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23RD CLINICOPATHOLOGICAL CONFERENCE ON PITUITARY DISEASE

ABSTRACT BOOK

Friday 17th March 2023

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CPCPD

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5.75 CPD POINTS

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Oral Presentations

A challenging case of persistent Syndrome of Inappropriate Antidiuretic Hormone secretion following traumatic subarachnoid haemorrhage

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Aims

To discuss a challenging case of chronic and persistent SIADH following a traumatic subarachnoid haemorrhage in a young female.

Method

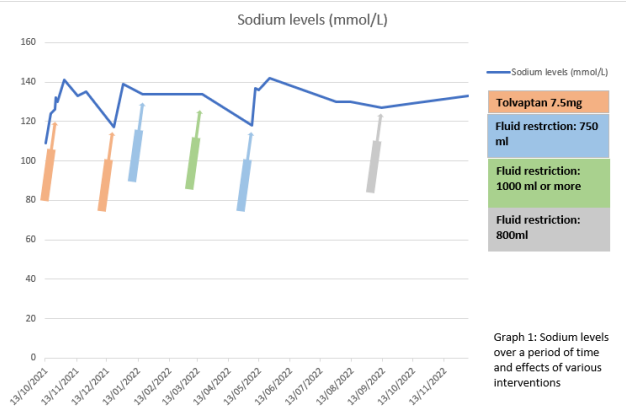
A 32-year-old female presented to hospital following a fall, headache, vomiting. She didn't have any other past medical history of note. She was not on any regular medications. On examination, her GCS was 15/15 with normal neurological examination. She was euvolemic. Her CT head revealed traumatic subarachnoid haemorrhage. Her blood results showed with severe hypotonic hyponatremia with serum sodium levels of 109 mmol/L, serum osmolality of 230 mosmol/kg, urinary sodium of 117 mmol/L and urinary osmolality of 502 mosmol/kg. Her previous bloods done at the GP surgery prior to this admission showed normal serum sodium. The initial clinical presentation seemed more compatible with cerebral salt wasting syndrome with very high urinary output of over 2.5 L per day, very high urinary sodium levels but she was clinically euvolemic. She didn't have any history of recurrent abdominal pain to suggest acute porphyria.

Results

She was given hypertonic saline infusion with difficulties in weaning her off it.
Subsequently, she was given a few doses of Tolvaptan with a good response to increase in sodium levels and she could be weaned of hypertonic saline infusion.Her results are compatible with the diagnosis of SIADH. Her CT Thorax, Abdomen and Pelvis was unremarkable except 1.2cm liver haemangioma. Her renin (0.8 nmol/h/l) and aldosterone levels (490 pmol/L), thyroid function test were with in normal range. Her 9 am cortisol levels were 411 nmol/L (mid-day off combined oral contraceptive pills). Trend of her sodium levels is depicted in graph 1 with corresponding interventions undertaken. She was discharged home with fluid restriction of 750 ml/24hours. Relaxation of fluid restriction to more than 1000 ml/day leads her sodium levels to drop. Currently she is on fluid restriction of 800 ml/24 hours and we are investigating other potential causes of SIADH.

Conclusion

Hyponatremia is one of the common complications following traumatic brain injury (TBI) due to impaired brain function and neuroendocrine dysfunction. SIADH accounts for >90% of cases of post-TBI. Most of TBI associated hyponatremia develops within a week following trauma and resolve within 6 months of insult. It is important to rule out other potential causes e.g., Cerebral Salt Wasting (CSW) syndrome and hypocortisolism. Our patient had normal 9 am cortisol which ruled out hypocortisolism and the fact she had responded to Tolvaptan and currently responding to fluid restriction rules out CSW. There is a case report in literature about prolonged and recurrent hyponatremia following TBI, however, in that case there was a trigger/aetiology for recurrent hyponatremia e.g. surgical intervention. Here with this case, we present a challenging and rare, case of chronic and persistent SIADH following a traumatic subarachnoid haemorrhage.



Final sodium picture.png

Case of cycling Cushing's - unpredictable and still challenging

Dr. Quazi Islam [1], ***Dr. Eleni Armeni*** [1], ***Prof. Stephanie Baldeweg*** [2], ***Ms. Anouk Borg*** [3], ***Prof. Pierre Bouloux*** [1], ***Dr. Randa Eltayeb*** [1], ***Ms. Glenda Jiminez*** [3], ***Ms. Inma Serrano*** [3], ***Dr. Xilin Wu*** [2], ***Dr. Dipesh Patel*** [1]

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Aims

Introduction: Cyclical Cushing's is known to cause fluctuating symptoms which are not always accompanied by classical clinical features of excessive cortisol production. Pathophysiological mechanism of intermittent hypersecretion of cortisol and/or adrenocorticotropic hormone remains unclear, but intermittent pituitary hypersecretion is often implicated. We report the case of a 64-year-old lady who initially was referred to our clinic for assessment of weight gain.

Discussion:

The patient gave a history of significant weight gain and swelling of her face and ankles, with facial plethora in early 2021. These symptoms subsided over a couple of weeks and recurred after few months. The first clinical assessment in July 2021, there was no clinical stigmata of Cushing's syndrome evident. A post overnight Dexamethasone suppression test cortisol measurement at 9am was 22nmol/L. In April 2022, she presented with clinical features compatible with florid Cushing's; significant ankle swelling, thinning of skin, facial plethora and rapid 5kg weight gain.

Method

Repeat blood sampling showed: 9am Cortisol of 1534 nmol/L, paired adrenocorticotrophic hormone (ACTH) 146ng/L (range: 7.2-63.3), 24-hour urinary Cortisol in 2.55 L was 18,192nmol (range: 0-125). Following the Low Dose Dexamethasone Suppression Test, 9am-cortisol was 127nmol/L at +48hrs. Initial MRI pituitary did not demonstrate a pituitary abnormality; a repeat scan 14 months later was suggestive of mild asymmetry and distortion of the infundibulum to the right.

She underwent an FDG PET CT and Ga68 Dotatate PET scans for further assessment, neither of which demonstrated any abnormal avidity. Incidental non-avid lung nodule was identified which raised the suspicion of possible ectopic ACTH. She underwent a bronchosopic lung biopsy, which showed no significant abnormalities, immunohistochemistry analysis revealed no abnormalities.

She was started on Ketoconazole and Dexamethasone as block-and-replacement regimen to manage glucocorticoid excess. She subsequently developed lower limb deep vein thrombosis (DVT) and was initiated on anticoagulation.

Results

At this stage, we planned for inferior petrosal sinus sampling (IPSS) to confirm centralization of her ACTH source.

The IPSS was carried out in November 2022. The basal ACTH was 36.3 nmg/L at periphery, with a Left Jugular value of 41.7 **(Ratio 1.15)** and Right Jugular value of 37.3 **(Ratio 1.02)**.

The 3-minute post CRH value showed a peripheral ACTH of 36.1 with a left Petrosal Sinus value of 207.0 **(Ratio 5.73)** and Right Petrosal Sinus value of 131.0 **(Ratio 3.63)**

8-minute post CRH value showed a peripheral ACTH of 45.3 with a left Petrosal Sinus value of 265.0 **(Ratio 5.85)** and Right Petrosal Sinus value of 219.0 **(Ratio 4.83).**

It suggestive of more Central than Peripheral response to CRH and hence a decision of Hypophysectomy was

made based on the pituitary multidisciplinary meeting discussion.

Conclusion

No clear pituitary tumour seen on surgey, decision for hypophysectomy made to have a greater chance of treating patient's Cushing's. Cleared pituitary fossa was visualised at the end.

Pituitary histology showed Crooke's hyaline changes with positive staining for ACTH, suggestive of Cushing's, a discrete adenoma was not confirmed by histopathology.

Post-surgical cortisol concentrations were low (73 nmol/l -day 1 and 38 nmol/L -day 2). She was started on replacement doses of Hydrocortisone and discharged with ongoing surveillance. On week two after the surgery, she reported typical recurrence of facial plethora and ankle swelling; contemporary blood cortisol level was elevated at 1138 nmol/l. Her case was re-evaluated in the MDT with plans for further pituitary investigation.

Conclusion: This is a description of a case of cyclical glucocorticoid excess and outlines some of the challenges and co-morbidity seen in such cases. Further contemporary information pertaining the case will be presented for discussion.

Results:

An IPSS was carried out in November 2022.

The result tabulated below

Time	Clock time	Test done	Peripheral	Central	Central
Basal	10:20	ACTH	Peripheral 36.3	Left jugular 41.7 Ratio: 1.15	Right jugular 37.3 **Ratio: 1.02**
Basal	10:40	**CRH given at 10:43**	Peripheral 35.1	Left petrosal sinus 69.6 **Ratio 1.98**	Right petrosal sinus 54.9 **Ratio 1.55**
3 min	10:46	ACTH	3' Peripheral 36.1	3' Left petrosal sinus 207.0 **Ratio 5.73**	3' Right petrosal sinus 131.0 **Ratio 3.63**
8 min	10:51	ACTH	8' Peripheral 45.3	8' Left petrosal sinus 265.0 **Ratio 5.85**	8' Right petrosal sinus 219.0 **Ratio 4.83**
15 min	10:58	ACTH	15' Peripheral 48.5	15' Left petrosal sinus 154.0 **Ratio 3.18**	15' Right petrosal sinus 147.0 **Ratio 3.03**

Ipss result.png

Diabetes Insipidus caused by Autoimmune Lymphocytic hypophysitis resulting in diagnosis and cure of Diffuse Large B-Cell Lymphoma

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1. *Barking, Havering and Redbridge University Trust*

Aims

An association between lymphoma and pituitary dysfunction is well documented, though most commonly through mechanism of infiltrative pituitary metastasis [1, 2, 3], or alternatively primary lymphoma of the pituitary gland [4, 5, 6], rather than through autoimmune lymphocytic hypophysitis.

Here we present a highly unusual case whereby investigation into the cause of central Diabetes Insipidus (DI) with radiological features of lymphocytic hypophysitis, led to diagnosis and cure of extranodal (bone marrow) Diffuse Large B-cell lymphoma (DLBCL).

This case highlights the importance of thorough investigation into pituitary dysfunction. Diabetes Insipidus with an MRI suggestive of autoimmune hypophysitis has been described [3]. However, to our knowledge this is the first case where this presentation has been attributed to a paraneoplastic phenomenon. In our patient's case the diagnosis was associated with a favourable outcome.

Method

A 67 year old lady presented to her GP in March 2019 with sudden onset polyuria and polydipsia associated only with headache lasting for three days. Her GP astutely diagnosed diabetes insipidus. MRI pituitary was highly suggestive of lymphocytic hypophysitis, with heterogeneous enhancement of the pituitary gland, thickening and heterogeneous enhancement of the pituitary stalk, and absence of the high T1 signal of the posterior pituitary lobe. Serum osmolality was 308 mosmol/kg; paired urinary osmolality was 132 mosmol/kg. Anterior pituitary function tests were normal.

She was commenced on Desmopressin, with complete and instantaneous resolution of symptoms. Comprehensive investigation into the underlying cause of her clinical and radiological presentation included CT Chest/Abdomen/Pelvis which revealed heterogenous sclerosis of multiple vertebral bodies. Subsequent MRI whole spine demonstrated widespread bone marrow changes throughout the spine supporting a diagnosis of metastatic disease, though the underlying diagnosis remained elusive.

Results

Clinical examination remained normal. Myeloma screen and Mammogram were normal. Several attempts at vertebral bone biopsy proved unfruitful, with non-diagnostic samples. Eventually PET-CT was arranged which demonstrated multi-focal FDG avid sclerotic foci throughout the axial and proximal appendicular skeleton. No primary lesion or organomegaly was identified to delineate an underlying cause for these findings. Finally, histopathology from a CT guided biopsy of the left anterior ilium revealed features of a high-grade Diffuse Large B-Cell Non-Hodgkin Lymphoma of Germinal Centre Cell (GCC) type.

