



ISSN: 0975-833X

Available online at <http://www.journalcra.com>

INTERNATIONAL JOURNAL
OF CURRENT RESEARCH

International Journal of Current Research
Vol. 15, Issue, 01, pp.23356-23359, January, 2023
DOI: <https://doi.org/10.24941/ijcr.44644.01.2023>

RESEARCH ARTICLE

TRACHEOMALACIA; A POSSIBLE SYSTEMIC EFFECT OF CHRONIC STEROID USE

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ARTICLE INFO

Article History:

Received 16th October, 2022
Received in revised form
10th November, 2022
Accepted 19th December, 2022
Published online 30th January, 2023

Key words:

Chronic use of Topical Steroids, Skin Whitening, Systemic Complication, Tracheomalacia, Tracheostomy.

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ABSTRACT

Background: Tracheomalacia is a structural deficit of the tracheal cartilages inducing excessive collapsibility of the trachea. Origin: Acquired conditions more common in adults. **Case report:** A report of a 23year old lady who presented with a benign, simple small sized goiter of 9 months duration scheduled for thyroidectomy. She had no retrosternal extension, pressure symptoms or features of respiratory insufficiency. Patient had thin, fragile depigmented (bleached) skin with discolouration. She had extensive skin striae (stretch marks) on the trunk, abdomen and pelvic region despite having a slim stature. She had been using a potent skin whitening cream containing two potent topical steroids for about 3 years. Prior to surgery, patient had intravenous exogenous dexamethasone to prevent the potential risk of the suppression of the hypothalamo-pituitary-adrenal axis with a subsequent inappropriate response to surgery. Intra-operatively the lead surgeons had called the teams' attention to the abnormal anatomy of the patients' trachea. The lumen of the trachea appeared narrowed as evidenced by the reduced anterior-posterior airway caliber with associated loss of the semicircular shape of the tracheal rings and bulging of the posterior membranous wall. The diagnosis of tracheomalacia was made and an emergency tracheostomy was performed. The endotracheal tube was extubated after proper suctioning. Surgery was concluded and patient was transferred to the high dependency unit for close monitoring. The serum cortisol and urinary cortisol levels were elevated (28mcg/dl and 55mcg/day respectively). This report highlights the unusual presentation of this patient and emphasizes the place of interprofessional team work required in her management. **Conclusion:** Tracheomalacia is a life threatening condition that is mostly of acquired origin in adults. Chronic use of topical steroids has been associated with systemic complications such as atrophy, and weakening of airway cartilages but these effects has been sparsely discussed or documented in literatures particularly relating to anaesthesia. This report presents an unusual presentation of suspected tracheomalacia secondary to chronic use of topical steroids.

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Citation: Fagbohun A.O., Akinyemi S.O, Eke G.N. and Fashina O.A. 2023. "Tracheomalacia; A Possible Systemic Effect Of Chronic Steroid Use". International Journal of Current Research, 15, (01), 23356-23359.

INTRODUCTION

Tracheomalacia is a life threatening condition characterized by a structural deficit of the tracheal cartilage leading to excessive collapsibility of the trachea. It occurs as a result of weakness of the tracheal wall caused by softening and thinning (atrophy) of the supporting hyaline cartilage, hypotonia of the interconnecting fibroelastic tissue and posterior myoelastic elements. Tracheomalacia occurs when the cartilaginous framework of the trachea is unable to maintain airway patency [Griffiths, 2000; Yang, 2022] Although there is no internationally agreed classification system for tracheomalacia, the origin of this condition can be divided into three main groups: congenital defects in tracheal integrity, secondary acquired conditions weakening the whole or part of the tracheal structure, and conditions causing extrinsic compression of the airway.

The first category are conditions that cause immaturity of the trachea; the other two categories are conditions in which a normally developed trachea undergoes a degeneration process [Yang, 2022; Wallis, 2019]. Secondary acquired tracheomalacia post thyroidectomy is usually associated with huge long standing symptomatic thyroids. Small sized, benign, recently occurring asymptomatic thyroid masses are not usually associated with the condition [Valizadeh, 2020]. Prolonged use of steroids is associated with various side effects. One of the documented complications is its effect on the airway with patient presenting with various symptoms from hoarseness of voice (dysphonia) to symptoms of respiratory insufficiency [Naseer, 2011]. There is evidence that topical steroids are absorbed systemically. The extent of absorption is small and associated problems are minimal except when strong steroids are used regularly on large surface areas. Small doses of a potent topical steroid can lead to side effect [Dhar, 2014].

