Tick induced facial nerve paresis

Jemma Porrett, Hashm Albarki, Adam Honeybrook

Department of Otolaryngology Head and Neck Surgery, Cairns and Hinterland Hospital and Health Service, Cairns, Australia *Correspondence to:* Dr. Jemma Porrett. Department of Otolaryngology Head and Neck Surgery, Cairns and Hinterland Hospital and Health Service, 165 The Esplanade, Cairns, Queensland 4870, Australia. Email: Jemma.Porrett@health.qld.gov.au.

> Abstract: We present a literature review examining the research surrounding tick paralysis resulting in facial nerve palsy. A novel case of an intra-aural paralysis tick bite resulting in unilateral facial nerve palsy is also discussed. A review of the literature was conducted using MEDLINE and EMBASE databases for relevant literature published between 1915 and 2020. Utilising the following key words: "Ixodes", "Facial paralysis", "Tick bite", and "Australia". Eighteen articles included in the review comprised a total of 48 patients. Patients' ages ranged from 1 to 84 years of age. Ten studies estimated the possible duration between tick bite and facial nerve palsy, averaging 8.9 days. Forty-one patients presented with a single tick within the external auditory canal (EAC), three had a single tick located on the temple or forehead region, three had post-auricular ticks and one patient had a remarkable 44 ticks removed from the face, scalp, neck, back and limbs. A complete ipsilateral facial nerve palsy was present in 45 patients, notably in 16 patients this occurred following tick removal. House-Brackmann classification was utilised in 7 patients; 4 patients with grade 4, 1 patient with grade 3, and 2 patients with grade 2 facial nerve palsy. Thirty-eight patients had complete recovery of facial palsy. Thirteen studies were analysed for time to recovery, with an average time of 19 days. Six patients had partial recovery at time of follow up. One patient died from respiratory arrest following generalized paralysis. Tick paralysis is a severe, but preventable disease. Careful examination of the face, scalp and EAC should be conducted in patients presenting with otalgia and facial nerve palsy, particularly in tropical areas, to exclude the possibility of tick infestation.

Keywords: Facial nerve palsy; tick bite; Australia; intra-aural; case report

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Introduction

Australia is a biodiverse continent centred between the Indian and South Pacific Oceans. It is populated by a unique variety of wildlife including approximately seventy species of ticks, sixteen of which feed on both humans and domestic animals (1). Only a few of these ticks are capable of human paralysis, with three species endemic to Australia; the most potent worldwide being the *Ixodes Holocyclus* located on the eastern coast of Australia and the less toxic *Ixodes Cornuatus* in Tasmania and Victoria and *Ixodes Hirsti* in South Australia (2-4).

The need for moist, humid conditions limits the distribution of ticks; most prevalently seen between September and March, associated with the Australian northern-easterly weather. Ticks have a life cycle consisting of four stages: egg, larva, nymph and adult. Each stage requires a new host in order to moult and evolve, thus requiring three hosts to complete their life cycle. Females host on human blood, whilst males host on female ticks. When fully engorged a tick can be anywhere from 200–600 times its original weight (1). The clinical presentation following tick bite can vary with some experiencing a mild localised allergic reaction and others anaphylaxis. Vector transmitted Rickettsia has been reported in Australia and most significantly neuromuscular paralysis has been well characterised (5-7).

Female *Lxodes Holocyclus* produce a holocyclotoxin from the salivary glands, a neurotoxin that acts similarly to botulinum toxin by inhibiting acetylcholine release presynaptically at the neuromuscular junction (4). Studies

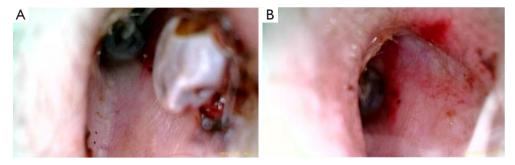


Figure 1 Patients external auditory canal. (A) Intra-aural tick; (B) post removal of tick from external canal.

in mice have shown worsening toxicity dependent on the duration of tick feeding, peaking at 4-5 days (8). Two types of tick paralysis have been described: generalised and isolated tick paralysis. Although rare, generalised tick paralysis can be fatal if left untreated (7). The onset of symptoms following *Ixodes Holocyclus* bite is slow with gradual progression of; lethargy, unsteady gait, slurred speech and blurred vision which subsequently can progress to ascending paralysis, bulbar palsy and potential respiratory paralysis if not recognised early (9). Isolated tick paralysis has been reported in patients with a tick infestation in the external auditory canal (EAC) (Otoacariasis) and surrounding scalp, resulting in facial nerve paresis (7). Thorough examination of the scalp and ear canal should be performed in these cases, with prompt removal of the tick for prevention of possible progressive paralysis.