Haematology commenced RCHOP with 50% doxorubicin/cyclophosphamide and GCSF in line with COVID pandemic attenuation guidance, increasing to standard dose after the first two cycles, and completing 6 cycles in July 2020. PET-CT in August 2020 showed complete metabolic response. She remained well up to February 2021 at which time she relocated.

Conclusion

Differential diagnosis of hypophysitis is broad.

We propose that in our patient's case, DLBCL presented with paraneoplastic hypophysitis. Alternative serendipitous identification of concurrent DLBCL in a patient with autoimmune lymphocytic hypophysitis is a possibility. Our patient's MRI findings were typical of lymphocytic hypophysitis, with characteristic stalk thickening and loss of T1 posterior lobe hyperintensity [7]. There was no cavernous sinus infiltration or sella sclerosis often seen with pituitary metastasis [8, 9]. Interval MRI four months later showed significant spontaneous regression of stalk changes, prior to lymphoma treatment commencing. Whilst low-grade lymphomas can be associated with waxing and waning phenomenon, this is not the case with CNS lymphoma. The pituitary was not FDG-avid on PET-CT.

Paraneoplastic autoimmune hypophysitis is becoming increasingly recognised [10]. Cases of anti-PIT-1 and anti-ACTH driven paraneoplastic autoimmune hypophysitis causing anterior pituitary dysfunction have been described [11,12]. Paraneoplastic posterior pituitary dysfunction may be an emerging area of interest.

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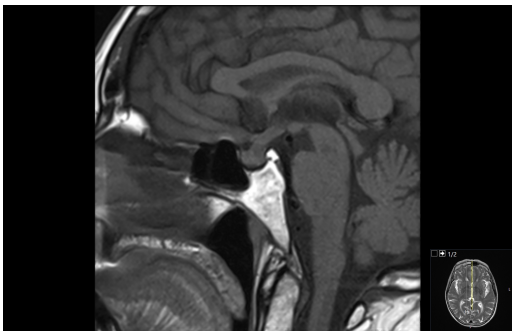
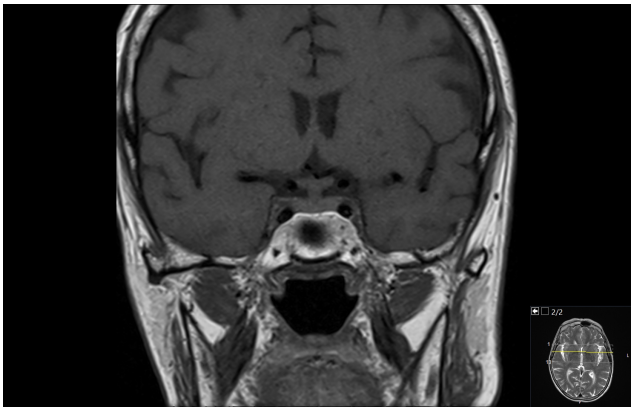


Mri pituitary sagittal.jpg



Mri pituitary coronal.jpg

Diabetes Insipidus in Pregnancy

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Aims

We present the case of a thirty-year-old woman who was diagnosed with diabetes insipidus at 20 weeks' gestation. A history revealed that her symptoms had started two years previously and were exacerbated by pregnancy.

Diabetes insipidus is a state of increased water loss due to the reduction of ADH secretion or activity, leading to concentrated blood and inappropriately dilute urine. Its presentation is characterised by polydipsia and polyuria. The diagnosis of DI is complicated by physiological changes to water homeostasis in pregnancy: changes in the sensitivity of the central osmostat mean that normal serum osmolality falls by 10 mosmol/L by 2-4 weeks' gestation. Pregnant women also feel thirst at lower serum osmolarities and pass higher volumes of urine.

There are currently no consensus guidelines on the diagnosis of diabetes insipidus during pregnancy and this case aims to highlight best practice. It also endorses MDT management of pituitary disorders.

Method

This 30-year-old woman was referred to Obstetric Endocrine clinic with excessive polyuria and polydipsia. She reported drinking up to twenty litres of water per day, and nocturia three to four times per night.

Her past medical history included gestational diabetes and surgical evacuation of a first trimester 'missed miscarriage' two years previously.

A history revealed that her symptoms began during the previous pregnancy. This pregnancy ended in a miscarriage at 11 weeks but her symptoms persisted, drinking 10 Litres of water per day. In her second pregnancy the symptoms became intolerable, and she presented to Endocrinology for the first time.

After review in Endocrinology clinic, her random serum osmolality was 286 mosmol/kg with paired urine osmolality of 37 mosmol/kg. She was normotensive with normal liver function and thyroid profile.

A non-contrast MRI at 30 weeks' gestation showed enlargement of the pituitary stalk.

Results

This patient was treated with nasal DDAVP on the strength of her history and her biochemical results, titrating the dose to her symptoms. She was monitored regularly for water intoxication and hyponatremia.

MRI at presentation revealed a symmetrically enlarged pituitary gland and thickened stalk. Her age, pregnancy, symmetrical gland and enlarged stalk point to a diagnosis of autoimmune lymphocytic hypophysitis. It seems likely from the history that this caused Diabetes Insipidus in her previous pregnancy.

She delivered a healthy baby and stopped taking DDAVP. A post-partum water deprivation test was stopped after three hours as she felt too unwell to continue (see image for results) and confirmed a diagnosis of persistent central diabetes insipidus.

The rest of her pituitary function was normal, apart from a moderately raised prolactin. Her MRI changes resolved on a contrast study post partum, but the D.I. persisted. She remains well on DDAVP treatment.

Conclusion

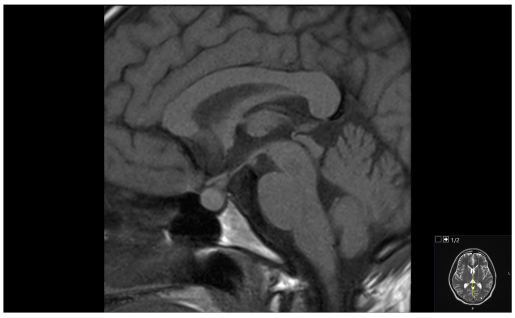
Diabetes Insipidus can be mediated by a failure of ADH secretion (central); increased metabolism of ADH; or insensitivity to its effects at the renal tubule (nephrogenic).

In the case of this patient, it is likely that she had a primary deficiency of ADH due to lymphocytic hypophysitis from a previous pregnancy. This diagnosis was made during her second pregnancy – in pregnancy the vaso-

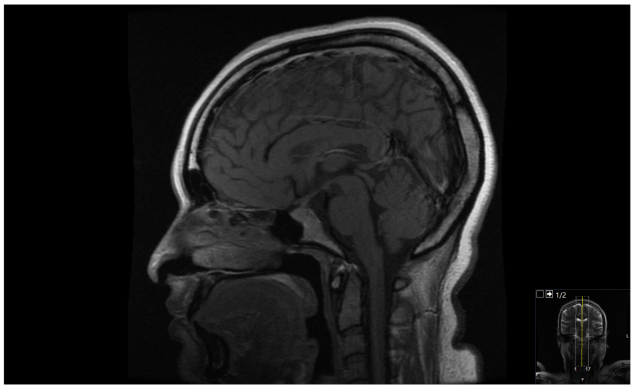
pressinase activity of placental cystine aminopeptidases leads to a 3-4 fold increase in ADH metabolism. Her pituitary was unable to meet this increased demand. Another mechanism of enhanced ADH metabolism in pregnancy is secondary to HELLP/ liver impairment but this was ruled out.
In this case our patient was treated successfully with desmopressin, which is safe in pregnancy and required regular monitoring for water intoxication and hyponatremia. dDAVP is more resistant to placental vasopressinases than endogenous ADH.

TIME	SERUM OSMOLALITY	URINE OSMOLALITY	URINE VOLUME	WEIGHT
	(mosmol/Kg)	(mosmol/Kg)	(ml)	(Kg)
09:00	294			86.2
09:30		71	390	
10:00	298			
10:30		79	460	
11:00	299			
11:30		107	530	
12:00	304			83.4
12:30		DDAVP given 2 ug IM		
13:00				
13:30				
14:00				
14:30	302	257	500	
15:00				

Water deprivation test report.png



Mri pituitary in pregnancy.png



Mri after pregnancy.png

Diagnosing and managing growth hormone (GH) excess in endocrine syndromes in children

Dr. Julie Park [1], ***Mr. Ajay Sinha*** [1], ***Dr. Christina Daousi*** [2], ***Prof. Jo Blair*** [1]

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Aims

Growth hormone (GH) excess is extremely rare in children. It can be associated with endocrine syndromes such as MEN1, carney complex and Mc Cune Albright syndrome. We aim to describe two cases of GH excess, their management thus far and potential treatment options including surgery, radiotherapy and medications such as somatostatin analogues.

Method

Patient L (15M) presented with fibrous dysplasia affecting his cranium (see Figure 1) and a large café-au-lait spot covering his right scapula. His height was 193cm (+2.6 SDS) and BMI 29.6kg/m^2(+2.3SDS). His IGF1 was 84.1nmol/L (22.5 – 65.9) and did not suppress his GH during an oral glucose tolerance test (OGTT) (GH nadir 1.43ug/L). Skeletal survey showed no other bony lesions. MRI of the pituitary was normal. Ophthalmology showed no effect on optic nerve or visual fields.

Patient T (15M) had an antenatal diagnosis of a PRKAR1A gene change consistent with Carney Complex. He had atrial myoxmas removed aged 3 and 8 years. He has evidence of large cell calcifying tumours of the testes, presenting at 8 years of age. Puberty commenced aged 10 years. Height SDS increased from 1SDS to 2.8SDS (See Figure 2). He did not suppress his GH in response to an OGTT (GH nadir 3.18ug/L).

Results

Both children were discussed within an MDT and with colleagues nationally. Treatment options including watchful wait, somatostatin analogues, growth hormone antagonists, radiotherapy and pituitary surgery were discussed. Both patients will commence Lanreotide®, a somatostatin analogue.

Patient L chose to commence medical therapy immediately as previous literature suggests that growth of fibrous dysplasia lesions will slow or stop. However, shrinkage has not been described. Surgical intervention of the craniofacial lesion, for patient L, may be considered for cosmetic reasons.

Patient T initially underwent a process of watchful waiting. However, a follow up MRI showed an enlarging pituitary lesion, therefore medical therapy was commenced. Both patients have opted for medical therapy to reduce the risks of multiple pituitary hormone deficiencies associated with surgery and radiotherapy.

Conclusion

GH secreting tumours are rare in childhood. They often present at the age when a pubertal growth spurt is also anticipated. Close observation is required to ensure that diagnoses are not missed including history, examination, auxology, biochemical testing, imaging of the pituitary and ophthalmology assessment. Treatment options include watchful wait requiring active observation; somatostatin analogues; GH antagonists which are rarely used in children and little data are available regarding their use; pituitary surgery comes with possible life changing complications and multiple pituitary hormone deficiencies. Remission rate is not 100%.

