Case Report

Post-operative acute circulatory collapse following craniofacial surgery: Rare event and rarer cause

Department of Oral and Maxillofacial Surgery, Maulana Azad Institute of Dental Sciences, [1]Departments of Anesthesiology and [2]Neurosurgery, Govind Ballabh Pant Hospital, New Delhi, India

Sujata Mohanty, Jitender Dabas, Monica S. Tandon[1], Daljit Singh[2], Ujjwal Gulati

ABSTRACT

We are presenting a case of a 13-year-old female patient diagnosed and operated for maxillary odontogenic myxoma extending to the anterior cranial base. The postoperative complication occurred in the form of acute circulatory collapse. The patient was bailed out with cardiopulmonary resuscitation and return of spontaneous circulation occurred. Investigations into the cause of the event led to the finding of an uncommon syndrome. Meigs syndrome is a triad of ovarian tumor mass, pleural effusion and ascites. It has been the topic of interest for the gynecologic fraternity since 1934, when the first case was reported by J. V. Meigs. According to the best of our knowledge, this is the first case report of an acute circulatory collapse due to Meigs syndrome in a maxillofacial patient.

Address for correspondence:
Dr. Sujata Mohanty,
Department of Oral and Maxillofacial Surgery,
Maulana Azad Institute of Dental Sciences, BSZ Marg,
New Delhi - 110 002, India.
E-mail: drsujatam@gmail.com

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Introduction

Surgery for pathologies encroaching the cranial base are associated with an array of operative and postoperative complications. Most of the acute and worrisome complications are either neurological or damage to adjacent vital structures. Acute hypotension in the postoperative period is a rare event in the absence of significant blood loss during surgery. However, in rare situations, secondary hemorrhage, visible or concealed, may account for massive loss of circulatory volume. We are presenting a unique case of acute circulatory collapse in the postoperative intensive care of a young female who underwent resection of odontogenic myxofibroma encroaching the anterior cranial base.

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Case Report

A 13-year-old female patient reported to our institution with a swelling on the right side of the upper jaw since 2 years. Contrast-enhanced computerized tomography (CECT) revealed a well-defined, nonenhancing, soft tissue density, expansile mass measuring approximately 5.6 × 4.6 × 4.1 cm [Figure 1]. The mass was seen to involve the orbital floor, medial wall of the orbit, ethmoidal sinuses, inferior orbital fissure and root of the right pterygoid process. Incisional biopsy revealed the lesion to be odontogenic myxofibroma. Magnetic resonance imaging revealed a T2 hyperintense mass with nonhomogenous enhancement. The tumor mass was excised and subtotal hemimaxillectomy was performed using the modified Weber Fergusson approach. Apt hemostasis and closure were performed. Total blood loss during the surgery was approximately 300 mL. The postoperative blood investigations revealed hemoglobin and other blood counts within normal limits. But, about 4–5 h after the surgery, there was an episode of severe hypotension. The blood pressure fell down to be undetectable on the monitor and the carotid pulse could not be felt. The patient was managed

effectively with cardiopulmonary resuscitation (CPR) and vasopressors. On the third postoperative day, oxygen saturation fell down due to respiratory distress and ascites developed. Ultrasound (USG) of the abdomen revealed a tumor mass of size 5.2 cm × 5.1 cm × 5.5 cm in the right ovary. The chest X-ray revealed left-sided pleural effusion [Figure 2]. CECT of the abdomen showed a cystic lesion in the right ovary and fluid in the peritoneum [Figure 3]. CECT of the chest was performed, which revealed left-sided pleural effusion along with collapsed lung [Figure 4]. In due course of time, the patient developed pericardial effusion, which was managed by peri-cardiocentesis and insertion of a pigtail catheter. The pleural, pericardial and peritoneal fluids were found to be transudate in nature. The blood sample revealed increased levels of tumor marker CA 125 (241 U/mL). All these findings established the diagnosis of Meigs syndrome. The patient was managed symptomatically and discharged after full recovery. The patient was then kept on regular follow-up. Surprisingly, at the 6-month follow-up visit, USG revealed no ovarian pathology. Currently, the patient has been on follow-up for 20 months and no recurrence has been noticed yet. CA 125 levels have also dropped down to normal (16 U/mL).