We present a novel case of otoacariasis with associated ipsilateral facial nerve palsy in accordance with the CARE reporting checklist (available at https://dx.doi.org/10.21037/ ajo-20-88). All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient. A review of the literature is also presented to summarise data surrounding tick bites with associated facial nerve palsy; its incidence, aetiology, presentation and treatment with special focus on the Australian paralysis tick *Ixodes Holocyclus*.

Case presentation

A 67-year-old male presented to the Cairns emergency department with a 2-day history of left sided facial weakness. The patient was seen at an outside facility and was prescribed a 6-day course of ototopical antibiotics for a presumed left sided otitis externa. Upon presentation, the patient recalled an initial incident of stinging in the ear whilst cleaning an outdoor awning at his property 10 days prior. He also reported a 3-day history of feeling unsteady on his feet but denied any vertigo. He had no change in his hearing, no reports of tinnitus, vertigo or otorrhea. He reported normal motor and sensory function in bilateral upper and lower limbs. He denied any diplopia or vision changes. He was otherwise fit and healthy with no notable medical history.

On examination, the patient's vital signs were all within normal limits. The left ear and auricle appeared normal with no pain on palpation of the pinna or tragus and no herpetic lesions were noted. Otoscopy revealed a live tick in the posterior superior aspect of the lateral EAC with an intact and aerated tympanic membrane (Figure 1A,1B). The tick was identified by a veterinarian as Ixodes Holocyclus more commonly known as the Australian Paralysis Tick (Figure 2). Cranial nerve examination revealed paraesthesia in the left V1 distribution to light touch and a Grade 3 House Brackmann lower motor neuron palsy (Figure 3A). All other nerves examined normally. Tone, power, reflexes and sensation were normal in upper and lower limbs and ataxia was not witnessed on examination. The patient was given an immediate dose of 8mg dexamethasone. The left EAC was flooded with olive oil for one hour prior to removal with the intention to kill the tick. Despite oil application the tick remained alive at time of removal. Micro Alligator Forceps were used to remove the tick in entirety from the EAC. Remaining faecal material was removed with suction. The patient was commenced on Ciprofloxacin Hydrocortisone ear drops for 7 days, Prednisolone 60 mg for 10 days with tailing taper, oral doxycycline and we recommended frequent eye lubrication and protection at night.

Follow up was performed 6 days post presentation. Rickettsia serology returned negative as did testing for

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mammalian meat allergy. The patient had documented a daily diary describing a gradual resolution of forehead numbness and facial weakness. He reported complete resolution of facial weakness by Day 3, resolution of forehead paraesthesia on Day 4 followed by resolution of imbalance on Day 5. At time of review the ear canal examined normally. A slight frontalis muscle asymmetry was noted with intense contraction, however the patient reported this as pre-existing (*Figure 3B*).

Literature review

Methods

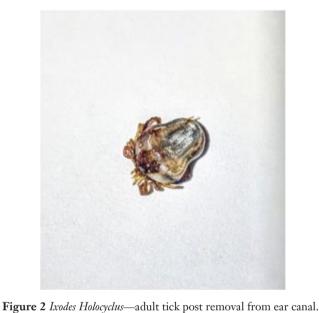
A review protocol was prepared in August 2020 and relevant studies were identified using MEDLINE and EMBASE. The aim of the review was to summarise all published literature between 1915 and 2020 to provide a succinct overview of tick induced facial nerve palsy. The search terms "Ixodes", "Facial paralysis", "Tick bite", and "Australia" were used. A language restriction was placed, with articles in English only. All articles describing facial nerve palsy in relation to Lyme Disease were excluded. Additional articles were screened using references of all studies reviewed for inclusion. Two independent authors reviewed abstracts for relevance and suitability for inclusion. Full texts were then reviewed.