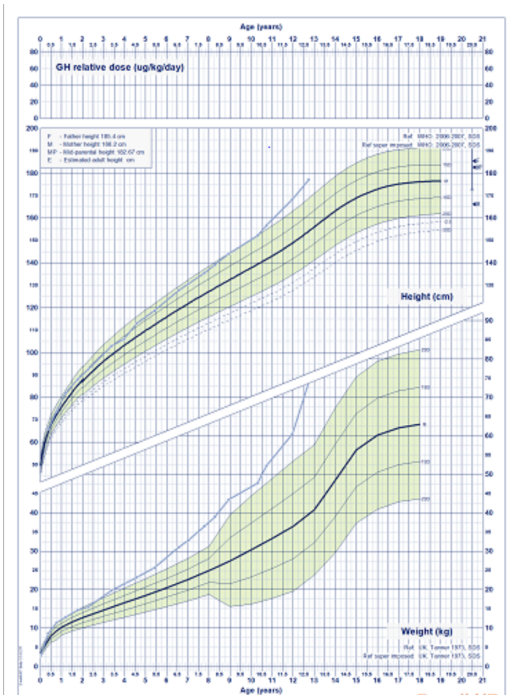


Figure 2 graph showing height and weight for patient t.png

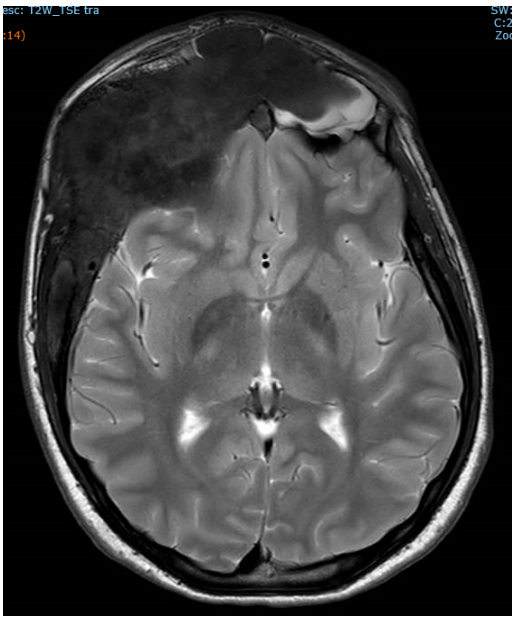


Figure 1 mri of patient l showing craniofacial fibrous dysplasia.png

Double Diagnosis Casting Doubt

Dr. Xilin Wu [1], ***Ms. Joan Grieve*** [2], ***Dr. Teng-Teng Chung*** [1]

1. *University College London Hospital, London,* ***2.*** *National Hospital for Neurology and Neurosurgery, London*

Aims

Case

A 45 -year-old lady was referred to Endocrinology clinic with primary hypothyroidism, on thyroxine replacement for over 20 years. Despite apparently good compliance with medications, blood results at referral were as follows: TSH 5.69 (0.27 - 4.2) mU/L, fT4 30.9 (12-22) pmol/L, fT3 7.2 pmol/L (4-6.8) pmol/L. She complained of weight gain, severe fatigue and oligomenorrhoea. She appeared clinically euthyroid. It transpired that for many years doctors in the UK and Greece struggled to keep her TSH within the reference range.

Method

Investigations with us demonstrated raised TSH and elevated fT4 and fT3 on two different assay platforms, confirming no TFT interference. The rest of her pituitary profile was unremarkable. Repeat bloods after stopping levothyroxine for 3 weeks showed TSH of 14.9 mU/L, fT4 16 pmol/L, fT3 5.6 pmol/L. SHBG and TPO antibodies were elevated at 173 (27-146) nmol/L and 122 (0-34) IU/ml respectively. α-subunit 1.31 (<1.0) IU/L. TRH stimulation test demonstrated a flat response (baseline TSH 11.7, peak 18.3 mU/L at 20 minutes), suggestive of a TSHoma. MRI confirmed a clear pituitary lesion extending towards and remodelling the clivus (Figure 1). Subsequent discussion at MDT recommended pituitary surgery and this was planned for February 2020.

Results

The patient was a Greek historian and was given a second opinion by an eminent Professor of Endocrinology when she was acting as a family “tour guide”. Doubt was cast on her diagnosis as another MRI pituitary performed in Greece did not demonstrate a pituitary lesion.

Ultimately, the consensus was this lady most likely had both primary hypothyroidism and a TSHoma, where partial gland failure from Hashimoto’s may have masked the full effects of the TSHoma (seen by fT4 and fT3 levels within the reference range when off thyroxine treatment). It was felt a methionine PET-CT/MRI and preoperative treatment with somatostatin analogue could offer both diagnostic clarity and therapeutic effect. She received Octreotide LAR 20 mg monthly for three months. Unfortunately, due to the COVID-19 pandemic, she was unable to have the methionine scan.

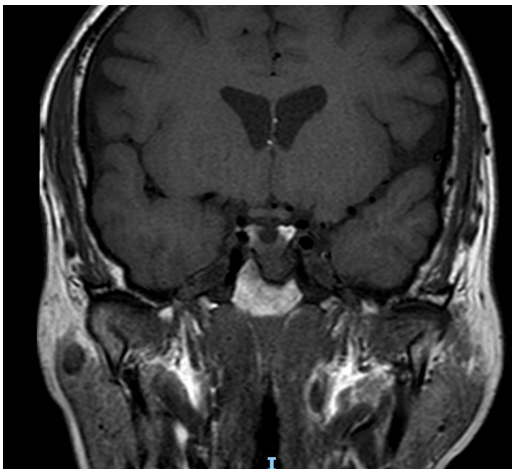
Conclusion

Pre-and post-Octreotide bloods demonstrated a good response to Octreotide treatment, with reduction in TSH, fT4 and fT3 levels (Table 1). She also developed symptoms of hypothyroidism on somatostatin analogue and required thyroxine replacement. There was no change in size of the adenoma on repeat imaging.

She proceeded to have endonasal resection of her pituitary tumour. Histology and immunohistochemistry were consistent with a TSH-expressing thyrotroph adenoma. She reported an improvement in her symptoms postoperatively and remains on levothyroxine 100 micrograms daily (TSH 0.92 mU/L, fT4 22 pmol/L).

Conclusion

This case highlights the importance of revisiting the biochemistry in the context of discordant TFTs, and the diagnostic difficulties associated with dual pathologies. Further points for discussion include the role of functional imaging in the diagnostic work-up of TSHomas, pre-surgical treatment with somatostatin analogues, and strategy for follow-up surveillance in the context of concomitant thyroid disease.



Mri.jpg

	Feb 2020	May 2020	June 2020	Reference Range
TSH	16.8	5.87	5.05	(0.49 – 4.67)
Free T4	1.03	0.71		(0.7 – 2.09)
T4			5.66	(4.5 – 12.0)
Free T3	4.01	2.56		(1.67 – 4.6)
T3			0.82	(0.49 – 1.58)
Treatment	Off thyroxine replacement	1 month Octreotide treatment	3 months Octreotide treatment	

Table 1.jpg

Handheld Robotic Device for Endoscopic Endonasal Skull Base Surgery: Cadaveric Feasibility Study

Dr. Nicola Newall [1], ***Dr. Emmanouil Dimitrakakis*** [2], ***Mr. Danyal Z Khan*** [1], ***Mr. James Booker*** [1], ***Dr. George Dwyer*** [2], ***Mr. John G Hanrahan*** [1], ***Dr. Siddharth Sinha*** [1], ***Dr. Danail Stoyanov*** [2], ***Mr. Hani J Marcus*** [1]

1. *National Hospital for Neurology and Neurosurgery, London, UK,* ***2.*** *Wellcome/EPSRC Centre for Surgical and Interventional Sciences (WEISS)*

Aims

The endoscopic endonasal transsphenoidal approach (eTSA) is emerging as a first-line approach for the treatment of various pathologies of the skull base. The eTSA offers several advantages over traditional transcranial approaches including improved visualisation and reduced brain exposure and retraction. One challenge, however, is the technical difficulties associated with endoscopic instruments. Current instruments lack articulation and offer limited surgical manipulation and dexterity. To enhance this surgical approach, a handheld surgical robot with articulated end effectors for the eTSA has been developed. The objective of this study was to evaluate the feasibility of the handheld robotic instrument for the eTSA in an IDEAL Stage 0 cadaveric study.

Method

One expert neurosurgeon, one intermediate neurosurgical trainee and four novice surgeons were recruited from a single centre UK neurosurgical unit.The eTSA including the durotomy was performed pre-task. Each participant was then instructed to enter and explore the pituitary fossa using the articulated robotic instrument and interact with soft tissue and bony structures. Feedback regarding the precision, dexterity, force application and robustness of the robotic device, and user experience, was obtained in a post-task questionnaire.

Results

All participants (n=6, 100%) reported the articulated robotic instrument provided greater dexterity than existing tools and maintained its structural integrity during the task, suggesting sufficient robustness. However, the ability for sufficient forces to be applied during the movement of the instrument and the lack of precise movements were noted by all participants. All six participants reported the instrument was intuitive and comfortable to use.

Conclusion

Our preliminary results suggest that the use of the handheld robotic instrument for the eTSA is feasible in a human cadaver. We have identified the need for iterative refinements, including increasing force delivery during motion and improving the precision of the end-effectors prior to first in human IDEAL stage 1 studies.

I Only Came in With Toothache

Dr. Gajawathana Sakthilingham [1], ***Dr. Jane Evanson*** [1], ***Dr. Katherine Miszkiel*** [2], ***Prof. William Martyn Drake*** [1], ***Mr. Neil L Dorward*** [3]

1. *St Bartholomew's Hospital,* ***2.*** *University College London Hospital,* ***3.*** *National Hospital for Neurology and Neurosurgery, London*

Aims

In 2013, a 48-year-old lady's complex tooth extraction subsequently required maxillofacial imaging, which revealed the caudal most aspect of a suprasellar lesion. This was further evaluated in detail by MRI, showing an 19 mm (Cranio-caudal), T1 hyperintense T2 hypointense non-enhancing mass lying within the hypothalamus just behind the chiasm and involving the upper aspect of the pituitary stalk. (Figure 1). Within the mass there was a T1 irregular hypo intensity area and on a subsequent non-contrast CT head (Figure 2) these were faintly hyperdense and consistent with small flecks of calcium. The signal characteristics suggested a mixed cystic and proteinaceous lesion.

Method

She was eupituitary clinically and biochemically and had no neuro-ophthalmic deficit on examination. Given the lack of symptoms and evidence of anterior visual pathway compromise, it was agreed that the lesion would be monitored by serial imaging, combined with clinical and ophthalmic review. Menses ceased 4 years later with elevated gonadotrophin levels indicating that this was not consequent upon hypothalamo-pituitary compression.

Results

Between 2013 and 2018, marginal (approximately 1 mm) annual increases in the cranio-caudal dimensions were noted, but apart from computer screen – related headaches she remained well. Minor displacement of the optic chiasm, without active compression was noted. At the latest image (Figure 3) the lesion measured 30 mm (CC) x 28 mm (AP) x 21 mm (TS), compared to 22 mm (CC) x 18 mm (AP) x 19 mm (TS) in 2018. Concern was raised about the proximity of the lesion to the Foramen of Monro and surgical discussions commenced about the advisability of decompressive surgery. The patient remains asymptomatic and anxious about the prospect of surgery and the potential for surgical and endocrine complications balanced against a difficult to quantify risk of developing hydrocephalus.

Conclusion

This case is being presented to canvas views from medical and surgical colleagues about the timing, the extend of and the strategy for any surgical intervention.

