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Abstract: Gestational gigantomastia (GG) or gravidic macromastia is a rare, severely debilitating condition characterized by massive enlargement of breasts and resulting in tissue necrosis, ulceration, infection, and, occasionally, haemorrhage. We herein report the anaesthetic management of a parturient presenting with GG, scheduled for caesarean section under single shot spinal anaesthesia. Although there is no reference in the literature regarding the anaesthetic considerations in gigantomastia, several implications must be considered, including the reduced chest wall compliance and reduced lung volumes, the increase in work of breathing, minute ventilation, and oxygen consumption. The increase in intrathoracic pressure caused by higher inspiratory pressure can impair ventricular filling and cardiac output. Moreover, breast enlargement predisposes the obstetric patient to a difficult airway during laryngoscopy. Our patient was a full term 25 yrs old female, second gravida, para 1, height 151cm, with history of previous caesarean section, planned for caesarean section. Intraoperative period was uneventful with minimal changes in hemodynamics. Patient was monitored postoperatively in the HDU and had an uneventful course.

Key words: gestational gigantomastia, caesarean section, single shot spinal anaesthesia

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I. Background

Gestational gigantomastia (GG) or gravidic macromastia is defined as a disorder characterized by a diffuse, extreme and incapacitating enlargement of one or both breast during pregnancy.¹ It has an incidence ranging from 1 in 28,000 to 1 in 100,000 pregnancies worldwide.² The first case of GG was described by Palmuth in 1648.³ Apart from being a social and emotional disability, it can lead to a myriad of physical symptoms as well, which include breast pain, infection, ulceration, postural problems and back pain. It can even lead to chronic traction and thereby causing temporary or permanent damage to fourth, fifth, or sixth intercostals nerves presenting in the form of loss of nipple sensation, further promoting infection and ulceration.² Age of the patients vary from 16 to 35 years, with majority of the cases reported from 26 to 30 years of age.¹ We herein report the anaesthetic management of a parturient having Gestational Gigantomastia scheduled for caesarean section.

II. Case Report

A full term 25yr old female, gravida 2, para 1, with a previous history of caesarean section 3 years back, was brought for caesarean section. She gave history of gradual progressive enlargement of bilateral breasts by the beginning of second trimester of the present pregnancy. She underwent various investigations, including imaging, pathology and biochemical. Ultrasonography of both breasts with axillae during the early part of second trimester suggested fibroadenoma (BIRADS category III- probably benign finding). Aspiration cytology done around the same time gave an impression of fibroadenoma. True cut biopsy done from bilateral breasts during second trimester suggested histopathological features of pregnancy related changes which led to the diagnosis of Gestational Gigantomastia being considered. MRI done in early third trimester revealed bilaterally enlarged breast with heterogenous signal intensities and ductal ectasia, with no sizeable adenopathy or obvious focal mass being noted. She also had her haemoglobin variant analysis, prolactin and ANA levels investigated, results of which were insignificant. The preoperative investigations revealed haemoglobin 8.4g/dl, total leucocyte count 5.18 thousand/mm³, platelet 2.66 lakhs/mm³, random blood sugar 76mg%, prothrombin time 12.6s, INR 1.03. The liver and kidney function tests were within normal limits. Serial abdominal ultrasounds in

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the 1st, 2nd and 3rd trimesters showed normal fetal development and the last ultrasound showed single live intrauterine fetus in cephalic presentation.

By the beginning of the third trimester, due to excessive enlargement of the breasts, she was mainly confined to bed and could get up and walk around only with the help of support (fig 1). There were multiple necrotic areas over bilateral nipple area for which she was undergoing regular dressing.

On physical examination, her pulse rate was 90/min with blood pressure of 100/70mmHg (right upper limb) and 104/70mmHg (left upper limb). The respiratory, cardiac and neurological examination showed normal findings. Her airway was graded as Mallampati class II and she was accepted for surgery in ASA Grade III. Engorged neck veins were noted over the entire chest wall, extending up to the neck. Bilateral breasts were grossly enlarged (approximating about 45cm x 35cm), the lower margins extending to the level of the 11th-12th ribs along with bilateral axillary lymphadenopathy and enlargement of right axillary tail (15cm x 10cm).

