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CASE REPORT:

CO-EXISTENCE OF GRAVES' DISEASE, BRACHIAL ARTERY PSEUDOANEURYSM AND RADIAL NEUROPATHY IN AN ADOLESCENT GIRL

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ABSTRACT:

Brachial artery aneurysm is rare but could be potentially both life-threatening and limb-threatening. It may lead to peripheral nerve injuries which cause disability and affect everyday function, both domestic and economic. We present a rare case of pre-menarcheal adolescent girl with Graves' disease co-existing with radial neuropathy due to radial nerve compression by a giant brachial artery pseudoaneurysm. The patient was managed surgically with aneurysmal resection and saphenous vein graft interpositioning. Prompt diagnosis and institution of appropriate surgical repair to prevent adverse outcome was imperative.

Keywords: Adolescence, Graves' disease, pseudoaneurysm, radial neuropathy, wrist drop

Running title: Co-existence of Graves' disease, brachial artery pseudoaneurysm and radial neuropathy

INTRODUCTION:

Hyperthyroidism due to Graves' disease is a common endocrine disorder in adolescence with a female preponderance. Often, it is treated with oral anti-thyroid medications such as carbimazole (or its active metabolite methimazole). The usual side effects of this medication are allergic-skin reactions and agranulocytosis but antineutrophil cytoplasmic

antibody (ANCA) vasculitis is a rare side effect [1]. In 2008, Mehindratta et al, [2] reported the first case of ANCA positive vasculitis complicating carbimazole therapy in a 20-year old Indian man. The risk of carbimazole-induced ANCA positive vasculitis is higher in females than males [1].

Pseudoaneurysm is a collection of blood formed as a result of vascular injury and

retained in tissues surrounding the breached vessel. The circulating blood is contained in a cavity surrounded by adjacent tissues, fascia and thrombus but not by normal arterial wall components [3]. Although most pseudoaneurysms result from penetrating injury, minor blunt trauma may cause pseudoaneurysm in individuals who are prone to haemorrhage [4,5]. Besides trauma, infection, polyarteritis nodosa and congenital arterial defects are other known causes of upper extremity pseudoaneurysm [6]. It can present as a new thrill or bruit, pulsatile haematoma, or marked pain or tenderness. A giant pseudoaneurysm can cause peripheral venous oedema and may compress the adjacent neurologic structures, causing sensory and motor deficits [7]. The first symptoms of upper extremity aneurysms can be nerve injury or adjacent nerve compression [8]. The potential associated complications of upper extremity pseudoaneurysm are local pain, local skin ischaemia, rupture and distal embolization with loss of hand and fingers [9,10]. Thus, early diagnosis and prompt surgical repair are essential to avoid complications.

Peripheral nerve injuries are of great economic importance because of their effect on quality of life. The radial nerve is one of the most frequently injured peripheral nerves because of its close anatomical relationship with the humerus. The radial nerve is composed of

branches of the C5 through T1 nerve roots and arises from the posterior chord of the brachial plexus within the axilla. Emerging from the axilla and in the arm, the radial nerve gives off three cutaneous sensory branches, namely posterior cutaneous and lower lateral cutaneous nerves of the arm. The third, posterior cutaneous nerve of forearm is responsible for sensation over a large area of the forearm [11]. Thereafter the radial nerve runs over the dorsal surface of the humerus in the spiral groove, supplying the extensor muscles of the forearm and fingers. Proximal to the elbow, it bifurcates into a sensory superficial radial nerve (SRN) and a motor posterior interosseous nerve (PIN) branch [11,12]. The sensory component (SRN) travels in the forearm over the radial bone, supplying most of the dorsal surface of the hand [11].

As a rule, pattern of clinical involvement is dependent on level of injury [11]. A radial nerve injury above the elbow may present with extensor weakness of the elbow, wrist and finger accompanied by sensory disturbance along PIN and SRN distribution [13,14]. Sensory loss over the posterior arm, forearm and the posterior lateral hand and thumb indicate a radial nerve lesion above the spiral groove [15].