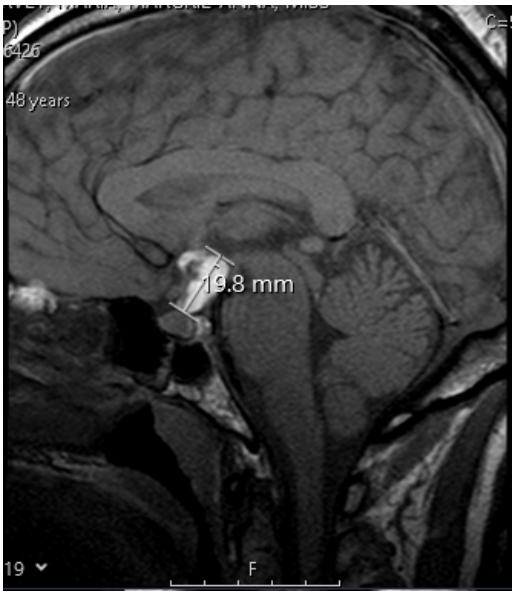
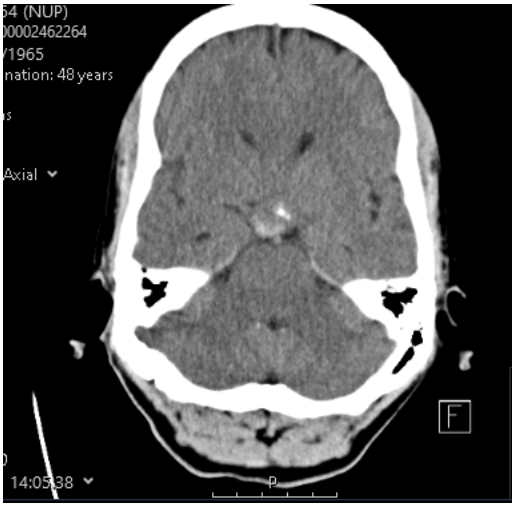


Image 1.png



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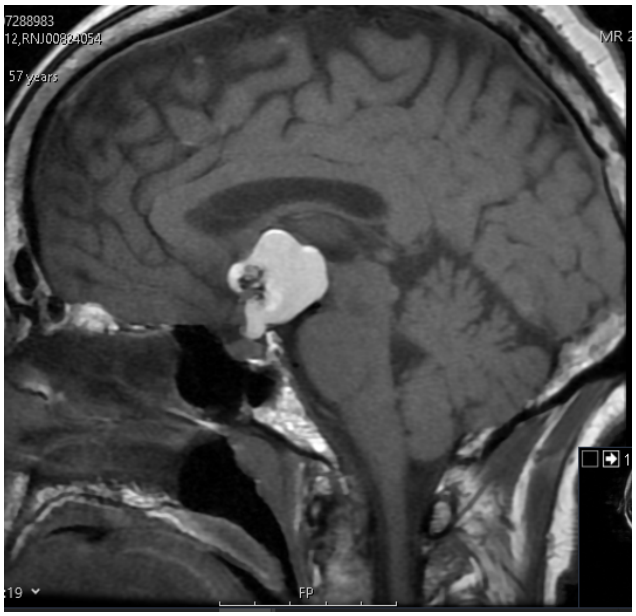


T1 sagital 2022 last .png

Operative Anatomy Recognition During Endoscopic Pituitary Surgery using Artificial Intelligence – A Pre-Clinical Analysis of Operative Videos

Mr. Danyal Z Khan [1], ***Mr. Adrito Das*** [2], ***Dr. Sophia Bano*** [2], ***Dr. Santiago Barbarisi*** [3], ***Ms. Anouk Borg*** [1], ***Dr. Lucy Culshaw*** [3], ***Mr. Neil L Dorward*** [4], ***Mr. John G Hanrahan*** [4], ***Dr. Karen Kerr*** [3], ***Dr. Imanol Luengo*** [3], ***Mr. Simon Williams*** [2], ***Dr. Danail Stoyanov*** [2], ***Mr. Hani J Marcus*** [1]

1. *National Hospital for Neurology and Neurosurgery, London, UK,* ***2.*** *Wellcome/EPSRC Centre for Surgical and Interventional Sciences (WEISS),* ***3.*** *Digital Surgery, Medtronic,* ***4.*** *National Hospital for Neurology and Neurosurgery, London*

Aims

Pituitary tumours are in an anatomically dense region of the body, and often distort or encase surrounding critical neurovascular structures, such as the optic nerves and internal carotid arteries. This, in combination with anatomical variations and limitations imposed by current scope technology, make safe identification and protection of these structures challenging intra-operatively. Advances in artificial intelligence (AI) have allowed accurate segmentation of anatomical structures of medical images. In this IDEAL Stage 0 study, we sought to develop and pre-clinically validate the application of this AI technology to endoscopic pituitary surgery.

Method

A video database of 64 consecutive endoscopic pituitary surgeries performed at a single tertiary neurosurgical centre was prospectively developed. Videos were uploaded and annotated in Touch Surgery™ Enterprise. Ten seconds of the sellar phase, immediately preceding sellotomy, were extracted from each video at 1 image per second. Images were labelled for ten critical anatomical structures through multi-round expert consensus (Figure 1). Labels outlined the visual boundary of each structure, and if not visible due to occlusions (e.g., an instrument), this boundary was excluded. Using Python, descriptive statistics were generated, and a segmentation artificial neural network (ANN) was trained to identify the structures. A dice score was calculated to assess the model performance by assessing the degree of overlap between the AI-predicted anatomy and expert-labelled anatomy.

Results

From the 640 images, the structures identified most through expert consensus were the sella (100%), clival recess (88%), and right optic protuberance (61%). Structures identified the least were the planum sphenoidale (23%), left optico-carotid recesses (40%), and right optico-carotid recesses (42%). The ANN achieved a 75% dice score on automatic sella segmentation, and is currently being refined to accurately recognise other key structures.

Conclusion

A library of over 600 sellar anatomy images usable for education and assessment has been generated. Certain critical landmarks proved difficult to identify consistently, despite multi-expert review, supporting the need for improved optics and surgical adjuncts in selected cases. The development of an AI capable of automatic recognition may assist with intra-operative decision support and synergise with complementary advances in augmented reality.

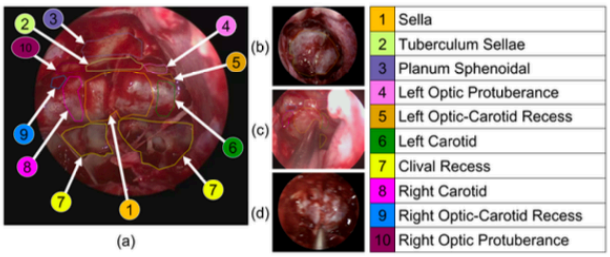


Fig. 1. 10-anatomical-structures semantic segmentation of the sellar phase in endoscopic pituitary surgery, with names given in the legend. Example images of the manual annotations, where each visible critical neurovascular structures' boundary is outlined by a neurosurgeon, are displayed: (a) displays an image where all structures are clearly seen; (b) displays an image where only one structure, the sella, is seen; (c) displays an image where some of the structures are occluded by an instrument and biological factors; and (d) displays an image where none of the structures are identifiable due to blurriness caused by camera movement.

Fig 1.png

Pituitary Ewing's Sarcoma: management of a pituitary tumour guided by DNA methylation and genetic analysis

Dr. ISHTA SHARMA [1], ***Dr. Adrian Li*** [2], ***Dr. Istvan Bodi*** [3], ***Dr. simon aylwin*** [2], ***Mr. Sinan Barazi*** [1], ***Mr. Nicholas Thomas*** [1], ***Ms. Eleni Maratos*** [1]

1. *1. Department of Neurosurgery, King's College Hospital NHS Foundation Trust, London, UK,* ***2.*** *2. Department of Endocrinology, King's College Hospital NHS Foundation Trust, London, UK,* ***3.*** *3. Department of Clinical Neuropathology, King's College Hospital NHS Foundation Trust, London, UK*

Aims

Ewing's sarcoma is a highly aggressive round cell neoplasm which usually occurs in children and adolescents, and has a survival rate of 70-80% in localized disease cases. Primary Ewing's sarcoma of the sella is a rare occurrence, with only one documented paediatric case in the current literature base. We report the first adult case of Ewing's sarcoma presenting as a primary pituitary tumour.

Method

This study is a case report of a 51-year-old female who presented with lethargy and bitemporal hemianopia to a neurosurgical department of a tertiary referral centre hospital. MRI imaging illustrated an 18mm sellar mass with suprasellar extension, displacing the optic chiasm. The absence of fossa enlargement was atypical for an adenoma. Pituitary biochemistry demonstrated no abnormalities. This report outlines the key operative findings in this unusual case, as well as the clinical and radiological features and patient management including surgery and chemoradiotherapy. The current study emphasises the role of genetic analysis and DNA methylation techniques in the diagnosis of this tumour type.

Results

An endoscopic trans-sphenoidal resection of the sellar mass was performed to obtain histological diagnosis. During surgery the tumour was found to be tough and fibrous. Normal pituitary gland was seen separately and preserved. The tumour was adherent to the cavernous sinus wall and resection was complicated by cardiac pauses during manipulation of the tumour. Subtotal resection was performed. There was no improvement in visual deficit postoperatively, with subsequent MRI scans confirming an increase in the lesion size to 24mm. Further surgical resection was planned via a supraorbital approach. Histological analysis was atypical for an adenoma and the Ki67 proliferation index was elevated at 10%. Subsequent next generation sequencing and methylation analysis correctly classified the tumour as Ewing's sarcoma. Chemotherapy and radical radiotherapy were delivered with excellent clinical and radiological response. No further surgery was required.

Conclusion

Unusual intraoperative findings should raise the suspicion of non-adenomatous pathology. The use of DNA methylation techniques confirmed the underlying diagnosis and enabled the successful application of a molecular targeted chemotherapeutic approach. Methylation markers have emerged as a novel tool to increase the sensitivity of prostate and lung cancer detection, and this success has been extended to improving the classification of brain tumours. Given the distinct epigenetic profile of sarcomas, this case exemplifies the diagnostic role of methylation assays for the management of such tumour types.

Shared decision making in Southmead Pituitary surgery

Mx. Rosemarie Bird [1], ***Mr. Daniel Thompson*** [2], ***Dr. Christin Hoffmann*** [3], ***Dr. Faisal Hasan*** [4], ***Dr. Karin Bradley*** [4], ***Dr. Parag Yajnik*** [4], ***Mr. Kumar Abhinav*** [1], ***Mr. Adam Williams*** [2], ***Mr. Angus McNair*** [3]

1. *Southmead Hospital Department of Neurosurgery,* ***2.*** *Southmead Hospital Neurosurgery department,* ***3.*** *University of Bristol,* ***4.*** *Southmead Hospital Department of Endocrinology*

Aims

High quality, patient centred shared decision making (SDM) is a central tenant of modern healthcare systems. SMD is particularly important for patients undergoing invasive procedures because, unlike many medical therapies, the effects are immediate and irreversible. This means that patients cannot discontinue treatment should the benefits fall short of expectations or side effects become unacceptable.

NICE (National Institute for Health and Care Excellence) has recently completed an evidence review and concluded that successful SDM relies on a combination of interventions that support organisations, clinicians, and patients. It recommends

further research to define how to best measure SDM and create a sustained culture change at an organisational/health service level.

Southmead Neurosurgery Pituitary department has taken steps to implement a patient centred consent process prior to to surgery with automated real-time monitoring of patient's experience of the process.

Method

The use of a standardised information booklet containing information on the process and procedure that the patient's are likely to undergo. Built into this is areas for patient's to detail what matters most to them about their health to help frame the clinic consultation. This allows for a patient centred consenting process and means that on the day of surgery only a signature is required rather than an in depth discussion in keeping with the expectations set by Montgomery.

Automated, real-time data collection of the patient experience questionnaire's CollaboRATE, SDM-Q-9 and SHARED are then used in order to ensure patient feedback aligns with the aims of the process. These are virtual questionnaires with virtual prospective assessment of scores. This allows service wide modelling of patient SDM experience and trends related to education of clinicians and allows pre-operative identification of poor SDM experience.

Results

Between April 2021 - December 2022 37 patients were booked for Pituitary surgery and were sent questionnaires. There were 19 non-responders. Of the 18 patients to complete the survey pre-procedure: 18 used CollaboRATE, 10 used SDM-Q-9 and 7 used SHARED.

CollaboRATE feedback recorded a median score of 96/100 with only one low scorer (<46/100). SDM-Q-9 revealed a median score of 83/100 with 2 top scores of 100. The SHARED tool showed a median score of 9/10 with again only 1 low responder.

Conclusion

In the post-Montgomery era clinicians need to be especially mindful of processes to best ensure that they can demonstrate working towards true shared decision making. We believe our process for Pituitary surgery consenting at Southmead aligns well with this and results generally support the conclusion that patient's feel listened to. Realtime feedback also allows for steps to be taken prior to surgery in the few cases when this process may not have functioned optimally. Work is ongoing to optimise acceptability and define ways to best feedback patient experience. We believe the ability to resolve poor SDM experience pre-operatively will reduce on the day cancellation, complaints and litigation.

