other illness except mild skin eczema in legs. Airway examination revealed mouth opening 4 cm, modified Mallampatti class II with normal thyromental distance and neck movements. The swelling was 6×5 cm in size, nontender, nonfluctuant, firm in consistency, palpable in the left anterior triangle of the neck, extending from the angle of the jaw to the left preauricular area. No warmth or redness was noted on the overlying skin. Histopathology of the mass revealed lymphoid follicles formation with prominent germinal centers, infiltration of eosinophils, and increased postcapillary venules and vascular proliferation suggestive of KD. Cervical lymph node biopsy, done to rule out malignancy, also revealed similar finding.

On further investigation, hemoglobin was 12.8 mg/dl, total leucocyte count 8100 with peripheral eosinophilia (eosinophil 18%, neutrophil 52%, basophil 1%, lymphocyte 25% and monocyte 4%), platelet count 2,43,000 and erythrocyte sedimentation rate was 18 mm fall in first hour. Liver function test showed bilirubin 0.8 mg/dl, total protein 6.9 g/dl, albumin 4.2 g/dl, and normal liver enzymes. Renal function test revealed elevated blood urea (60 mg/dl) and serum creatinine (1.1 mg/dl). Proteinuria 2+ was present in the urine dip-stick test. Serum electrolytes were Na 137 mEq/l and K 4.2 mEq/l. Serum IgE level was markedly elevated (559 IU/ml; normal 0-14 IU/ml). Ultrasound of the abdomen showed that both the kidneys were of normal size and echo-texture. The chest X-ray was normal.

The patient was administered oral alprazolam 0.25 mg and ranitidine 150 mg the night before and on the morning

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of surgery. In the operating room, intravenous access was obtained and anesthesia was induced with fentanyl 120 μg and propofol 60 mg intravenous. Oro-tracheal intubation was facilitated with atracurium 30 mg intravenous and mechanical ventilation was initiated. Monitoring included pulse oximetry, continuous electrocardiography, noninvasive blood pressure, end-tidal CO_2 , temperature and neuro-muscular blockade depth by train-of-four (TOF). Anesthesia was maintained with oxygen nitrous oxide, and isoflurane (end-tidal concentration 0.6-0.8%). Analgesia was provided with fentanyl infusion 50-70 $\mu\text{g}/\text{h}$ and intermittent boluses of atracurium maintain a TOF count of 1 to 2.

The patient underwent a selective neck dissection to excise the right buccal space mass, including right superficial parotidectomy, and excision of multiple lymph nodes involving the peri-parotid tissues. The facial nerve was preserved by surgeons without resorting to facial nerve stimulation, as the nerve was quite thick and easily identified. Duration of surgery was 4 h. The glands were quite vascular; intra-operative blood loss was 750 ml which was adequately replaced by crystalloids. The patient made 500 ml urine.

Intra-operatively, heart rate was 70-85/min, blood pressure 94/60-128/80 mmHg, oxygen saturation 98-100% and EtCO_2 35-38 mmHg. At the end of surgery, fentanyl infusion was stopped, neuro-muscular blockade was reversed and trachea extubated. He was awake and comfortable in the post-anesthesia care unit. Intravenous tramadol 50 mg 8 hourly and intravenous paracetamol 1 gm 8 hourly were prescribed for post-operative analgesia. The post-operative course was uneventful and patient was discharged home after 6 days.

Discussion

KD is a rare disease characterized by eosinophilic lymphoid infiltration with secondary vascular proliferation which usually involves subcutaneous tissues, lymph nodes (periauricular, cervical, axillary, and inguinal), parotid and submandibular salivary glands, and rarely, oral mucosa. Other unusual sites of involvement include the auricle, scalp, and orbit.^[3] KD can easily be mistaken for a malignant disorder. Fine needle aspiration cytology can be misleading and diagnosis is established only on histopathological examination.