On the day of surgery, intravenous (IV) access with 18G IV cannulae were secured on the dorsum of both hands. Premedication was done with injection ranitidine 50mg IV and injection metoclopramide 10mg IV. Patient was preloaded with 500ml of ringer lactate and shifted to operation theatre (OT). One unit of PRBC was kept ready. Extra theatre personnel were on standby to help with the shifting and positioning of the patient inside the operating theatre. The operating table was prepared with wide arm boards placed parallel to the table. After shifting to OT, BP cuff, ECG electrodes and pulse oximeter were connected and baseline hemodynamic parameters recorded. Baseline parameters were BP 100/68 mmHg, PR 94/min and SpO2 98% on room air respectively. Two dhotis (long cotton loincloth) were joined together and applied over the chest area in a figure of 8, so as to provide support for the enlarged breasts (fig 2). The ends of these two dhotis were attached to two heavy IV stands so as to lift and support the breasts in the same plane as that of the operating table. Patient was continuously kept in verbal contact enquiring about the position where she was comfortable, prior to proceeding for spinal anaesthesia. Then under aseptic conditions, a 25G quincke needle was introduced in the L3-L4 interspace and subarachnoid block was performed (single shot spinal anaesthesia) with bupivacaine heavy 0.5% 8mg and fentanyl 25µg, in the sitting position (fig 3). The patient was then placed supine and oxygen 4L/min was administered via facemask. A head ring was placed under the patient’s head and 15 degree left lateral tilt was maintained by applying wedge under right hip. After assessing sensory level of T6, the operating table was positioned in semi-Fowler’s position, to improve respiratory comfort of the patient. There was minimal fall in blood pressure for which use of vasopressor was not required. Following delivery of a single live baby, injection oxytocin 10units IM (intramuscular) was given followed by infusion (20units in 500ml crystalloid at a rate of 10-12 drops/min). Surgery lasted for approximately 50 minutes and was uneventful. Intraoperatively, the variation in blood pressure was maintained within 20% of the baseline BP. After the end of surgery, patient was monitored for 30 minutes in post anaesthetic care unit (PACU) and later shifted to HDU (high dependency unit) for further monitoring. Bolsters were kept on the sides of the cot to support the breasts from sagging laterally and the head end was raised by about 30 degree to prevent respiratory discomfort.

Postoperative pain was managed with injection paracetamol 1gm IV 8hrly and injection tramadol 100mg IM 12hrly. After observation in the HDU for 24hrs, patient was later on shifted to post natal ward.

**III. Discussion**

Gestational gigantomastia (GG) or gravid macromastia, is a rare condition that presents as enlargement of one or both breasts during pregnancy. The first case of GG was described by Palmuth in 1648. It has an incidence ranging from 1 in 28,000 to 1 in 100,000 pregnancies worldwide. The definition of this rare disease is although not clear, Lewison et al.1960, used beautiful words to describe a typical case “True gigantomastia develops rapidly during pregnancy, undergoes regression after delivery, and recurs with subsequent pregnancies.” In 2011, Dafydd et al. defined gigantomastia as excess breast tissue that contributes >3% of a patient’s total body weight. Another more discrete definition used is an enlargement of breast, where more than 1500 g of breast tissue needs to be removed from the breast.

Risk factors for GG are not well understood, however occurrence is more common in Caucasian and multiparous women; however maternal age and fetal gender do not seem to have any association. Although GG can occur in any pregnancy, the occurrence of GG in one pregnancy is probably the strongest risk factor for its recurrence in subsequent pregnancies.

Many theories have been proposed about the etiology and pathogenesis of GG, including excessive production of estrogen or prolactin, hormone receptor sensitivity, and underlying autoimmune disease triggered by pregnancy. There are also reported cases of penicillamine induced breast gigantism with subsequent treatment with danazol.

Dancey et al. proposed a classification wherein gigantomastia can be divided into three subgroups according to aetiology (table 1). Group 1 is idiopathic in nature, quiescent and can be managed with a breast reduction in the first instance and tend to have a good prognosis. Group 2 is a result of endogenous hormone imbalance (pregnancy or juvenile) and presents with very aggressive and unremitting breast growth. They often
require multiple reductions and consideration should be given to a primary mastectomy with breast reconstruction. There is a familial tendency in juvenile gigantomastia and patients should therefore be made aware of the possible genetic implications. Group 3 is drug induced and responds well to cessation of therapy with or without breast reduction.