Discussion

Acute circulatory collapse in a patient can have numerous causes. But, the main causes of its occurrence after a surgery include an inadequately treated hypovolemia after excessive blood loss, any preexisting cardiac/peripheral vascular disease, use of hypotensive drugs during/after the surgery, pulmonary or coronary embolism or some unknown systemic abnormality. But, in our patient, there was no active bleed either intraorally or on the face. The oropharynx was negative for any concealed hemorrhage. The patient was normotensive throughout the surgery along with normal oxygen saturation and all vital parameters within normal range. Chest radiograph and

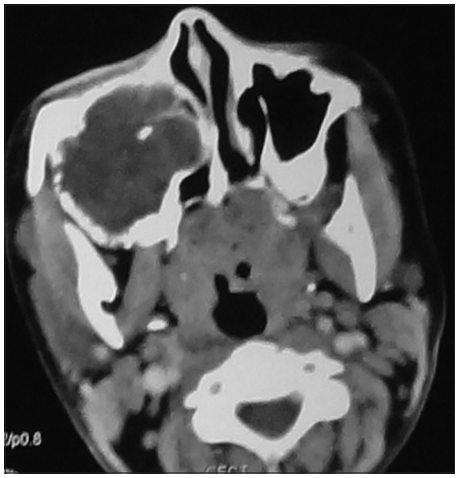

Figure 1: Contrast-enhanced computed tomography showing an expansile mass in the right maxilla measuring approximately 5.6 cm × 4.6 cm × 4.1 cm

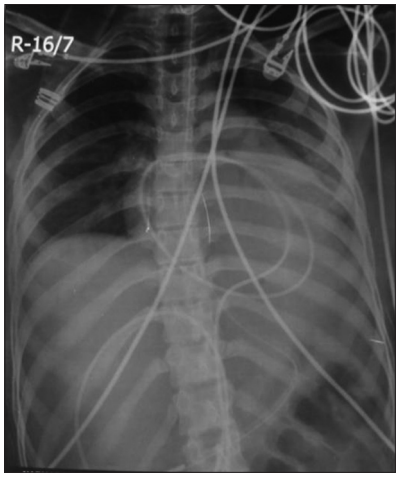

Figure 2: Chest radiograph showing left-sided pleural effusion

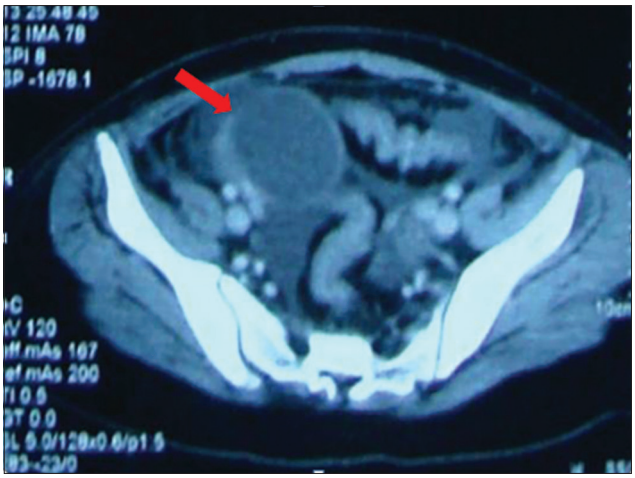

Figure 3: Contrast-enhanced computed tomography of the abdomen showing cystic lesion in the right ovary and ascitic fluid

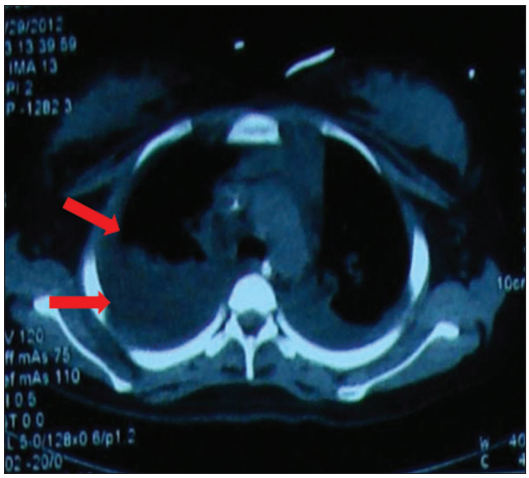

Figure 4: Contrast-enhanced computed tomography of the chest showing left-sided pleural effusion along with lung collapse