Wrist drop is the most common presentation of radial nerve palsy which can occur because of external compression secondary to a giant pseudoaneurysm [7].

CASE REPORT:

We report a case of a 15-year-old pre-menarcheal Nigerian girl who presented with anterior neck swelling of 4 years duration, protrusion of both eyes of 3 years duration, painful swelling of left arm of one week duration. The size of neck swelling progressively increased but it is not associated with pain. There is no family history of thyroid disorder. About one year after onset of neck swelling, the eyeballs were noticed to be more prominent, revealing an increased area of the sclera. No history of eye discharge or redness. One week before presentation the patient developed painful progressive swelling of the left arm and this was followed three days later by loss of sensation and paralysis of the forearm. This prompted parents to seek medical help. One year ago, she was commenced on carbimazole because of the anterior neck swelling. Her appetite has remained good. She is the youngest of 3 surviving children; two younger siblings died from unknown cause. Her family was displaced from Borno State, Northeast Nigeria to internally-displaced-person's camp in Edo State as a result of the Boko Haram terrorist insurgency. She denied any history of trauma to the left arm, even while they were fleeing from the terrorist group who invaded their homes five months earlier. She was in class 2 in Junior Secondary School with good academic performance. The whereabouts of

her father is currently unknown. At present, the mother has no source of livelihood. Permission to report this case was obtained from the patient and her mother.

On examination she was afebrile (37.4°C), not pale or jaundiced and there was no peripheral lymphadenopathy. No vasculitic rash was seen. Anthropometric measurements showed weight 37kg (5th percentile), height 149cm (5th percentile) and body mass index 16.8kg/m² (10th percentile). Her sexual maturity rating was Tanner stage 2 [16]. She had bilateral exophthalmos with uniform swelling in the anterior triangle of the neck. The neck swelling moves with deglutition, smooth, non-tender and no bruit. Examination of the left upper limb revealed uniform swelling of the forearm with wrist drop and weakness at the elbow joint. Sensory examination of the radial nerve with pin prick and light touch testing of the posterior arm, forearm, posterior lateral hand and thumb revealed sensory loss in these areas. Figure 1 shows the left radial wrist drop with burns in the left wrist region (burns due to contact with hot pot as a result of sensory loss). She had a resting right radial pulse rate of 120 beats/minute (tachycardia), regular, full volume. The left radial pulse volume was small. Her blood pressure was elevated (150/100mmHg) with hyperactive precordium. The apex beat was in the 5th left intercostal space (LICS), mid-clavicular line (MCL). The heart sounds were normal. Examination of the other body systems was unremarkable. The peripheral blood film

showed anisocytosis, microcytosis, severe hypochromasia, neutrophilic hypersegmentation with toxic granulations and thrombocytosis. Complete blood count showed a total white cell count of $22.0 \times 10^3/\mu\text{l}$, lymphocytes 8.9%, granulocytes 84.1%, Haematocrit 39.3%, platelets $324 \times 10^3/\mu\text{l}$. Thyroid scan showed diffuse enlargement of thyroid lobes, multiple anechoic lesions within

both lobes (worse in the right) and mild displacement of vascular structures. Serum urea and electrolyte results showed no abnormality. The electrocardiogram showed sinus tachycardia and the echocardiogram revealed a structurally normal heart. The thyroid function tests (TFT) results are summarized in Table 1.

Table 1: Summary of thyroid function test results

Laboratory parameter	Results	Comments
Total serum T3	10ng/ml	High
Total serum T4	247mmol/L	Very high
TSH	0.05 $\mu\text{U}/\text{ml}$	Very low