The Pituitary Outcome Score: Proposal & Recruitment for a National Validation Study

Mr. John G Hanrahan [1], ***Mr. Danyal Z Khan*** [2], ***Ms. Elika Karvendi*** [3], ***Dr. Nicola Newall*** [2], ***Mr. Neil L Dorward*** [1], ***Prof. Stephanie Baldeweg*** [4], ***Mr. Hani J Marcus*** [2]

1. *National Hospital for Neurology and Neurosurgery, London,* ***2.*** *National Hospital for Neurology and Neurosurgery, London, UK,* ***3.*** *Department of Neurosurgery, University of Cambridge, Cambridge, UK.,* ***4.*** *Department of Diabetes & Endocrinology, University College London Hospitals NHS Foundation Trust, London, UK*

Aims

Patient-reported outcome measures (PROMs) can evaluate treatment efficacy, providing outcome measures that align closely with patient objectives. For patients diagnosed with a pituitary adenoma, they are a useful means of assessing quality of life (QoL) which can be used to determine effectiveness of treatment and capture variations in patient experience. In previous work, we developed and validated a PROM tool called the Pituitary Outcome Score (POS) for patients with pituitary adenoma undergoing surgery in a single-centre setting. The benefit of this tool includes the facilitation of outcomes research and quality improvement. Yet, there are wider benefits if scaled nationally, including a standardised and unified measure for assessing QoL. Such a tool would allow national data comparisons, mirroring the success of the Oxford Hip and Knee PROMs. Therefore, we report the protocol for a national validation study of the pituitary outcome score.

Method

We will conduct a multi-centre centre prospective cohort study recruiting centres across the United Kingdom. All neurosurgical units (NSUs) performing the transsphenoidal approach (TSA) for pituitary adenoma will be eligible. Individual centres will register the study as a service evaluation with local governance committees. Consultant neurosurgeons with a subspecialist interest in pituitary surgery will be contacted to recruit consecutive patients attending a pre-operative neurosurgical clinic for TSA to resect a pituitary adenoma. Inclusion criteria will be patients aged 18 or over scheduled for TSA, diagnosed with pituitary adenomas and able to complete the questionnaire. Data collection timepoints will include two pre-operative surveys and two post-operative surveys (3 months and 6 months).

Results

Reliability will be assessed through Cronbach's alpha to evaluate the internal consistency and the intraclass correlation coefficient (ICC) to determine test-retest reliability. The POS will be correlated with the SF-36 questionnaire, a validated QoL tool, to assess validity. Responsiveness to the PROM will assessed through the Spearman's correlation coefficient calculated from the baseline questionnaire and the 6-month post-operative questionnaires, according to patient responses to the Global Perceived Effects (GPE) scale. Interpretability of the POS will be determined through determining the smallest detectable change, minimal important change, minimal important difference and floor and ceiling effects.

Conclusion

This proposal aims to devise the first nationally valid PROM for patients undergoing surgery for a pituitary adenoma. This would provide a nationally valid patient-centred outcome measure, offering a standard UK pituitary surgery PROM.

Title: To treat or not to treat...

Dr. Shani Apsara Dilrukshi Mathara Diddhenipothage [1], ***Dr. James MacFarlane*** [2], ***Dr. Heok Cheow*** [3], ***Dr. Jonathan Jones*** [4], ***Dr. Susan Oddy*** [5], ***Dr. David J Halsall*** [5], ***Prof. Mark Gurnell*** [6], ***Dr. Helen Simpson*** [7]

1. *Department of Diabetes and Endocrinology, University College London Hospital NHS foundation trust, London, United Kingdom,* ***2.*** *Addenbrooke's Hospital, Cambridge University Hospitals NHS Foundation Trust,* ***3.*** *Cambridge Endocrine Molecular Imaging Group, Metabolic Research Laboratories, Wellcome-MRC Institute of Metabolic Science, University of Cambridge, Addenbrooke's Hospital, Cambridge, United Kingdom,* ***4.*** *Cambridge University Hospitals NHS Foundation Trust,* ***5.*** *Clinical Biochemistry, Cambridge University Hospitals NHS Foundation Trust, Addenbrooke's Hospital, Cambridge, UK,* ***6.*** *Cambridge Endocrine Molecular Imaging Group, Metabolic Research Laboratories, Wellcome-MRC Institute of Metabolic Science, University of Cambridge, Cambridge Biomedical Research Centre,Cambridge, United Kingdom,* ***7.*** *University College London Hospital*

Aims

Background

Macroprolactinomas are rare in adolescent age groups and management can be challenging. We describe a case of a macroprolactinoma presenting in a young person aged 12 years, demonstrating the importance of reassessment at transition to adult services, challenges of deciding long term management and, in particular here, with discordance in prolactin assay results.

Clinical presentation and initial evaluation

A 12-year-old girl was referred to paediatric services for evaluation of bilateral upper quadrantanopia that was detected by an optician when assessed for recurrent headache. Subsequent evaluation with magnetic resonance imaging (MRI) showed a large pituitary macroadenoma with suprasellar extension and chiasmal compression without cavernous sinus invasion. Serum prolactin 38,531 mU/L (RR 63-318), LH 0.5 IU/L, FSH 2.6 IU/L, Free T4 16.3 pmol/L, TSH <0.1(0.4-4) mU/L, Glucagon stimulation test; peak growth hormone(GH) <0.2µg/L, peak cortisol 510 nmol/L.

Method

One paternal uncle was diagnosed with large non-functioning pituitary adenoma and panhypopituitarism. Genetic studies were negative for FIPA and MEN-1. Awaiting genome wide studies.

Initial management

Cabergoline 0.25mg/twice weekly was started along with levothyroxine and GH replacement. Rapid normalization of vision was noted along with tumour shrinkage on MRI (Figure 1) imaging. Prolactin level dropped to 638mIU/L in 3 months with cabergoline dose of 1.25mg/ weekly.

However, serum prolactin level started to rise to >2000 mU/L and cabergoline dose was increased despite progressive reduction of tumour size on imaging. In the meantime, oestradiol replacement was started to support puberty aged 15 years. She was then managed with femoston 2/10 and levothyroxine, GH replacement was stopped at the age of 18 after completion of linear growth.

Results

Review of the treatment plan

At transition to adult care, her prolactin level was >1500mU/L (Roche chemiluminescence immunoassay) whilst on cabergoline 14mg/week. Cabergoline dose was reduced. Further investigations were considered to see if there were any foci of residual active tissue and if definitive treatment, such as surgery or proton-beam-therapy (PBT) appropriate.

She underwent C-11 Methionine PET/CT scan November-2021 after stopping cabergoline for 1 month: this revealed only physiological uptake, with no focal tracer uptake at site of the suspected remnant. Surprisingly prolactin level was normal in the Siemens assay: 357mIU/L(RR <619mU/L), whilst 752mU/L(RR 102-496mU/L)

with the Roche assay. She remained off cabergoline and repeat C-11 methionine scan in December-2022(Figure 2) revealed a small tongue of active tissue in the left side of the sella, congruent with tissue seen on MRI. Prolactin remained discordant (754 mU/L in Siemens assay (1.2×ULN) vs 1806mU/L in Roche assay(3.6×ULN). Macroprolactin test was negative throughout.

Conclusion

Discussion

Treatment of a macroprolactinoma in this young female with dopamine-agonist therapy was thought not to result in normalisation of prolactin. However, as the prolactin level was discordant between the 2 assays, this may not be the case. Discordance has previously been described between Roche and Siemens assays. Questions remain now how to manage this young person long term. As she is hypopituitary already, and no significant change in size of tumour the plan currently is to stay off cabergoline and observe.

Clinical team is faced with following questions in view of future care,

1. What is the reason for higher prolactin in the Roche assay and is this affecting management in other patients?
2. Is there value here for treating small volume residual disease by surgery or PBT if disease progresses?
3. Should management be ‘observe’ and then treat in the usual way with low-dose cabergoline if prolactin increases

Figure 1: Series from MRI Pituitary 2013-2014

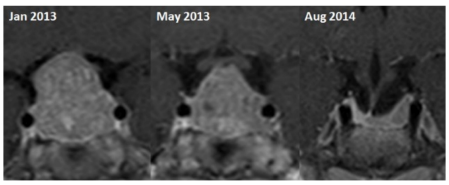


Figure 1.jpg

Figure 2: Co-registration for C-11 Methionine PET/CT 2021 and 2022

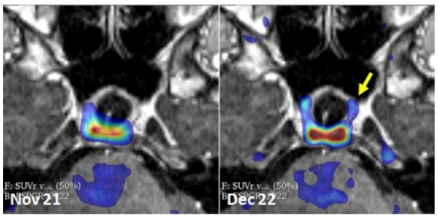


Figure 2.jpg

When plan B works …

Mr. Kevin Anthony Huynh [1], ***Dr. Olympia Koulouri*** [1], ***Dr. James MacFarlane*** [1], ***Dr. Waiel Bashari*** [1], ***Dr. Daniel Scoffings*** [1], ***Dr. Jonathan Jones*** [1], ***Mr. Daniel Gillett*** [1], ***Mr. Göran Darius Hildebrand*** [2], ***Dr. Shaun Wilson*** [2], ***Mr. Jayaratnam Jayamohan*** [2], ***Dr. Fiona Ryan*** [2], ***Prof. Mark Gurnell*** [1]

1. *Cambridge University Hospitals NHS Foundation Trust,* ***2.*** *Oxford University Hospitals NHS Foundation Trust*

Aims

Background

TSH-secreting pituitary adenomas (thyrotropinomas) are rare, accounting for 0.5–2% of all pituitary adenomas. Paediatric thyrotropinomas are even rarer. In most cases surgery is the preferred treatment option following normalisation of thyroid function, typically with somatostatin receptor ligand (SRL) therapy. The latter may also be used following incomplete surgical resection.

Method

Case details

A nine-year-old boy developed progressive visual field loss. Pituitary MRI revealed a 6 cm giant pituitary adenoma compressing the optic chiasm and extending into the right lateral ventricle (Fig 1A). Surgical exploration was limited by bleeding and only a biopsy of the tumour was performed. Histology revealed diffuse staining for TSH. Pituitary function tests demonstrated anterior hypopituitarism, but with the notable exception of the hypothalamic-pituitary-thyroid axis. A subsequent thyrotropin releasing hormone (TRH) stimulation test showed an attenuated response (serum TSH: 0 min, 4.76 mU/L; 20 min, 5.38 mU/L; 60 min, 5.49 mU/L), in keeping with autonomous TSH secretion (Fig. 1C). The parents of the patient expressed a wish to explore treatment options other than further surgery.

Results

Management

An octreotide suppression test (OST: 100 µg SC) showed a partial reduction in serum TSH (Pre: 2.81 mU/L; Post 1.71 mU/L) (Fig. 1D). Sandostatin LAR® 10 mg every 28 days was commenced, and subsequently titrated to 20 mg). SRL therapy proved to be so effective in suppressing TSH that he required levothyroxine supplementation (Fig. 1E). He experienced no significant gastrointestinal side-effects, dysglycaemia, or cholelithiasis. He was also treated with hydrocortisone and growth hormone therapy and in subsequent years testosterone replacement was commenced to induce puberty. Serial surveillance MRIs showed a dramatic reduction in tumour volume (>75%) (Fig. 1B). Repeat visual field testing reassuringly demonstrated reversal of the visual field loss. The patient – currently 18 years of age and eight years on from the original diagnosis – remains well on monthly Sandostatin LAR® injections and complete anterior pituitary hormone replacement.

