Case Report:

Mucoepidermoid Parotid Cancer Rapidly Progressing During Pregnancy

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\textbf{Introduction}

Malignant neoplasm of the salivary gland is rare, with frequency ranging from 0.4 to 2.6 cases per 100,000 population. Of these tumors, mucoepidermoid carcinoma (MEC) is the most common type, and most frequently found in the parotid gland (45\%). However, the incidence in pregnancy is rare and has not been well documented. Endogenous hormones such as estrogen and progesterone have been reported in neoplastic salivary glands, but the results have been conflicting.\textsuperscript{1} In this case, we present a case of MEC that progressed rapidly during pregnancy.

\textbf{Case Description}

A 27-year-old lady, G2P1, presented with a mass on the right cheek that has rapidly enlarged since three months before. She admitted to be eight months pregnant and forgot her last menstrual period. She had antenatal care at midwife and doesn’t have any problems regarding her current pregnancy.

After delivery of her first child five years ago, she felt a small, painless lump around the size of a corn seed, on her right cheek. It did not enlarge until three months ago when she was five months pregnant.

The mass rapidly enlarged until it reached approximately 17 cm in diameter. Since one month ago, the mass ruptured, with active bleeding and pus production. She felt pain, with VAS 2-3. She has normal appetite and could still eat solid food. Neither dysphagia nor dyspnea was found. She did not know about her weight gain/loss during this pregnancy. Her current ECOG performance status was 2.

Her past medical history was unremarkable. She denied smoking, chewing betel leaves, and alcohol consumption. She had history of 3-monthly injectables, works in a garment factory, and exposure to textile fumes was denied.

On physical examination, patient was moderately ill with stable hemodynamics. Her BMI was 23.5 kg/m\textsuperscript{2}, but the middle upper arm circumference was 20 cm. Her conjunctiva was pale; other examination
was within normal limit. On the right buccal region, there was a 17 cm mass with irregular surface and border, ulcerated, firm consistency, and immobile. There was active bleeding (oozing) from the surface. Trismus was positive.

![Figure 1. (a) Tumor mass on the right buccal area (b) Positive trismus](image)

The fundal height was 28 cm with no contractions. Ultrasound revealed EFW 2213 g corresponding to 33-34 wga, with normal AFI and umbilical Doppler studies. No signs of IUGR was found.

Initial lab results revealed normocytic normochromic anemia (Hb 8.3 g/dl) possibly due to bleeding from the ulcerated mass, and hypoalbuminemia (1.8 g/dl). Biopsy result revealed mucoepidermoid carcinoma of the right parotid gland.

Patient was hospitalized for blood transfusion and fetal lung maturation with dexamethasone. A joint conference was held with obstetrician, oncologist, and perinatologist, and decided to perform termination of pregnancy after fetal lung maturation because of recurrent anemia due to bleeding, and to optimize further treatment for the mother. Patient was delivered by cesarean section, born a 2200 g boy with APGAR score 8/9. After cesarean section, patient was treated by oncologic surgeon, and she underwent tumor resection and chemoradiation. Paraffin block and immunohistochemistry examination revealed a mucoepidermoid carcinoma which did not express estrogen nor progesterone receptors.
Discussion
The incidence of head and neck tumors in pregnancy is rising. Mucoepidermoid tumors most frequently present as firm, fixed, and painless swellings. Other symptoms may include pain, otorrhoea, paraesthesia, facial nerve palsy, dysphagia, bleeding, and trismus. In our case, the tumor rapidly progressed since the second and third trimester of pregnancy. This phenomenon leads to the question whether hormones may be related to the tumor growth.

Reports regarding estrogen (ER) and progesterone receptor (PR) expression in salivary gland tumors have been conflicting. IARC reported that ER have been found in minority of mucoepidermoid carcinoma, and PR in 30% of cases. Another study by Nasser (2003) showed that 91% of salivary gland tumors did not express ER nor PR. In the subtype study of mucoepidermoid carcinoma, 1/10 cases each showed weak expression of ER and PR. Pires (2004) examined ER expression in 136 cases of MEC, and all of them showed negative expression for ER. In our case, both ER and PR expression was negative. Palluch (2011) reported a similar case of pleomorphic adenoma of the parotid gland rapidly growing in the third trimester of pregnancy, which also did not express ER nor PR. Although we could not define a direct relationship between pregnancy and the rapid tumor growth, it is possible that other hormones produced by the placenta such as growth factors and human placental lactogen, may contribute to the tumor growth. False-negative results may also be a contributing factor, due to differences in tissue fixation, sensitivity and specificity of the antibodies used, methods used, or criteria used for judging a tumor positive for the marker.

Decision for pregnancy termination in cases of malignancy is an individual decision and depends on the parental option, oncologic prognosis, and the urgency of cancer treatment initiation. In general, termination could be postponed until when fetal maturity has been achieved if there is not urgent indication. All cases of pregnancy complicated by malignancy should be managed by a multidisciplinary team of specialists. In our case, the multidisciplinary team consisted of obstetricians, perinatologist, and oncologic surgeon. Decision to terminate the pregnancy at 34 weeks of gestation after a course of antenatal steroids due to recurrent anemia due to bleeding and to optimize further treatment.

Figure 2. Surgical resection of tumor after delivery
for the mother. It has been reported that high basal metabolic rates associated with malignancy may lead to a catabolic state, causing secondary detrimental effects on the fetus.2

Conclusion
Mucoepidermoid carcinoma progressing rapidly in pregnancy may be due to other factors produced by the placenta besides estrogen and progesterone. Management should be conducted with a multidisciplinary team and individualized to the condition of each patient.

References