USG suggested pleural and pericardial effusion. CECT of the abdomen revealed a right ovarian cystic lesion along with ascites. The presence of ovarian cyst along with ascites and pleural effusion redirected the thought process toward Meigs syndrome. Further blood work-up for CA 125 reassured the diagnosis. Once a holistic view of the patient with a complex triad of ovarian cyst, ascites, pericardial and pleural effusion was made, diagnosis and management was streamlined.

Meigs syndrome is a condition characterized by ovarian tumor, pleural effusion and ascites, commonly seen in the early menopausal life, but can occur at any age. It was first described by Meigs in 1934 in a series of three cases.[1] It was designated as "Meigs syndrome" by Rhodes and Terrel in 1937.[2] Ovarian tumor can be a fibroma, myxofibroma, fibromyxoma, thecoma or a granulose cell tumor. Other than these solid tumors, benign cysts of the ovary, leiomyomas and teratomas may also be associated with pleural effusion and ascites, a condition called as "Pseudo Meigs syndrome."[3] Abdominal hypertension refers to the raised intraabdominal pressure because of the accumulation of fluids that can cause the visceral organs to function improperly, leading to decreased cardiac and urinary output, and, in extreme instances, cardiovascular collapse. Cases with abdominal hypertension in Meigs' syndrome have well been documented in the literature.[4] In the present case, the intraoperative blood loss was within the limits of Maximum Allowable Blood Loss (MABL); hence, the authors ruled out the possibility of hypovolaemia causing this episode of circulatory collapse. The abdominal and chest findings directed the authors' attention (after literature research) toward the possibility of this rare syndrome. To confirm the diagnosis, tumor marker levels were assessed in this patient as raised CA 125 level is another significant finding in this syndrome. Normal CA 125 level in the serum is about 35 IU/mL, but in Meigs syndrome it may be elevated significantly.[5] The CA 125 level in our patient was found to be 241 IU/mL. Thus, the triad of ovarian cyst, ascites and pleural effusion, along with raised level of CA 125, confirmed the diagnosis. It has been seen that levels of CA 125 have a direct correlation with the volume of ascitic fluid.[6] The level of CA 125 falls down as the ascites resolves. Ascites usually resolves after surgical removal of the ovarian pathology, but in extremely rare cases, resolution of symptoms may occur without any surgical intervention. Pascal termed this condition as "Regressive Meigs syndrome."[7] He found that acute torsion caused necrosis of the adnexae, which resolved the effusion. This might have been the reason for resolution of symptoms in the present case without any surgical intervention. However, we would refrain from commenting on the same due to lack of sequential abdominal CECT. Another possible explanation for the resolution of symptoms in this case can be self-regression of the ovarian cyst, as has been found in the study by Sasaki *et al*., where they concluded that 73% of simple ovarian cysts usually regress on their own and 48% of them regress within the first 6 months of their diagnosis.[8] This hypothesis is supported by the absence of ovarian cyst on 6-month follow-up USG. However, the exact reason for this bizarre behavior of the ovarian cyst is still unknown. There is also a probability that the Meigs' syndrome was a coincidental finding unrelated to hypotension. But, in the absence of any preexisting peripheral or central cardiovascular disease and blood loss during surgery, which was well within the limits, this possibility seems unlikely. The patient is still on gynecology follow-up and no recurrence of the symptoms have been noted yet.

Conclusion

Being a maxillofacial surgical unit, it was a unique experience for the authors and the reason for this case report is to attract the clinician's attention toward the ovarian pathologies. We would like to conclude that if the medical history given by the patient reveals any symptomatic/asymptomatic ovarian pathology, or if the pertaining symptoms develop during any stage of treatment, then the clinician should always keep these possible consequences in mind and should take appropriate measures.

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