Renal involvement can occur in 60% patients in the form of membranous glomerulonephritis, minimal change glomerulonephritis, diffuse proliferative glomerulonephritis, mesangial proliferative glomerulonephritis, and also nephrotic syndrome (12% of cases).^[5,6]

Significant elevation of serum IgE and peripheral eosinophilia are not uncommon;^[5] patients may be mistreated as tropical eosinophilia.^[1] The differential diagnoses of KD include eosinophilic granuloma, Mikulicz's disease, acute lymphocytic leukemia, Hodgkins disease, angioimmunoblastic lymphadenopathy, and angiolymphoid hyperplasia with eosinophilia (ALHE). These usually can be differentiated on clinical and histological bases.^[7]

The mechanisms of these manifestations continue to be the subject of controversy and research. Several mechanisms have been postulated, which include atopy to persistent fungal or parasitic antigenic stimulation^[5] and alterations of immune regulation,^[8] but none have gained widespread acceptance to date. The onset of KD is insidious and lesions are benign, which follow an indolent course, gradually increasing in size over months or years. The overall prognosis is good. Although spontaneous involution is rare, malignant transformation has not been documented.^[9]

Radiological features, particularly on computed tomography (CT) and magnetic resonance imaging, have been reported to be useful in the diagnosis of KD. Parotid gland and lymph nodes show intense enhancement on CT reflecting the vascular nature of the lesions. The borders are ill-defined and there is usually adjacent enhancing cervical lymphadenopathy.^[10]

The optimal treatment for KD is not well established. However, treatment should aim at cosmesis and function preservation while preventing recurrences and long-term sequelae. The treatment options include conservative medical treatment, radiotherapy, cryotherapy, laser fulguration, and surgical excision. Medical management involves oral or intra-lesion steroids, cyclosporine, oral pentoxifylline, all-trans-retinoic acid; all with limited success.^[11] At initial presentation, surgical excision is the choice for both diagnosis and therapy. However, lesions often recur after excision. If recurrence is frequent or there is symptomatic nephrotic syndrome, systemic steroids should be started. Unfortunately, there is a tendency for lesions to recur when steroid therapy is stopped.^[12] For recalcitrant cases or lesions not amenable to surgery due to size or unacceptable resultant morbidity, radiotherapy can be considered.^[13] Low-dose local irradiation (about 25 to 30 Gy) has been reported to yield good control and obviates need for long-term corticosteroids.^[12,13]

Anesthesiologist may encounter a patient of KD for biopsy of lymph node or kidney, excision of subcutaneous mass or salivary gland.^[3] Anesthetic concerns are increased chance of bleeding due to vascular nature of the mass; airway involvement by the mass; presence of concurrent nephrotic syndrome and any allergic symptoms. Airway compromise may occur due

to compression by enlarged subcutaneous mass, cervical or mediastinal lymph nodes. A mass involving the hard palate has been reported.^[14] Our patient did not have any mass involving the airway.

Our patient did not have any clinical feature suggestive of renal insufficiency but routine urine dip-stick test revealed 2+ proteinuria and blood biochemistry showed elevated renal parameters. Subsequent work-up revealed marginally elevated 24-h urinary albumin with preserved serum albumin but ruled out any ultra-sonographic features of chronic kidney disease. A renal biopsy was not advised before surgery as it would not have affected subsequent anesthetic or surgical management.

Despite a markedly elevated eosinophil count and IgE, the patient did not have any allergic symptom except mild skin eczema over the left lower limb for which he occasionally used topical steroid as advised by dermatologist. Before surgery, the lesions were not active and the patient was not using steroids. Perioperative steroid supplementation was not considered. There was no intraoperative hypotension.

Balanced anesthesia was used with the aim to maintain hemodynamic stability and provide adequate analgesia. Urine output was monitored to ensure adequate renal perfusion for two reasons; there was an increased risk of blood loss as the USG and histo-pathologic features were suggestive of highly vascular mass and as the patient had compromised renal function.

In conclusion, the present case highlights the need for awareness of KD by clinicians and pathologists to avoid unnecessary and potentially harmful investigations and anesthesiologists to be avoid massive blood loss.

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