The initial diagnostic consideration was Graves' disease with cellulitis of the left arm. The patient was commenced on IV Amoxicillin-Clavulanic acid, Gentamicin and oral Labetalol and Propranolol with continuation of Carbamazepine. The paediatric endocrinology team reviewed the patient and considered Multiple Endocrine Neoplasia (MEN) type 2A with metastasis to left arm. The investigations requested for were serum calcium, phosphate, parathyroid hormone (PTH), free para-metaneprine level, magnetic resonance imaging (MRI) of adrenal glands and fine needle aspiration of thyroid gland. These investigations could not be carried out because of financial constraints. The result of serum calcium concentration

obtained later was normal. By the 6th day on admission, the swelling and pain in the left arm worsened and ultrasound scan of the left upper limb was requested. The scan revealed a complex mass at the medial aspect of left arm measuring 4.52 x 4.51cm with both solid and cystic components. Cystic components appear vascular but dilated in the proximal part tapering into a tubular structure in the distal part. Within the cystic component, there was a high turbulence with an associated extensive soft tissue swelling in this region. The diagnosis was modified to Graves' disease co-existing with left brachial artery pseudoaneurysm. At this point, the Cardiothoracic surgical team was invited and a Doppler ultrasound was

performed. The Doppler ultrasound findings were as follows: (i) Aneurysm measuring 32 x 44.9 x 40mm located at the proximal part of the left brachial artery with its thinnest wall thickness of 4.70mm; (ii) Organized thrombus crescentic in shape measuring about 37.2 x 18.4mm with 54.7mm- luminal patency; (iii) Thrombus is at the distal end of the aneurysm and occludes the draining brachial artery; and (iv) Multiple collateral channels are seen which re-enters the draining brachial artery increased peak systolic velocity of brachial artery of about 82.8cm/s. Having confirmed the diagnosis, aneurysmal resection together with salphenous vein graft interpositioning was performed. At surgery, the following were found: (i) Huge

false aneurysm of the proximal left brachial artery, filled with clots, necrotic tissue and pus; (ii) aneurysmal sac measuring about 10x 8cm (iv) 16cm gap between both ends of the artery;(iv) surrounding soft tissue oedema and cellulitis (Figure 2).

At surgery, the great saphenous vein of the left leg was harvested up to the knee, reversed and anastomosed to the proximal and distal ends of the brachial artery. Visible and palpable arterial pulsations were noted post repair. She was transfused with two pints of blood (One pint intra- and post-operatively, respectively).The surgical wound healed satisfactorily but sensory loss was still present at discharge. She is being followed up in the outpatient clinics.



Fig. 1: A 15-year-old girl with left wrist drop and burns (arrow) at the left wrist region.



Fig. 2: Exploration of the pseudoaneurysmal sac during surgery

DISCUSSION:

In the index case, the cause of the brachial artery pseudoaneurysm is not clear. Although most pseudoaneurysms result from penetrating injury, minor blunt trauma may cause pseudoaneurysm in individuals who are prone to haemorrhage [4,5]. In our patient, history of trauma was negative. Is it possible that the patient may have sustained a minor blunt trauma without noticing it? Alternatively, could the index case have carbimazole-induced antineutrophil cytoplasmic antibody positive vasculitis which is known to be more frequent in females [1]. Our patient is a female. We could not investigate for antineutrophil cytoplasmic antibody (ANCA) because of inadequate laboratory facilities in our centre.

With regard to the radial nerve injury, the pattern of clinical involvement is dependent on level of injury [11]. The index case had extensor weakness of the elbow, wrist and fingers, suggesting radial nerve injury above the elbow [13,14]. In addition, sensory loss over the posterior arm, forearm and the posterior lateral hand and thumb were present. This distribution of sensory loss indicates a radial nerve lesion above the spiral groove [15].

At surgery, the pseudoaneurysmal sac was found to be located at the proximal part of the left brachial artery, in keeping with the pattern of clinical involvement in our patient. Wrist drop is the most common presentation of radial nerve palsy which can occur because of

external compression secondary to a giant pseudoaneurysm [7] as is the case in our patient. Evaluation of the patient at one of her follow-up visits (12 weeks post repair), revealed that the patient was just beginning to regain ability to extend interphalangeal joints. The sensory loss was still present, accounting for the burns she sustained at home from contact with hot pot. These findings suggest a slow regain of function. In most cases of compressive radial neuropathy, the type of injury is a neuropraxia which does not involve damage to the axon [15].