Conclusion

Conclusions

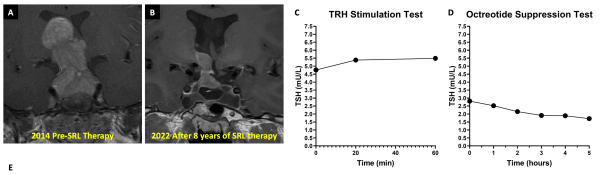
Currently, less than 20 cases of paediatric thyrotropinoma have been described in the literature, with only eight reported to have received treatment with SRL therapy, and the longest duration of medical treatment being 4 years. To our knowledge, this is the first case of long-term primary SRL therapy (now >8 years) for a giant paediatric thyrotropinoma, with dramatic and sustained biochemical, ophthalmological and radiological improvement. This case demonstrates that SRL therapy may be effective in cases where surgery is not preferred or possible.

Questions

1. Is it reasonable to continue long-term, open-ended, SRL therapy? If not, which criteria are recommended to guide a safe trial of medical treatment withdrawal?

2. Should further surgery now be considered, or should radiotherapy or proton beam therapy be advised?

Figure 1



E

	24/3/15	7/4/2015	19/5/2015	4/11/2016	17/1/2017	7/4/2020	23/12/2021
TSH *(0.35 - 5.50 mU/L)*	2.81	0.39	0.29 (L)	0.07 (L)	0.06 (L)	0.04 (L) *(0.3-4.2)*	0.07 (L) *(0.3-4.2)*
Free T4 *(10.0 - 19.0 pmol/L)*	16	8.6 (L)	10.6	16.5	18.0	13.7 *(10.0-17.0)*	12.5 *(10.0-17.0)*
Free T3 *(3.5 – 6.5 pmol/L)*	6.4	3.2 (L)	3.1 (L)	5.2	5.0	- *(3.4-6.9)*	4.9 *(3.4-6.9)*
Medication	Pre-SRL	2 weeks post-Sandostatin LAR 10 mg	Sandostatin LAR 10 mg 4 weekly	Sandostatin LAR 20 mg 4 weekly Levothyroxine 75mcg daily	Sandostatin LAR 20 mg 4 weekly Levothyroxine 75 mcg daily	Sandostatin LAR 20 mg 4 weekly Levothyroxine 100 µg daily	Sandostatin LAR 20 mg 4 weekly Levothyroxine 175 µg daily

Huynh et al - figure - pit cpc march 2023.png

Poster Presentations

Craniopharyngioma: 'Less Is More'

Dr. Desiree Seguna [1], ***Prof. William Martyn Drake*** [1], ***Dr. Zane Jaunmuktane*** [2], ***Dr. Katherine Miszkiel*** [2], ***Mr. Hani Marcus*** [2]

1. *St Bartholomew's Hospital,* ***2.*** *University College London Hospital*

Aims

We report the case of a 19-year-old Lithuanian gentleman who first presented at the age of 12 with failure to thrive and short stature. No organic cause was identified at the time and the patient was discharged on a high-calorie nutrition plan. Aged 16 the patient was seen for the first time at the paediatric endocrinology department, but defaulted subsequent appointments.

One year later, the patient presented with headache, nausea and intractable vomiting. CT brain scan showed a 43x40x27 mm predominantly cystic sellar/suprasellar mass, with peripheral areas of calcifications. Mass effect involving the midbrain and third ventricle was resulting in obstructive hydrocephalus (Figure 1).

The patient underwent emergency endoscopic septum pellucidotomy with cyst biopsy and fenestration (Figure 2). A ventriculo-peritoneal shunt was inserted in view of persistent ventriculomegaly, despite cyst drainage. Histology was in keeping with an adamantinomatous craniopharyngioma.

Method

The patient weighed 41.3 kg (<5th centile) and was 157 cm tall (<5th centile). There was sparse pubic hair and 5 mL testes bilaterally, but no axillary hair (Tanner stage II).

A combined primed insulin tolerance + GnRH test performed by the paediatric endocrinologists 9 months prior to presentation to emergency, showed biochemical evidence of pan-anterior hypopituitarism but without accompanying diabetes insipidus.

Post-operatively the patient was treated for pan-hypopituitarism with hydrocortisone, thyroxine, growth hormone and eventually testosterone replacement. A this stage, the patient was adequately responding to hormonal treatment, growing taller and gaining weight. Vision was normal and there was no hypothalamic or cognitive deficit.

Five months later the patient presented with forgetfulness and difficulty concentrating. CT Brain scan showed recollection of a further suprasellar cyst and an Ommaya reservoir was inserted.

Results

Excision of the residual mass was discussed at the level of the multidisciplinary team. Further surgery was felt to pose too great a threat to vision and hypothalamic function. Subsequently, the patient went on to receive proton beam radiotherapy. Three months following treatment the mass had decreased in size to 42x30x21 mm. The patient expressed wishes for further debulking of the residual calcified solid tumour and 8 months later (against the advice of the multidisciplinary team), underwent frontal craniotomy and debulking in Lithuania (Figure 3). The immediate post-operative course was complicated by the development of a dense homonymous hemianopia, as well as polyuria and polydipsia. The patient was diagnosed with new cranial diabetes insipidus, requiring long-term treatment with DDAVP. The patient also suffered hypothalamic damage in the form of dysregulated appetite, gaining 36 kg over a span of 9 months.

Conclusion

This case highlights the potential hazards of offering 'too much surgery' for craniopharyngioma. The strategic objectives of surgery should be to alleviate pressure-related mass effects whilst preserving vision, hypothalamic, neurological and cognitive function. Although more conservative surgical strategies may result in greater potential for regrowth, carefully planned radiation therapy (proton beam in young patients) substantially re-

duces this risk and is not associated with the same risk of hypothalamic injury. ‘Less is more’.

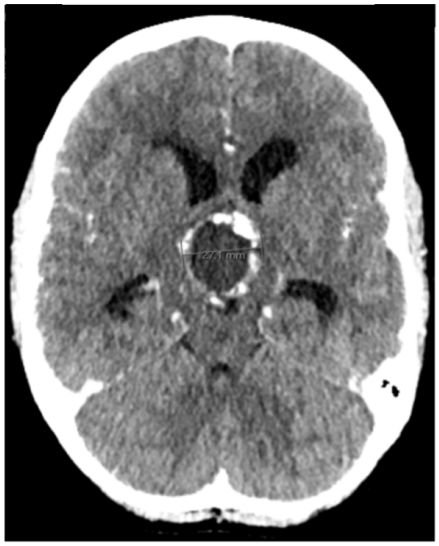


Figure 1. Pre-operative CT Brain scan

Pre-op ct scan.png

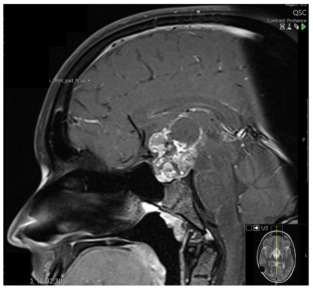


Figure 2. T1-weighted MR Pituitary with contrast (6 weeks post-op)

Mr pit 6 weeks post-op.png

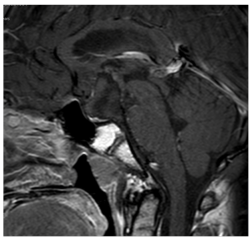


Figure 3. T1-weighted MR Pituitary with contrast 9 months following debulking operation

Mr pit post-craniotomy.png

Expansion of Left Cavernous Sinus Meningioma secondary to pregnancy

Dr. Yuvanaa Subramaniam [1], ***Dr. John V Anderson*** [2], ***Dr. Jane Evanson*** [3], ***Dr. Nick Plowman*** [4], ***Prof. William M Drake*** [5]

1. *1. Department of Endocrinology, St Bartholomew's Hospital,* ***2.*** *4. Department of Endocrinology, Homerton University Hospital,* ***3.*** *3. Department of Radiology, St Bartholomew's Hospital,* ***4.*** *2. Department of Oncology, St Bartholomew's Hospital,* ***5.*** *1.Department of Endocrinology, St Bartholomew's Hospital*

Aims

We present a 39-year-old lady, who was fit and well with no past medical history and was on no regular medications.

She developed left-sided headache, left ocular pain and diplopia on left lateral gaze during the 3rd trimester (39/40) of her second pregnancy. Her first pregnancy was conceived without difficulty. Her 4-year-old son was well with no medical issues. She had no symptoms to suggest a primary pituitary disease.

Clinical examination revealed a left 6th nerve palsy, but was otherwise unremarkable. She was clinically eupituitary with normal biochemical thyroid function tests.

MRI head showed a 22.5mm lesion centred predominantly in the left cavernous sinus. The lesion had high T2 signal and intermediate T1 signal. Medially, the lesion extended into the pituitary fossa partially displacing the pituitary gland. The internal carotid artery was patent but displaced anteriorly. Radiologically, the lesion appeared to be a meningioma (Figure 1).

Method

She chose to be induced, rather than having a caesarean section and delivered a baby girl 48 hours after presentation. The second stage of labour was swift and there were no untoward consequences of her pushing.

Following the birth of her daughter, her diplopia, and visual symptoms significantly improved. She had mild 6th nerve palsy with diplopia on extreme left lateral gaze. Her baby was well and was largely breastfed.

Her follow-up imaging two-months post pregnancy showed a less bulky lesion with an expected physiological reduction in the volume of the pituitary.

Our presumed diagnosis at this stage was that oestrogen receptors on the meningioma were stimulated during her pregnancy leading to expansion of the lesion, in turn causing headaches and diplopia.

Results

She was advised against taking the combined oral contraceptive pill, and refrain from further pregnancy until she had received definitive treatment. A repeat MRI pituitary a year post-partum showed a slight enlargement of the meningioma from 20mm to 23mm.

Following this, she was referred for stereotactic radiotherapy (gamma knife treatment (15Gy) achieving 95% coverage of a 6.12cc lesion 14 months after her initial presentation. This led to a substantial reduction with the tumour size (14mm on her repeat MR brain done 3 months following treatment) (Figure 2). At that stage, she expressed wishes to have her third pregnancy in 2019.

She conceived spontaneously post-IUCD removal, and her 3rd baby was delivered in 2021. There was no untoward pituitary, neurosurgical or neurological events through the pregnancy.

Her repeat MRI pituitary in 2022 showed a stable appearance of the (still presumed) left cavernous sinus meningioma.

Conclusion

There are reports describing the effects of pregnancy on intracranial meningioma. The aggravation of signs

and symptoms may be related to fluid retention, vascular engorgement and increased oedema or the presence of hormone receptors leading to a dramatic growth during pregnancy. As our patient presented at term, the decision to deliver her baby was straightforward. The advice to avoid oestrogen-containing contraception was empirical, given the presumed mechanism of VI nerve compression and the radiosurgery facilitated sufficient volumetric reduction to facilitate an uneventful subsequent pregnancy.

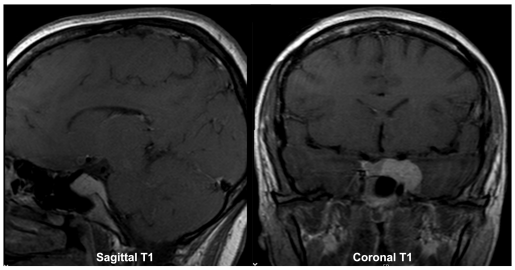


Figure 1.jpg

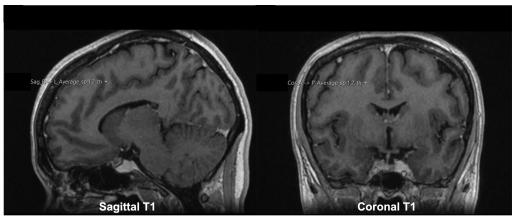


Figure 2.jpg

Gigantism – Genetics, Localisation Techniques and Management Options in Persistent Residual Active Disease

Dr. Amy Morrison [1], ***Dr. Narendra L Reddy*** [1], ***Dr. Ian Scott*** [2], ***Mr. Ian Robertson*** [2], ***Dr. Miles J. Levy*** [1]

1. *Department of Endocrinology, University Hospitals of Leicester,* ***2.*** *University Hospitals of Nottingham*

Aims

We present a case of Gigantism, in a young man initially diagnosed secondary to hearing loss and cardiac impairment. Management has been challenging, involving recurrent surgery, in addition to medical management. Despite which this patient has persistent residual active disease.