Results

Electronic searching identified 133 citations in MEDLINE

Figure 3 Facial nerve function. (A) Initial presentation; (B) day 6 follow up review (this image is published with the patient's consent).





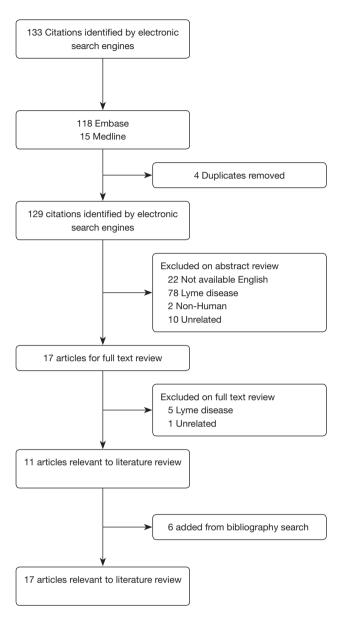


Figure 4 Flow diagram showing identification, screening, eligibility, and inclusion in this review.

and EMBASE from January 1, 1946 to November 1, 2020. Four duplicate papers were removed. The remaining papers were reviewed for relevance to the topic by abstract and keyword search with 18 articles meeting the criteria. Full texts were subsequently sourced for all articles with a further 5 texts excluded. A bibliographic search was completed on each full text, using these articles a further 7 articles were included in this review. A summary is included using the PRISMA (Preferred Reporting Items

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for Systematic Reviews and Meta-Analyses) flowchart (*Figure 4*). The level of evidence for each article was assessed per the Oxford Centre for Evidence-Based Medicine (OCEBM) 2011 Levels of Evidence criteria and the included studies largely ranged between level 4 and 5 OCEBM.

Eighteen articles, including 16 case studies and two case series were included, published between 1915 and 2020, comprising a total of 48 patients. Nine articles were published in Australia, three from Turkey, two from Malaysia and one article from both India and Sri Lanka. Interestingly, a study published in Singapore involved an Australian who had travelled just days prior to diagnosis and a further study published in New Zealand involved a New Zealander returning from Australia (10,11). The largest case series was from Sri Lanka involving 29 patients (12). Seven of the articles identified the tick type; including two cases with *Ixodes Holocyclus* and one presentation with *Ixodes Cornuatus*, three international cases of *Dermacentor* and two of *Hyalomma*.

Patients' ages ranged from 1 to 84 years of age. Ten studies estimated the possible duration between tick bite and facial nerve palsy, averaging 8.9 days (range, 3– 20 days). Forty-one patients presented with a single tick within the EAC, three had a single tick located on the temple or forehead region, three had post-auricular ticks and one patient had a significant 44 ticks removed from the face, scalp, neck, back and limbs.

A complete ipsilateral facial nerve palsy was present in 45 patients, in 16 patients this occurred following tick removal (range, 1–7 days). The ipsilateral temporal branch of the facial nerve was affected in 3 patients. House-Brackmann classification was utilised in 7 patients; four patients with grade 4, one patient with grade three and two patients with grade 2 facial nerve palsy.

Of those with ticks in the ear canal, 81% complained of associated otalgia. One patient with a tick to the forehead had associated facial swelling thought to be due to an allergic reaction. Three reported altered sensation of the ipsilateral face. One patient had an associated ipsilateral sensorineural hearing loss, confirmed on formal audiometry, and had complete recovery at one-month follow up. A 3-year-old patient suffered from a generalized tick paralysis requiring a period of mechanical ventilation and after extubation the child had a complete recovery.

Ticks were removed in all cases, one requiring incision prior to removal and two children required removal in theatre. Six studies reported the use of topical agents prior to removal including; alcohol, ether, Vaseline, 4% lignocaine and 10% cocaine. Thirty-four patients received antibiotics following removal, the majority receiving penicillin for prophylaxis or treatment of secondary skin infection. In 10 patients, doxycycline was given for treatment of possible Rickettsia pending serology and in a single pregnant patient azithromycin was given with this intention. Thirty-two patients received prednisolone 1mg per kg and one patient received intravenous steroids. Seventeen patients received acyclovir.

