Unusual association of diseases/symptoms

Does lightning strike twice?

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Summary

The authors present an interesting case under our follow-up who has had five different forms of tumours with different pathologies throughout his lifetime. He started off with hepatoma, followed by pleomorphic sarcoma of the thigh, adenocarcinoma of the prostate, meningioma and finally schwannoma. He is still alive to this date.

BACKGROUND

Tumour is defined as an abnormal mass of tissue, the growth of which is basically autonomous and exceeds that of normal tissues. It could be broadly categorised into benign and malignancy. We often see primary lesions (tumour) with metastases, but reports of mixed pathologies (both benign and malignant) occurring in a same patient at different times of their life is rare. We report an interesting case of five different abnormal proliferations causing five different pathologies in the same patient who is still surviving to date.

CASE PRESENTATION

The patient is a 76-year-old gentleman who is on our regular follow-up. He first presented at the age of 42 years with right-sided abdominal pain associated with fever. Physical examination at that time revealed tenderness over the right upper quadrant of his abdomen on deep palpations. Liver enzymes were raised. Ultrasound of hepatobiliary system was suggestive of cholelithiasis. Provisional diagnosis of acute cholecystitis secondary to cholelithiasis was made. He underwent elective cholecystectomy 3 months later. Intraoperatively, there is incidental finding of irregular mass over his liver; it was removed along with cholecystectomy. The histopathological examination of the liver mass was consistent with hepatoma. He remained symptom free postoperatively and was on regular follow-up on his liver function and did not receive any adjuvant treatment.

At the age of 62, he noticed a right posterior thigh swelling. It was gradually increasing in size and causing discomfort. He had no constitutional symptoms. He underwent an incisional biopsy and the histopathology was consistent with pleomorphic sarcoma. Subsequently, he underwent wide resection. CT of the thorax showed lung metastases (two lung nodules). Postoperatively, he underwent 35 cycles of radiotherapy for the operative site and the lung nodules were observed, as they were still small. However, they progressively increased in size over next 5 years on regular CT follow-up and a new nodule was found at the left apical lobe in 2003. He was also suspected of having a recurrence of right thigh sarcoma during the same period, whereby he complaint of swelling over the previous scar

site, and the MRI done showed suspected lesion over the hamstring compartment. The patient refused further surgery at the time.

In the year 2001, he presented to the urology unit with urinary hesitancy and frequency. His total prostate specific antigen (PSA) was 0.57 ng/ml and a clinical diagnosis of benign prostate hyperplasia was made. He underwent a transurethral resection of the prostate. Unfortunately, the histopathology was reported as prostate carcinoma (Gleason 1+1). The patient opted for conservative treatment.

Over the next 3 years, he was on regular follow-up and the thigh swelling and the lung metastasis were closely monitored. In the year 2004, his right thigh swelling started to increase in size. A repeat MRI showed features consistent with recurrence at right thigh sarcoma within the hamstring compartment. He underwent another wide excision of tumour, after which he was symptom free. The three lung nodules were continued to be observed, as they appeared to be stable and non-progressive.

In 2006, he complaint of giddiness on and off, with headache radiating to the back and neck for 4 months duration. He was referred to the ear, nose and throat surgeons and their examination was uneventful. Clinically, he did not exhibit any neurological deficits. CT scan of brain was done and it showed small tumour at left parietal region. This was followed by a MRI examination of the brain. It was suggestive of a meningioma. The patient had craniotomy and tumour excision done concomitantly with thoracotomy and excision of right pleural base mass. The histopathological examination (HPE) confirmed the diagnosis of meningioma (brain tumour) and schwannoma (right pleural base mass).

In the year 2010, he was referred back to urologist as his PSA showed an increasing trend; 6.67 ng/ml (April 2009) to 7.78 ng/ml (June 2009) to 14.9 ng/ml (January 2010). Clinically, he was asymptomatic. Transrectal ultrasound biopsy of both left and right lobe of the prostate was done which showed poorly differentiated carcinoma, likely adenocarcinoma (Gleason 4+5=9). He was immediately started on zoladex injection.

This gentleman is currently in stable condition and on urology and oncology follow-up. The chronologies of events are presented in table 1.

Table 1 Chronology of events

Year (age)	Diagnosis (histopathology)	Treatment
1977 (42 years)	Hepatoma	Surgical removal
1997 (62 years)	Pleomorphic sarcoma of right thigh	Wide excision
2001 (66 years)	Adenocarcinoma of prostate	Conservative treatment
2004 (69 years)	Recurrence of right thigh pleomorphic sarcoma	Wide excision
2006 (71 years)	Meningioma (brain)	Craniotomy with excision of tumour
2006 (71 years)	Schwannoma (right pleural base mass)	Thoracotomy and excision of mass
2010 (75 years)	Adenocarcinoma of prostate	On zoladex injection

OUTCOME AND FOLLOW-UP

The patient is still alive and under our follow-up.

DISCUSSION

This is an interesting case of multiple tumours occurring in a single individual. It is extremely rare for a person to have five different pathologies over his life time and still alive with stable disease. This is a different entity compared with collision tumours that consist of two histologically distinct neoplasms present at one location.¹ This patient had a mixture of three malignancies (hepatoma, pleomorphic sarcoma and prostate adenocarcinoma), and two benign tumours (meningioma and schwannoma). In all of these conditions except the prostatic carcinoma, the tumours were resected surgically and adequate treatment and follow-up was done. Normally when lesions appear in other areas in a person with a known primary, they are assumed to be metastasis and the prognosis is poor.

There have been many publications that describe concomitant schwannomas and meningiomas coexisting and this implies NF 2 gene mutations.²⁻⁵ It has been shown that heterozygous mice can exhibit multiple tumours and this is attributed to the tumour suppressors.⁴ In this patient, we postulate that he probably has a mutation of NF 2 gene with p53 gene linked, which causes him to have five different tumours at five different sites.⁵

In conclusion, tumours suppressor can promote variable mutations; therefore we should always consider the possibility of second pathology even if the patient has a known primary lesion. It is crucial that proper examination and

investigation especially biopsy is performed to provide an accurate diagnosis so that the correct treatment can be instituted.

Learning points

- It is possible to have tumours from different pathology over a lifetime.
- We must not assume that a tumour occurs in an individual with a previous history of tumour as a recurrence of metastasis.
- Having more than one tumour does not necessarily mean a poor prognosis.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Tang GC** Piao YS, Zhao L, *et al*. Lung adenocarcinoma metastasizing to cerebellopontine angle schwannoma (collision tumor). *Acta Neurochir (Wien)* 2007;**149**:87–90.
2. **Kley N**, Seizinger BR. The neurofibromatosis 2 (NF2) tumour suppressor gene: implications beyond the hereditary tumour syndrome? *Cancer Surv* 1995;**25**:207–18.
3. **Haneman CO**. Magic but treatable? Tumours due to loss of merlin. *Brain* 2008;**131**:606–15.
4. **McClatche AI**, Saotom I, Merce K, *et al*. Mice heterozygous for a mutation at the Nf2 tumor suppressor locus develop a range of highly metastatic tumors. *Genes Dev* 1998;**12**:1121–33.
5. **Gao X**, Porte AT, Honn KV. Involvement of the multiple tumor suppressor genes and 12-lipoxygenase in human prostate cancer. Therapeutic implications. *Adv Exp Med Biol* 1997;**407**:41–53.

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