We aim to discuss investigations to date, specifically imaging methodologies utilised in this patient's management. Questions remain surrounding optimal targeted management going forwards, in order to achieve definitive cure of Growth Hormone (GH) excess.

This case additionally raises discussion points surrounding potential underlying genetic aetiology of Gigantism and the appropriate avenues to explore contributory somatic and germline mutations.

Method

A 33 year old professional basketball player presented to ENT clinic with unilateral hearing loss. CT Petrous bones revealed an enlarged pituitary gland. Clinical evidence of gigantism was evident; 2.32m(7ft 7), macroglossia and enlarged hands and feet. Cardiac impairment was apparent, with reduced left ventricular ejection fraction(EF 31-35%) and multifocal premature ventricular complexes resulting in palpitations. Management was commenced with Bisoprolol and Amiodarone.

Initial investigation revealed an elevated GH (34 ug/L) and IGF-1 (>1000 ug/L), which failed to supress with OGTT (remained GH>40ug/L). MRI Pituitary highlighted a 23mm pituitary macroadenoma, with possible extension into the left cavernous sinus but no suprasellar extension. Plans for surgery were delayed due to availability of an appropriately sized operating table and COVID-19 pandemic.

Pre-operative Somatostatin analogue therapy was commenced (Octreotide 30mg) and Hydrocortisone(20/10/10mg) following suboptimal Short Synacthen Test(55 to 441nmol/L). Carbimazole was additionally required in management of Amiodarone induced thyrotoxicosis(TSH<0.05, FT4 99, FT3 >30).

Results

Transphenoidal hypophysectomy surgery took place in December 2020, with histology revealing a densely staining somatotroph tumour, with PIT1 apparent in almost all cells. One week post-operative; GH 4.8, IGF-1>1000. Following this, this gentleman travelled to New Zealand for work, with ongoing Endocrinology care coordinated there. An MRI in New Zealand revealed residual disease. On his return to the UK, Methionine PET in Cambridge highlighted a left posterior lesion within the sella, adjacent but not within the cavernous sinus in line with repeat OGTT in August 2021; GH 6.7 ug/L.

A second transphenoidal hypophysectomy took place in February 2022, debulking occurred with histology revealing mixed PIT-1 positive mammo-somatotroph densely granulated adenoma with slightly elevated p53. Immediately post-operative GH (8) and IGF-1 (853) levels remained elevated. OGTT six weeks post-operatively; GH 5.9 ug/L, was consistent with persistent residual disease.

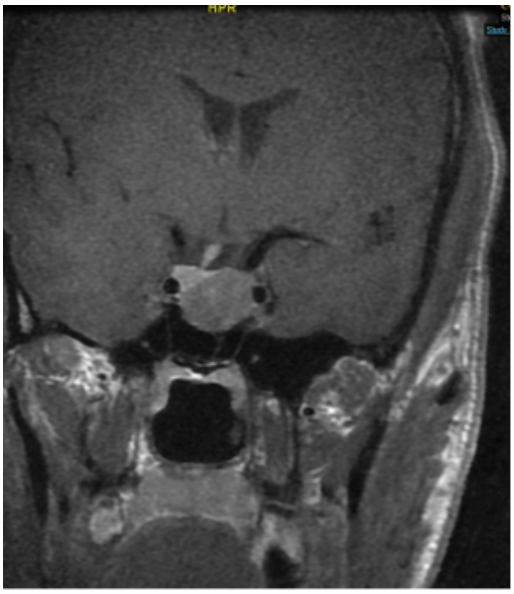
Conclusion

This gentleman has active GH excess despite two attempts at definitive surgical management.

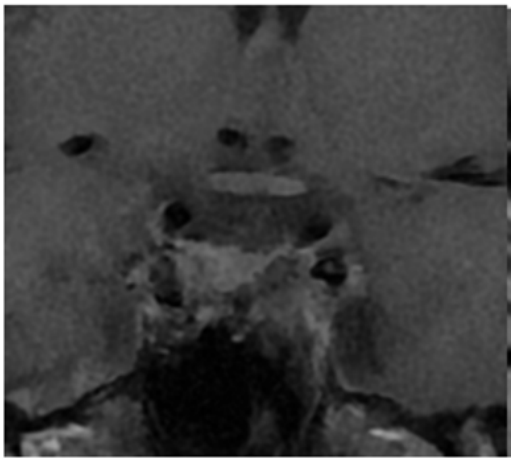
Management options now available include: 1) further surgery, 2) medical therapy 3) Fractionated or stereotactic radiotherapy

A novel method of incorporating gonadotroph and ACTH suppression (with GnRH and Dexamethasone respectively), between interval Methionine PET/MR scans 28days apart, in order to maximise the somatotroph signal,

as part of the Cambridge OPTIMUM trial is planned.
Genetic investigation is yet to take place, as this gentleman has not wished to proceed with this to date. He has no known relevant family history but discussion regarding familial genetic counselling and screening in needed, given that he is planning children, as well as predict the behaviour of his own disease.
We hope this case allows a good discussion, and for the expert panel and wider audience to aid ongoing management of this case from an endocrine, genetic, radiological, surgical and radiotherapy perspective.



Initialmripituitary.png



Postoperativex2mrimay22.png

Timepoint (minutes)	Glucose (mmol/L)	Growth Hormone (ug/L)
0	5.3	>40
30	5.3	>40
60	6.9	>40
90	4.1	>40
120	4.8	>40
150	5.4	>40
180	5.5	>40

Initialogttresults.png

Hey doc have you seen such a big thing before

Dr. Muhammad Iftikhar [1], ***Mr. Hani Marcus*** [2], ***Dr. Katherine Miszkiel*** [2], ***Dr. Helen Simpson*** [2]

1. *UCLh,* ***2.*** *University College London Hospital*

Aims

INTRODUCTION:

Large skull base prolactinomas are rare, and diagnosis could easily be delayed by considering skull base tumours as tumours of non-pituitary origin. Early recognition and timely medical management can prevent neurological complications.

Method

PRESENTATION.

We present a case of 17 year old male who presented to his local emergency department with history of headache for 18 months, initially treated as migraine in community. He had progressive left sided central field loss for 1 month, diagnosed as optic neuritis and was on methylprednisolone, and a one-week history of increasing ataxia and drowsiness. He did not report any issues with energy, growth, weight, pubertal development, erections.

On examination he had right eye squint, pupil dilatation, diplopia, ptosis, and right-sided tongue deviation along with dysarthria suggestive of 3rd, 4th, 6th and 12th nerve palsy.

Results

INVESTIGATIONS:

Prolactin 195,000, Cortisol 811, Testosterone 0.6, FSH 1.2, LH 1, T4 11.3, TSH 0.72, GH, and IGF1 97.

CT head and subsequent MRI head reported a 6.4x4.6 cm extensive intra-, supra-, and para-sellar lesion causing compression and upward displacement of prechiasmatic optic nerves (R>L) and optic chiasma. There was also extension to the cavernous sinus, temporal lobes and posterior fossa, resulting in brainstem compression.

DIAGNOSIS AND MANAGEMENT:

He was started on cabergoline 1mg/day. Neuro-ophthalmology examination confirmed complete bilateral 6^{th} nerve palsies, partial right 3^{rd} nerve palsy, pupil involvement and right 4^{th} nerve involvement.

PROGRESS.

A repeat MRI pituitary after 1 week showed shrinkage of tumour and hence decision was made to continue with high dose of Cabergoline and to repeat MRI at 3 months interval. He was started on testogel in the usual way. Prolactin came down to 40,000 at 3 months. Symptoms of ataxia, diplopia dysarthria also improved.

Conclusion

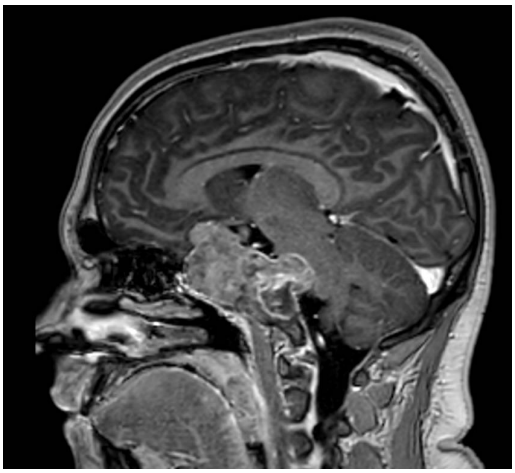
Follow up:

We regularly followed up this case in endocrine clinic, including further MRI in 4 months' and subsequently 6 months' time. MDT discussion made after each scan and treatment deemed working successfully, it lead to resolution of symptoms and signs, reduction in prolactin levels, and improvement in radiological features.

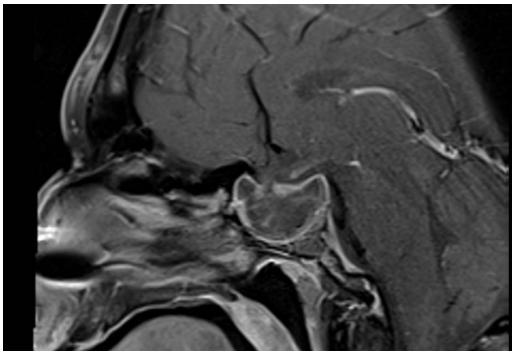
He is currently continuing with cabergoline 500ug/day and latest prolactin is 5300. (Peak 195000). He is now on a gap year after doing his A levels, planning to join drama college.

DISCUSSION:

Skull base giant prolactinomas are rare presentation accounting for 2-3 % of all prolactinomas, they are challenge to diagnose and intervene early due to uncommon presentation. Rapid progressive nature can lead to cavernous sinus, temporal lobe and posterior fossa invasion, with corresponding mass effect. Markedly raised serum prolactin with these tumors should prompt medical treatment with dopamine agonists because of good prognosis and excellent serological and clinical response.



Before.png



After.png

Pituitary Metastasis from a renal cell carcinoma: primum non nocere

Dr. Jessica Lee [1], ***Dr. Thomas Powles*** [1], ***Dr. Thomas Campion*** [1], ***Dr. Katherine Miszkiel*** [2], ***Prof. William M Drake*** [1], ***Mr. Hani Marcus*** [2]

1. *St Bartholomew's Hospital,* ***2.*** *University College London Hospital*

Aims

A 69 year old man with metastatic renal cell carcinoma was referred for endocrine evaluation because of worsening lethargy and a random serum cortisol of 59nmol/L. His most recent cross-sectional imaging had shown an increase in the size of known mediastinal lymphadenopathy, worsening pulmonary metastasis and an enlarging (now 5cm) right adrenal nodule despite therapy with sunitinib and nivolumab. Further endocrine evaluation revealed hyperprolactinaemia (Prolactin=5744; Reference: 0-323mU/L) secondary hypothyroidism (TSH=1.51,FT4=7.2; Reference: TSH=0.27-4.2pmol/L, FT4=10.5-24.5pmol/L) and hypogonadism (LH<1, FSH<1, Testosterone<0.5; Reference: LH=1.7-8.6 unit/L, FSH=1.5-12.4 unit/L, Testosterone=6.7-25.7 nmol/L). Hydrocortisone and, later, thyroxine replacement were commenced with good clinical effect. Urgent pituitary imaging was arranged but whilst waiting he reported deteriorating vision and rapidly became unable to work, drive and subsequently read.

Method

MRI pituitary (Figure 1) identified a suprasellar lesion which had a large superior component which was indenting the optic chiasm. The lesion was suspicious for a pituitary metastasis and had characteristics suggestive of intra-lesional haemorrhage. The MRI also revealed metastatic deposits in both cerebellar hemispheres. In addition, the patient reported new symptoms consistent with diabetes insipidus and was started on desmopressin at night with resolution of nocturia. A combination of the vascular nature of the tumour, its suprasellar anatomical conformation, and the presence of multiple brain metastases dictated that, on balance, endonasal surgery was not recommended. The patient was started on cabazantinib as a palliative measure.