Two patients underwent testing for Lyme's disease, in both cases this was negative. Ten patients were tested for Rickettsia with six testing positive.

Thirty-eight patients had complete recovery of facial palsy, thirteen studies were analysed for time to recovery, with an average time of 19 days (range, 3 days to 15 weeks). Kularatne *et al.* reported ranges for recovery in their cohort of 39 patients with recovery time ranging from 1 to 55 months duration (12). Six patients had partial recovery at time of follow up. One article reported improvement in facial nerve palsy at 24 hours but no further follow up was reported. One patient was lost to follow up and one article failed to mention any resolution of facial nerve palsy. One patient died from respiratory arrest following generalized paralysis (*Tables 1,2*).

Discussion

Unlike other intra-aural insect bites, ticks tend to have a delayed presentation due to reduced pain at the time of attachment. This is due to the numerous pharmacologically active substances contained in tick saliva. This includes anaesthetics, anti-nociceptives and anti-inflammatory compounds (13).

Otalgia is the most common symptom, reported to occur in 81% of cases included in this review. This has previously been reported in up to 90% of cases of otoacariasis (14). The mean time of otalgia to diagnosis amongst previously reported case studies was 10 days (range, 3–20 days). Kularatne *et al.*, however, reported a mean duration of 5 days (range, 1–13 days) (12). This may be due to the higher prevalence within the region. As such, clinicians may have a higher index of suspicion of otoacariasis. In the case presented, the tick was removed 10 days after otalgia was reported. This was likely delayed by 6 days as the patient was initially diagnosed with otitis externa.

Eight percent of cases reported ipsilateral paraesthesia associated with the facial nerve palsy. It is unclear how a

trigeminal nerve palsy can be caused by otoacariasis. The holocyclotoxin has only been shown to cause a disorder of motor neurones (4). Miller *et al.* [2002] suggests that the paraesthesia is secondary to altered proprioception, as seen with Bell's palsy (15).

In one case, a sensorineural hearing loss confirmed on audiometry was also reported (16). The reporting authors suggest that the toxin may be able to diffuse through the tympanic membrane and the round window, leading to a sensorineural hearing loss. The patient made complete recovery at 1 month follow up (16).

There is limited research surrounding the pathophysiology of tick-induced facial nerve paralysis. The earliest documented explanation suggests that the paralysis is caused by a localised urticaria, oedema and subsequent pressure on the nerve (17). However, urticaria has been shown to occur within an hour of the tick bite and onset of facial nerve paralysis occurs between 3–20 days. Additionally, the associated oedema of the EAC appears less severe than that seen in otitis externa or furunculosis of the ear canal (18).

The female *paralysis tick* produces a holocyclotoxin from the salivary glands. This neurotoxin acts similarly to botulinum toxin, inhibiting acetylcholine release presynaptically at the neuromuscular junction (4). Indudharan and colleagues propose that the toxin gains access to the middle ear through a perforation following the tick bite. When combined with a natural dehiscence of the facial canal, the toxin may cause a facial nerve palsy (14,19). However, this explanation does not account for patients with an intact tympanic membrane. Kularatne and colleagues state that given holocyclotoxin is a highly diffusible protein, it is unlikely to cause an isolated nerve palsy. Alternatively, they found that treating the associated Rickettsia following a tick bite, there was a faster resolution of the facial nerve palsy (12).

There is a paucity of data surrounding the management of otoacariasis and subsequent facial nerve palsy. In general, most patients had the tick removed on diagnosis with no medications provided. A small group of patients had their ear canal flooded prior to removal. Once the tick is identified, prompt removal should be performed. The use of chemical agents prior to the removal of ticks varied considerably. On review of the literature, one patient received petroleum jelly, three received alcohol, one received 4% lignocaine and one further patient had 10% cocaine instilled into the EAC.