<table>
<thead>
<tr>
<th>Group</th>
<th>Characteristics</th>
</tr>
</thead>
<tbody>
<tr>
<td>1a</td>
<td>Idiopathic, spontaneous condition of excessive breast growth in a patient with a BMI &gt;30.</td>
</tr>
<tr>
<td>1b</td>
<td>Idiopathic, spontaneous condition of excessive breast growth in a patient with a BMI &lt; 30.</td>
</tr>
<tr>
<td>2a</td>
<td>Excessive breast growth related to an imbalance of endogenous hormone production occurring during puberty</td>
</tr>
<tr>
<td>2b</td>
<td>Excessive breast growth related to an imbalance of endogenous hormone production occurring during pregnancy</td>
</tr>
<tr>
<td>3</td>
<td>Excessive breast growth induced by a pharmacological agent.</td>
</tr>
</tbody>
</table>

Apart from being a social and emotional disability, GG can lead to a myriad of physical symptoms as well, which include breast pain, infection, ulceration, postural problems and back pain. It can even lead to chronic traction and thereby causing temporary or permanent damage to fourth, fifth, or sixth intercostals nerves presenting in the form of loss of nipple sensation, further promoting infection and ulceration. Lymphatic and venous stasis can further predispose to the development of ulcers. Severe sepsis, renal dysfunction, multiorgan dysfunction syndrome, and even death have been have been reported in rare patients with GG.

Although only rare cases have been reported in literature that underwent complete spontaneous resolution after pregnancy, majority of cases need either medical or surgical treatment. Though medical management is the first line of treatment, surgery is the mainstay.

Anaesthesiologists may encounter these patients either during obstetrical anesthesia or corrective plastic surgery procedures.

Airway problems are more common in pregnancy than in the general population due to anatomical and physiological changes during pregnancy. Some anatomic changes that may affect the obstetric airway include upper airway edema, breast enlargement and excessive weight gain. Although there is not much reference in the literature regarding the anaesthetic considerations in gigantomastia, several implications must be considered, including the reduced chest wall compliance and reduced lung volumes, the increase in the work of breathing, minute ventilation and oxygen consumption. These changes can lead to hypoxemia and rapid desaturation. The increase in intrathoracic pressure caused by higher inspiratory pressure can impair ventricular filling and cardiac output. Moreover, in the supine position, the enlarged breasts tend to fall back against the neck, which can interfere during laryngoscopy and intubation. During general anaesthesia, adequate preoxygenation, ramped head position, the use of a short-handled laryngoscope, and acid aspiration prophylaxis are mandatory.

Regional anaesthesia is a desirable technique for caesarean section. Single shot spinal anaesthesia (SA) is preferred for elective caesarean delivery, due to its simplicity, ability to provide adequate surgical anaesthesia, ease of administration, faster onset of action, and safety. SA also allows higher maternal satisfaction due to early skin-to-skin contact with the baby. The operating table should be appropriately sized. Wide arm boards placed parallel to the operating table and a semi-Fowler’s position to improve respiratory comfort for the patient are recommended.

A detailed preanaesthetic evaluation is undertaken to look for any significant obstetric or anaesthetic risk factors, requiring a prior consultation between the attending obstetrician and anaesthesiologist. The multidisciplinary team should communicate closely and formulate plans according to risk stratification if any. Since most of the elective caesarean sections are performed under neuraxial block, the back should be examined noting the presence of any comorbidity like spinal deformities, obesity, and coagulation abnormalities. Any contraindications to spinal anaesthesia (SA) should be identified, and the patient should be evaluated thoroughly for an alternative plan of management.

In our case, we had planned to take it up under single shot spinal anaesthesia. Extra theatre personnel were on standby to help with the shifting and positioning of the patient inside the operating theatre. The operating table was prepared with wide arm boards placed parallel to the table. We arranged for locally available dhotis (cotton loincloths) which were applied over the chest area to support and prevent lateral sagging of the breasts intraoperatively. Postoperatively, bolsters were kept on the sides of the cot to support the breasts and the head end was raised by about 30 degree to prevent respiratory discomfort.

### IV. Conclusion

Gestational gigantomastia (GG) or gravid macromastia is a rare, severely debilitating condition characterized by massive enlargement of breasts. We recommend a thorough and meticulous preoperative evaluation as well as communication with the obstetrical team and theatre personnel in taking up surgical cases of GG for caesarean sections. Proper preparation of the operation theatre is of prime importance in the management of such cases. Appropriately sized operating table as well as wide arm boards placed parallel to the
Regional anaesthesia is a desirable technique of anaesthesia. We recommend using locally available resources like dhotis (cotton loincloths), wrapped in a figure of 8 across the back and breasts, intraoperatively, and bolsters kept on the sides of the arms, postoperatively, to support the breasts of the patient and decrease her respiratory discomfort.

References

Fig 1. Gestational Gigantomastia