CASE REPORT

Acinic cell carcinoma of the parotid gland in pregnancy: an approach to cancer in pregnancy

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SUMMARY

A 27-year-old woman presented with an enlarging painless right preauricular mass at 28 weeks' pregnant. The mass had been stable for more than 10 years, but showed rapid growth during pregnancy. Imaging and biopsy were consistent with parotid gland malignancy, with surgical resection undertaken at 33+4 weeks' gestation. Histopathology confirmed acinic cell carcinoma. Labour was induced without complication at 36+6 weeks' gestation and adjuvant radiotherapy commenced 2 weeks postpartum. At 9 months follow-up, both mother and baby were well, with no signs of disease recurrence. Rapid progression in pregnancy, of a previously stable salivary gland mass, is a common feature among reported cases and was also observed in the current case. This suggests an aetiological link between pregnancy and salivary gland tumour progression. We demonstrate successful management of a parotid gland malignancy in pregnancy and review guiding principles for cancer management in pregnancy.

BACKGROUND

Cancer complicates approximately 1:1000 pregnancies.¹ This rate is increasing as maternal age increases. Cancer in pregnancy presents complex medical, ethical and psychological challenges and should be managed by a multidisciplinary team.² The most common cancers seen in pregnancy include: breast cancer, cervical cancer, lymphoma, ovarian cancer and melanoma.¹

Cancer of the salivary glands is rare, with an incidence of 1.3 per 100 000 people per year.³ The coincidence of salivary gland cancer with pregnancy is even more rare, with only a handful of cases reported internationally.⁴⁻⁶ Rapid progression in pregnancy, of a previously stable salivary gland mass, is a common feature of these cases,⁴⁻⁷ suggesting an aetiological link. We report a case of parotid gland acinic cell carcinoma which demonstrated rapid progression in pregnancy and describe its successful multidisciplinary management.

CASE PRESENTATION

A 27-year-old East-Asian non-English speaking woman presented with an enlarging painless right preauricular mass at 28 weeks' pregnant. The mass had been stable in size and asymptomatic for more than 10 years and had not previously been imaged or biopsied, but showed rapid growth during pregnancy.

INVESTIGATIONS

Ultrasound and MRI were undertaken, demonstrating an 18×22×34 mm heterogeneous right parotid mass (figure 1). Fine needle aspiration biopsy suggested high grade primary salivary gland malignancy.

TREATMENT

Following multidisciplinary team assessment, superficial parotidectomy and selective cervical lymph node dissection was undertaken at 33+4 weeks' gestation (figure 2). Histopathology confirmed dedifferentiated acinic cell carcinoma that was oestrogen and progesterone receptor negative, with no nodal involvement. Labour was induced close to term (36+6 weeks' gestation), to facilitate timely commencement of adjuvant radiotherapy, which had been delayed due to pregnancy. Labour progressed to normal vaginal delivery of a 2985 g term female infant, with no maternal or neonatal complication. Adjuvant radiotherapy directed to the right parotid bed was commenced 2 weeks postpartum, within 6 weeks of surgery.

OUTCOME AND FOLLOW-UP

At 9 month follow-up, 6 months post radiotherapy, the patient was disease free, the surgical wound had healed well and there was no evidence of any adverse effect of radiotherapy. A mild facial nerve neurapraxia present in the initial postoperative period resolved completely. The 8-month-old female infant was healthy and meeting developmental milestones.

DISCUSSION

Cancer in pregnancy

The management of cancer in pregnancy requires consideration of the effect of (1) pregnancy on the cancer; (2) the cancer on the pregnancy and (3) cancer investigation and treatment on the pregnancy.⁶ It raises complex moral and ethical issues,² as the goals of preserving both maternal and fetal well-being are often in direct conflict. Given the relative rarity of these cases, an individualised, multidisciplinary management approach should be adopted, taking into account the type and location of tumour; grading, staging and prognosis of disease; sensitivity to chemotherapy and radiotherapy; gestational age at presentation; pre-existing pregnancy complications and patient wishes.²

The principles of management of cancer in pregnancy include^{1,2}:

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Figure 1 Coronal and axial T2-weighted MRI showing right parotid mass at 30+5 weeks' gestation, 2.5 weeks after fine needle aspiration biopsy.

- Pregnancy should be continued to close to term in the majority of cases.
- Ultrasonography and MRI are preferred imaging modalities.
- Surgical management is possible throughout pregnancy, but preferred in the second trimester.
- Chemotherapy is possible in the second and third trimesters, but should be ceased 3 weeks prior to delivery.
- Radiotherapy of the upper body is possible, with shielding of the abdomen, in the first and second trimesters.