Petroleum jelly has previously been reported as

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Table 1 Case study outcome

Article	Country of publication	Age (years)	Tick type	Tick location	Facial paralysis at presentation	Otalgia (Y/N)	Associated symptoms	Canal flooded (Y/N)	Adjunct treatment	Resolution of paralysis	Outcome
Strickland 1915	Australia	11	N/A	EAC	Y	Y	Generalised paralysis	Ν	N/A	10 days	Resolved
Ferguson 1924	Australia	1	N/A	Post- auricular	Y	N/A	Generalised paralysis	Ν	N/A	N/A	Death
Foster 1931	Australia	45	N/A	EAC	Y	Y	N/A	Ν	N/A	3 days	Resolved
Crossle 1932	Australia	2	N/A	EAC	Y	Ν	N/A	Y	N/A	3 weeks	Resolved
Hamilton 1940	Australia	7	N/A	Temple	Y	N/A	N/A	Ν	N/A	15 weeks	Resolved
Pearn 1977	Australia	6	N/A	Post- auricular	Y	N/A	N/A	Ν	N/A	5 days	Resolved
Tibballs <i>et al.</i> 1986	Australia	3	lxodes Cornuatus	Post- auricular	Y	N/A	Generalized paralysis	Y	Ventilation; tick; anti- toxin	11 days	Resolved
Barber <i>et al.</i> 1994	New Zealand	60	N/A	Forehead	Y	N/A	Facial swelling	Ν	Antibiotics	1 month	Resolved
Indudharan <i>et al.</i> 1996	Malaysia	65	Dermacentor sp.	EAC	Y	Y	N/A	Y	N/A	N/A	N/A
Miller 2002	Australia	48	N/A	Upper Body	Y	N/A	Facial paraesthesia	Ν	N/A	7 days	Resolved
Patil <i>et al.</i> 2010	India	3	N/A	EAC	Y	Y	N/A	Ν	Antibiotics	1 week	Resolved
McGrath 2012	Australia	84	N/A	EAC	Ν	N/A	Facial paraesthesia	Ν	N/A	7 days	Resolved
Doğan <i>et al.</i> 2012	Turkey	33	Hyalomma sp.	EAC	Y	Y	Nil	Y	Steroids	N/A	N/A
Uguz <i>et al.</i> 2015	Turkey	47	Hyalomma Marginatum	EAC	Y	Y	Facial paraesthesia	Ν	Steroids	2 weeks	Resolved
Pek 2016	Singapore	63	lxodes Holocyclus	Temple	Y	N/A	Facial paraesthesia	Ν	Antibiotics	3 weeks	Resolved
Kutuk 2020	Turkey	45	lxodes Holocyclus	EAC	Y	N/A	SNHL	Y	Steroids; antibiotics	4 weeks	Resolved

EAC, external auditory canal; Y, yes; N/A, not applicable; N, no; SNHL, sensorineural hearing loss.

successful in inducing detachment of tick. This has been assumed to be secondary to the disruption of the tick's breathing (20). We attempted this method with the use of olive oil, however detachment was unsuccessful. Needham *et al.* found that occlusion of the tick's air supply for several hours had no effect, likely due to their extremely slow respiratory rate (20,21). The use of alcohol has also been shown to have little effect on tick removal, however, there is some benefit in disinfecting the canal prior to removal (20). It should be noted that ethyl alcohol and chlorhexidine are ototoxic, especially in the context of a tympanic membrane perforation. Therefore, an alternative of half strength