Neuropraxia is classified as a transient conduction block of motor or sensory function without neuronal degeneration [15]. Within this context, radial nerve injury caused by brachial artery pseudoaneurysm is expected to be a neuropraxia. As such, regain of function is expected within a few weeks after surgical repair [15].

The reason for the delay in regain of function in our patient is not clear. The presence of an infected pseudoaneurysmal sac in our patient may have contributed to the slow regain of function by altering the usual nature of the nerve injury and subsequently, the process of regain of function. An alternative explanation is that peripheral nerves are embedded in epineural tissues which are different in each individual [13]. Therefore, apparently identical nerve injury type may be associated with different rates of regain of function.

In conclusion, prompt diagnosis and institution of appropriate surgical repair to prevent adverse outcome was imperative.

REFERENCES:

1. Gunton JE, Stiel J, Clifton-Bligh P, Wilmshurst E, McElduff A. Prevalence of positive antineutrophil cytoplasmic antibody (ANCA) in patients receiving antithyroid medication. *Eur J Endocrinol* 2000; 142:587-596.
2. Mehindratta MM, Pandey S, Kushwaha RS, Mehndiratta P. Carbimazole induced ANCA positive vasculitis. *J Assoc Physicians India* 2008; 56:801-803.
3. Hall HA, Minc S, Babrowski T. Peripheral artery aneurysms. *Surg Clin N Am* 2013; 93(4):911-923.
4. Johnston KW, Rutherford RB, Tilson MD, Shah DM, Hillier L, Stanley JC. Suggested standards for reporting on arterial aneurysms. Subcommittee on Reporting Standards for Arterial Aneurysm, Ad Hoc Committee on Reporting Standards, Society for Vascular Surgery and North American Chapter, International Society for Cardiovascular Surgery. *J Vasc Surg* 1991; 13:452-458.
5. Kumar S, Agnilotri SK, Kharima SK. Brachial artery pseudoaneurysm following blood donation [Letter]. *Transfusion* 1995; 35(9):791.
6. Yetkin U, Gurbuz A. Post-traumatic pseudoaneurysm of brachial artery and its surgical treatment. *Tex Heart Inst J* 2003; 30:293-297.
7. Robbs JV, Naidoo KS. Nerve compression injuries due to traumatic false aneurysm. *Ann Surg* 1984; 200(1):80-82.
8. Ergungor MF, Kars HZ, Yalin R. Median neuralgia caused by brachial pseudoaneurysm. *Neurosurgery* 1989; 24(6):924-925.
9. Nugud OO, Hedges AR. Axillary artery pseudoaneurysm. *Int J Clin Pract* 2001; 5:494-499.
10. Singh A, Kumar V, Tandon S. Brachial artery pseudoaneurysm causing median nerve compression: A case report. *J Med Sci Clin Res* 2015; 3(5):5742-5745.
11. McNamara B. Clinical anatomy of the radial nerve: *Anatomy Primer*. ACNR 2003; 3:28-29.
12. Kimbrough DA, Metha K, Wissman RD. Case of the season: Saturday Night Palsy. *Semin Roentgenol* 2013; 48:108-110.
13. Düz B, Solmaz L, Civelek E, Onal MB, Pusat S, Daneymez M. Analysis of proximal radial nerve injury in the arm. *Neurol India* 2010; 58:230-234.
14. Posner MA. Compressive neuropathies of the median and radial nerves at the elbow. *Clin Sports Med* 1990; 9:343-363.
15. Han BR, Cho YJ, Yang JS, Kang JH, Choi HJ. Clinical features of wrist drop caused by compressive radial neuropathy and its anatomical considerations. *J Korean Neurosurg Soc* 2014; 55(3):148-151.
16. Nakamoto JM, Franklin SL, Geffner ME. Puberty. In: Kappy MS, Allen DB, Geffner ME (Eds) *Pediatric Practice: Endocrinology*, McGraw Hill Company Inc. New York, 2010: 257-298.