Results

Although metastasis of renal cell carcinoma to the pituitary gland is rare, it presents particular challenges. Pituitary hormone deficits can be easily replaced, but in this case the vascular nature of the tumour and its unfavourable anatomy dictated that, after careful consideration, pituitary surgery was not advised/offered.

Conclusion

We present this case to encourage exchange of experience– both surgical and medical – of this and other, related scenarios in which the desire to offer sight-preserving treatment within a palliative setting must be balanced against the maxim 'primum non nocere'.

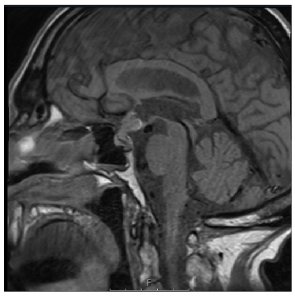


Figure: pre contrast T1 Sagittal: sellar lesion with suprasellar extension

Renal pit image-page0001.jpg

Sellar collision tumour – a rare entity presenting with rapidly progressive visual deficits

Mx. Adrian Zammit [1], ***Mx. Christina Daousi*** [2], ***Mx. Piyali Pal*** [3], ***Mx. Nitika Rathi*** [3], ***Mx. Frederico Roncaroli*** [4], ***Ms. Catherine Gilkes*** [4]

1. Department of Neurosurgery, The Walton Centre NHS Foundation Trust, Liverpool, 2. Department of Endocrinology, Liverpool University Hospital NHS Foundation Trust, Liverpool, 3. Department of Neuropathology, The Walton Centre NHS Foundation Trust, Liverpool, 4. Department of Cellular Pathology, Salford Royal NHS Foundation Trust, Manchester

Aims

Tumour-to-tumour metastases involving metastatic disease spread into a pre-existing pituitary adenoma (PitNET), also known as sellar collision tumours, are a rare entity. We present a case of metastatic high-grade neuroendocrine (small cell) tumour from a likely radiologically-occult lung primary into an endocrinologically-silent pituitary adenoma (PitNET).

Method

The patient was identified after discussion at the pituitary MDT. A retrospective review of the patient's medical records, including diagnostic imaging, laboratory results, and treatment history was performed.

Results

74-year-old lady presented with rapid progressive bilateral ocular motor nerve palsies, decrease in visual acuity and anterior hypopituitarism. Serum prolactin was 1,698mIU/L. An MRI revealed an extensive tumour centred on the sella turcica invading both cavernous sinuses. A retrospective review of an MRI performed years prior to presentation identified an unreported smaller pituitary adenoma. A CT chest-abdomen-pelvis was normal.
She was offered an endoscopic transsphenoidal excision of the lesion. The tumour was associated with significant bony involvement, displaying both necrotic and fibrotic areas. Post-operatively the patient experienced an improvement in the cranial neuropathies and visual acuity.
Histology was initially interpreted as a poorly differentiated pituitary carcinoma. Further review revealed two separate cell populations, one with uniform cells with rounded hyperchromatic nuclei lacking mitotic figures consistent with pituitary adenoma (PitNET) and another with larger, atypical cells which was highly mitotically active suggestive of a metastatic high-grade neuroendocrine (small cell) tumour deposit.

Conclusion

Pre-operative differentiation between pituitary adenoma (PitNET) and collision tumour on the basis of radiological and clinical characteristics is difficult. The presence of rapidly evolving neurological deficits could be representative of an aggressive pituitary carcinoma or collision tumours with invasive metastatic disease. Histological diagnosis presents its own challenges. This case report also clearly describes the pathogenesis of collision tumour with metastatic seeding into a pre-existing pituitary adenoma.

Severe hyperprolactinaemia associated with domperidone with normal MRI pituitary

Dr. Kerrie Thackray [1], ***Dr. Felicity Kaplan*** [1]

1. *East and North Herts NHS Trust*

Aims

Domperidone, a peripheral dopamine antagonist, is widely used as a treatment for nausea and vomiting. A well recognised side effect is mild hyperprolactinaemia in up to 50% of patients, although only 1% of patients will experience associated clinical endocrinological side effects.

Prolactin levels in females are generally <500 mU/L with drug induced hyperprolactinaemia usually associated with only a modest elevation in prolactin of <2000 mU/L. Prolactin levels above this may indicate a possible pituitary adenoma.

We present a case of a patient in whom short term administration of domperidone was associated with much more significant hyperprolactinaemia than would normally be expected with domperidone alone, with complete normalisation of prolactin on discontinuation of the drug. MRI pituitary was unremarkable. The extent of hyperprolactinaemia may have been exacerbated by co-prescription of a proton pump inhibitor and by ongoing use of marijuana.

Method

A 28 year old female initially presented to gastroenterology with a six year history of vomiting, dyspepsia and weight loss. Past medical history included mild hirsutism with previously normal biochemistry, surgery for a 14 cm ovarian dermoid cyst ten years prior, and anxiety/depression. Repeat medication included esomeprazole and mebeverine. Domperidone 10mg three times daily had been started in the community 20 days prior to specialist review. The patient was not using hormonal contraception and was not pregnant or lactating. She smoked marijuana four times weekly. Examination was unremarkable and she had no new signs or symptoms of hyperprolactinaemia or hormone excess. Bloods were within normal limits except for prolactin of 3833 mU/L. Macroprolactin studies confirmed true hyperprolactinaemia. Following endocrine review, domperidone was discontinued. Subsequent prolactin levels were 211 mU/L and 151 mU/L demonstrating complete normalisation. MRI pituitary demonstrated a normal pituitary gland.

Results

Domperidone is a peripherally selective D2 receptor antagonist acting at the lactotrophs of the anterior pituitary to reduce intracellular cyclic AMP thereby increasing prolactin production. It is generally thought to cause only mild hyperprolactinaemia. Previous work by Bouwers et al demonstrated that a single dose of domperidone might increase prolactin to 157-2638 mU/L with sustained hyperprolactinaemia at a lower level following two weeks of treatment.

Variants of the dopamine D_2 receptor gene in patients on risperidone may be associated with an exaggerated hyperprolactinemic effect, so it is reasonable that this also might be seen with domperidone.

Esomeprazole has been suggested as a cause of hyperprolactinaemia in several case reports, potentially by inhibition of CYP3A4 thereby elevating oestrogen levels. Marijuana has been similarly implicated in case reports although literature is mixed with reports of decreased, increased and unchanged prolactin associated with cannabinoids.

Conclusion

Domperidone is usually associated with mild hyperprolactinaemia but, in this case, was associated with severe

hyperprolactinaemia, although without clinical signs or symptoms, following 20 days of treatment for nausea and vomiting. This may have been exacerbated by co-administration with long-term esomeprazole and/or by cannabinoid use by the patient. In this case, prolactin level normalised with discontinuation of domperidone with no other changes to prescribed or self-administered drugs.

While severe hyperprolactinaemia is well-recognised as a side effect of metoclopramide, risperidone and phenothiazines, we demonstrate that domperidone may also be a causative factor in the absence of primary pituitary pathology.

Spontaneous resolution of pituitary tumour in a post-partum woman - Would you recognise it?

Dr. Bashir Mahamud [1], ***Dr. Belayet Hossain*** [1], ***Dr. Shaan Sahota*** [1], ***Dr. Lydia Sinclair*** [1], ***Mr. Ayodeji Oyesanya*** [1], ***Dr. Mohammed Fathalrahman*** [1], ***Dr. Gideon Mlawa*** [1]

1. *Barking, Havering and Redbridge University Trust*

Aims

1. To present an interesting case of spontaneous resolution of pituitary macroadenoma in a post-partum patient. The patient was reviewed by the neurosurgical team and was scheduled for surgical intervention. On the subsequent follow up her pituitary macroadenoma had resolved and no surgical intervention was necessary.
2. To allow attending delegates to discuss this case and share their experience in managing cases with similar presentation.

Resolution of the pituitary tumour without surgical intervention is rare. The mechanism of spontaneous resolution of pituitary adenoma is unclear but may be explained by either pituitary apoplexy or hypophysitis responding to steroids. Bray BP, et (2021) reported the case of vanishing pituitary mass due to pituitary apoplexy. Park SM, et al (2014) reported 22 cases of pituitary mass due to hypophysitis which improved mostly with medical therapy.

Method

The causes of non-functioning tumour include neoplastic, inflammatory and vascular. Non-functioning pituitary macroadenoma are usually managed surgically if patients are symptomatic, unless contra-indicated.

Case Report

A 29-year-old lady presented to the hospital in January 2014 with vomiting, abdominal pain, headaches and profound hyponatraemia of 112. She had undetectable LH (<0.2), FSH (1), SST normal, and Prolactin 407. IGF-1 was normal. She was treated with IV fluids. She had an MRI pituitary which showed homogenous enhancing pituitary macroadenoma measuring (1.0x1.5x1.8 cm). She was 1 year postpartum. The second baby was born in 2012 and stopped breastfeeding in October 2013. Her periods were regular.

Results

She was seen initially by neurosurgeons for surgical intervention. She was started on hydrocortisone 10mg then reduced to 5mg twice a day then stopped in July 2014 at 9 am cortisol was normal at 300.

A repeat MRI after 10 months reported a normal appearance of the pituitary gland and surgery was not required.

On the subsequent follow up her pituitary macroadenoma had resolved and no surgical intervention was necessary. She remains stable and asymptomatic.

Conclusion

Spontaneous resolution of pituitary tumours is rare. The mechanism of resolution of pituitary adenoma is unclear but may be explained by either pituitary apoplexy or hypophysitis responding to steroids.

The time of onset of spontaneous regression of pituitary adenoma following pituitary apoplexy is unclear, some cases were previously reported to be as early as 1 week.

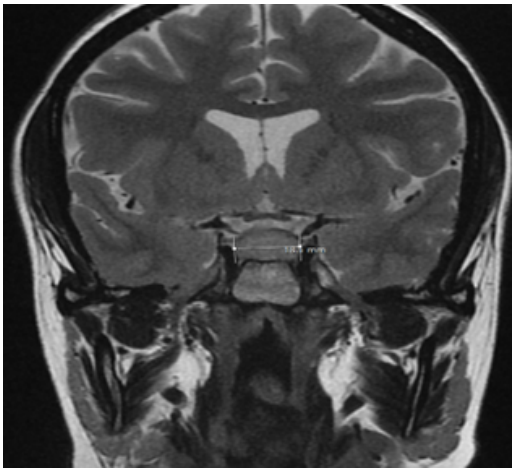
Hypophysitis is an inflammatory process which may respond to steroids. It can be postulated that giving

steroids facilitates the regression of pituitary adenoma.

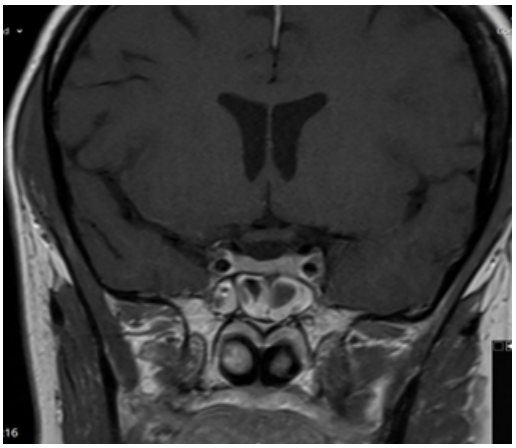
Spontaneous regression of pituitary adenoma is uncommon and possible causes discussed above should be considered. It is unclear whether patients with spontaneous regression of pituitary adenoma are at risk of recurrence and whether they need long-term follow-up. The case raises the question of how long non-function pituitary adenomas can be monitored before intervening surgically.

Question to the Panel

How would you manage this patient? How often, how long do you monitor?



Picture1 2023.png



Picture2 2023.png

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