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Table 2 Case	Table 2 Case series outcome	ıe									
Article	Country of publication	z	Age mean (range)	Tick type [N]	Tick location	Facial paralysis at presentation (%)	Otalgia (%)	Canal flooded (Y/N)	Adjunct treatment	Resolution of paralysis (range)	Outcome
Kularatne et al. 2018	Sri Lanka	12	Sri Lanka 12 45 [19-76]	Dermacentor Auratus [8]	EAC	67	93	z	Antibiotic; steroid	-, (1–55 months)	Resolved 83%; partial recovery 17%
		9	50 [37–72]	N/A	EAC	50		z	Antibiotic; steroid; anti-viral –, (1–3 months)	-, (1–3 months)	Resolved 60%; partial recovery 40%
		÷	49 [33–68] [49 [33-68] Dermacentor sp. [1]; Hyalomma sp. [1]; Amblyomma sp. [1] 	EAC	45		Z	Antibiotic; steroid; anti-viral	-, (1- 13 months)	Resolved 82%; partial recovery 18%
Amin <i>et al.</i>	Malaysia	-	5	N/A	EAC	100	100	≻	Steroid	7 days	Resolved
2007		-	-	N/A	EAC	100	100	z	Antibiotic	7 days	Partial recovery
		-	78	N/A	EAC	100	100	z	N/A	N/A	Lost to follow up
EAC, externa	al auditory car	, Y ;ler	yes; N/A, not	EAC, external auditory canal; Y, yes; N/A, not applicable; N, no.							

betadine should be used with caution (22). The use of local anaesthetic for tick removal has been examined, with no effect seen on tick release. Local anaesthesia may make removal more comfortable (23). This should also be used with caution due to risk of severe position induced vertigo if contact is made with the middle ear (24).

The tick detected in our case was removed with alligator forceps. Careful removal of the tick was performed, avoiding head rotation as this may lead to further saliva being released. Thorough examination of the area was conducted following removal to ensure complete removal. Remnant mouth parts may lead to further inflammation, infection and granuloma formation (25). Symptomatic management for patients with facial nerve palsy is essential including; eye care, lubricating drops in daytime, eye ointment and taping at night.

A single dose of 8mg intravenous dexamethasone was given to our patient, as well as a 10-day course of oral prednisolone 60 mg daily with a taper. This management approach was adopted from the treatment guidelines for Bell's palsy. Steroids have been shown to improve recovery time in patients suffering from idiopathic facial nerve palsy (26).

Kularatne *et al.* attempted to improve time to recovery with the addition of aciclovir within their case series with little success (12). The use of antiviral therapy in Bell's palsy stems from the suspicion of herpes simplex virus as the causative factor of idiopathic facial nerve palsy. Its use however is controversial at best, with two of the largest randomised control trials showing no benefit with use of antiviral versus placebo or antiviral with glucocorticoid versus glucocorticoid alone (26,27).

The patient discussed in the case presented received doxycycline as advised by the local clinical toxicologists for the treatment of possible Rickettsia (12). Literature suggests routine antibiotic prophylaxis following tick bite is not indicated unless the patient is pregnant or in an area endemic to tick-borne disease (28).

A single case reported in the literature received tick antitoxin. This patient received the anti-toxin after developing a generalised tick paralysis. Tick antitoxin is heterologous in nature and has been shown to induce serum sickness and anaphylactic shock in humans and is therefore not recommended (4).

The prognosis remains excellent in patients presenting with tick induced facial nerve palsy. An initial worsening of the palsy may occur following tick removal and patients should be advised on the expected clinical course.

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Consideration should be made for tick induced mammalian meat allergy. Tick bites may transfer galactose-alpha-1,3-galactose, a carbohydrate found in mammalian cell membranes except for apes and humans (29). A delayed anaphylaxis may occur due to IgE anti-gal antibody formation and therefore discussion with an immunologist is advised.

Our review has its limitations, due to the lack of availability of research surrounding tick bite induced facial nerve palsies. As a result of this, articles dated back to 1915 were sourced for inclusion. The articles published were assessed for levels of evidence and largely ranged between level 4 and 5 OCEBM. This evidence is subject to bias, leading to a limitation of treatment recommendations. Further research in this area is recommended with higher quality studies required to determine if adjunct treatments are warranted.

Conclusions

Facial nerve palsy is a debilitating complication following paralysis tick bite, albeit with a good overall prognosis. Careful examination of the face, scalp and EAC should be conducted in patients presenting with otalgia and facial nerve palsy. Prompt removal of ticks should be conducted, preventing progression of disease. The use of steroids may be of benefit. Australian clinicians working in tick endemic areas are encouraged to have a high degree of suspicion for tick paralysis.

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Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://dx.doi. org/10.21037/ajo-20-88

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://dx.doi. org/10.21037/ajo-20-88). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are

appropriately investigated and resolved. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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