Salivary gland tumours in pregnancy

Rapid progression during pregnancy, of a previously stable salivary gland mass, is a common feature among the small number of cases of salivary gland tumour in pregnancy reported in the literature.^{4 5 7} This observation suggests an aetiological link

Figure 2 Intraoperative photograph after right superficial parotidectomy, prior to neck dissection, showing preservation of the facial nerve.

between pregnancy and salivary gland tumour progression, which could reflect physiological changes of pregnancy, such as increased immune tolerance or represent an hormonal influence on tumour progression. Our study adds to the body of evidence for the association between pregnancy and salivary gland tumour progression.

Due to the small number of reported cases, there are no established risk factors for salivary gland tumour progression in pregnancy. In addition, the epidemiology of salivary gland malignancy, and salivary gland tumour progression in general, is not well documented due to the heterogeneity of tumour types and sites and the inconsistencies in data gathering internationally.⁸ Exposure to ionising radiation is a known risk factor for salivary gland tumour. Females are more commonly affected, but this is dependent on tumour type.⁸ There is some evidence for predisposition based on ethnicity, association with viral infection (such as Epstein-Barr virus and cytomegalovirus) and association with certain occupational exposures, but none of these have been demonstrated conclusively.⁸

There are several lines of evidence hinting at a role for sex hormones in the progression of salivary gland tumours. Studies have shown the presence of oestrogen and progesterone receptors in a proportion of normal and neoplastic salivary glands.⁸ There is some evidence to suggest that salivary gland malignancy in women may share risk factors with endometrial, ovarian and breast cancers, such as early menarche and nulliparity.⁹ A more recent, larger case-control study did not bear this out, except for a non-significant relationship between salivary gland cancer and older maternal age at first birth, a trend shared with endometrial and ovarian cancers.¹⁰ In der Maur *et al*¹¹ demonstrated an increased risk of breast cancer, within a median follow-up period of 5.4 years, among women with salivary gland tumours. In the face of this evidence for an hormonal influence on salivary gland tumour progression, our case demonstrated rapid progression in pregnancy despite being oestrogen and progesterone receptor negative.

We demonstrate successful multidisciplinary management of a salivary gland malignancy, with definitive surgery during pregnancy, delivery at close to term with good neonatal outcome and minimal delay to adjuvant therapy. We suggest that the obstetrician should be a full and active member of the multidisciplinary team, a factor that was critical to the optimal management of our case.

Learning points

- There is a possible aetiological link between pregnancy and salivary gland tumour progression.
- This link may relate to increased immune tolerance or hormonal changes in pregnancy, but occurred in our case despite oestrogen and progesterone receptor negativity.
- Cancer can be managed successfully in pregnancy (including judicious use of surgery, chemotherapy and radiotherapy), without unnecessary risk to maternal or fetal well-being, by respecting certain guiding principles.

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REFERENCES

- Salani R, Billingsley CC, Crafton SM. Cancer and pregnancy: an overview for obstetricians and gynecologists. *Am J Obstet Gynecol* 2014;211:7–14.
- Amant F, Han SN, Gziri MM, *et al.* Management of cancer in pregnancy. *Best Pract Res Clin Obstet Gynaecol* 2015;29:741–53.
- Australian Institute of Health and Welfare. *Head and neck cancers in Australia. Cancer series no. 83 Cat. no. CAN 80.* Canberra: AIHW, 2014.
- Al-Zaher NN, Obeid AA. Acinic cell carcinoma in pregnancy: a case report and review of the literature. *J Med Case Rep* 2011;5:91.
- Prabhu RV, Dinkar A, Spadigam A, *et al.* Low-grade papillary adenocarcinoma of minor salivary glands in pregnancy. *Indian J Cancer* 2015;52:644–5.
- Atabo A, Bradley PJ. Management principles of head and neck cancers during pregnancy: a review and case series. *Oral Oncol* 2008;44:236–41.
- Palluch F, Lehmann M, Volz J, *et al.* The rapid growth of a pleomorphic adenoma of the parotid gland in the third trimester of pregnancy. *J Med Case Rep* 2011;5:141.
- Barnes L, Eveson JW, Reichart P, *et al.* *World health organization classification of tumours: pathology and genetics of head and neck tumours.* IARC Press: Lyon, 2005.
- Horn-Ross PL, Morrow M, Ljung BM. Menstrual and reproductive factors for salivary gland cancer risk in women. *Epidemiology* 1999;10:528–30.
- Lu Y, Lagergren J, Eloranta S, *et al.* Childbearing and salivary gland cancer: a population-based nested case-control study. *Epidemiology* 2009;20:780–2.
- In der Maur CD, Klokman WJ, van Leeuwen FE, *et al.* Increased risk of breast cancer development after diagnosis of salivary gland tumour. *Eur J Cancer* 2005;41:1311–5.

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