Whitening creams contain steroids in doses up to a thousand times more than topical creams used in treatment of skin conditions like eczema. Some manufacturers combine two or more potent topical steroids [Maneli, 2016]. Patients who are in the habit of using these agents are at potential risk of complications of prolonged or long term steroid use like suppression of hypothalamic-pituitary-adrenal axis and iatrogenic Cushing's syndrome with suppression of adrenocorticotropic hormone (ACTH) [Naseer, 2011; Riegelman, 1974]. Patient may present with features such as diastolic hypertension, diabetes, buffalo hump, hirsutism, striae, telangiectasia, skin fragility and discoloration and others. It may also cause thinning (atrophy), softening and hypotonia of airway cartilages and surrounding structures leading to rare conditions like laryngomalacia, tracheomalacia [Naseer, 2011]. Correct diagnosis of tracheomalacia is made by the observation of the collapse of the cartilagenous and membranous part of the trachea (4).

CASE REPORT: A 23 year old female who was 1.5m tall and weighed 60kg with a BMI of 26.6kg/m² presented with a simple goitre and was scheduled for thyroidectomy. She had a history of use of a skin whitening cream for about 3 years duration. On examination, the patient had an anterior neck swelling about 4.5cm by 3cm in diameter that moved with deglutition. It had gradually increased in size over a period of about 9 months (no period of accelerated growth) and had well defined margins. There was no retrosternal extension in this patient. She was assessed as American Society of Anesthesiologists physical status (ASA) 11 and her airway was assessed as a Mallampatti I. Cardio-respiratory assessment was normal with a heart rate of 82 beats /min, blood pressure of 120/68mm of Hg. Heart sounds were normal. Blood investigations revealed haemoglobin of 13.6 gm%, fasting blood sugar 84 mg%, Electrolyte urea and creatinine was normal. Thyroid function was in keeping with a euthyroid state. Pulmonary function tests were not indicated as patient did not have compression or respiratory symptoms. There was no pathology seen on chest X-ray, Electrocardiography, and Echocardiography. On indirect laryngoscopy both vocal cords were mobile.

On the morning of surgery, patient was taken to the operation theatre. Two wide bore intravenous accesses were secured with 18-G cannulas. Standard monitors including a non invasive blood pressure, Electrocardiogram and pulse oximeter, capnograph and temperature probes were connected. She was premedicated with 8mg dexamethasone which was to continue every 6 hours for 48 hours. Airway management cart was kept ready. Premedication included glycopyrrolate 0.2 mg, fentanyl 100 µg. Patient was pre-oxygenated for about 5 minutes and anaesthesia was induced with propofol at 2.5mg.kg⁻¹ intravenously with a total dose of 140mg given. After confirming ventilation, succinylcholine at 1.5mg.kg⁻¹ up to a total dose of 100mg was given intravenously for ease of intubation. Patient was intubated with ease at the first attempt with a 7.5 mm ID reinforced endotracheal tube (ETT). After confirming tube placement, ETT was secured. Anaesthesia was maintained with Isoflurane at 1-2% in 100% oxygen. Intravenous atracurium was given at a dose of 0.5 mg.kg⁻¹ Patient remained hemodynamically stable till the end of surgery. The mass excised weighed about 10grams. At about 1hour.10mins into surgery, the lead surgeon, a consultant General surgeon called the attention of the team to the abnormal appearance of the trachea as evidenced by the narrowing of the lumen of the trachea, reduced anterior-posterior airway caliber with associated thinning and loss of the semicircular shape of the tracheal rings with bulging of the posterior membranous wall. The tip of our endotracheal tube was located about 5cm above the carini below which the suspected collapse was evident. A diagnosis of tracheomalacia was made and the Otorhinolaryngologist team was immediately invited and an emergency tracheostomy was performed. A serum cortisol sample was thereafter taken which later revealed a serum cortisol level of 28mcg/dl (reference range is 10-26mcg/dl) while her 24 hour urinary cortisol sample yielded a cortisol level of 55mcg over 24 hours (reference range of 3.5 to 45mcg/24 hour). The result of both urine and serum cortisol were both above the reference values. Patient was extubated post tracheostomy after thorough oropharyngeal and

tracheostomy suction. The tracheostomy tube was then connected to the anaesthetic machine till surgery ended. At the end of surgery, isoflurane vaporizer was switched off, oxygen was continued till patient was fully awake. Neuromuscular blockade was reversed with neostigmine, and glycopyrrolate in the doses of 2.5 mg and 0.4 mg respectively She was taken to the recovery room breathing room air spontaneously and maintaining oxygen saturation between 98-100%. The tracheostomy tube was suctioned as frequently as was required. Patient was eventually transferred to the high dependency unit. She was discharged home on the 5th day post surgery for follow up clinic on outpatient basis with both the general surgery team and the Orthorhinolaryngologists.

DISCUSSION

The trachea is the tube like structure that connects the larynx to the distal airways. Its structural support is derived from 16 to 20 rings of hyaline cartilages interconnected by fibroelastic tissue anterolaterally and a fibromucular wall posteriorly. The rings form a "U" shape as they are incomplete at the posterior aspect, which is covered by a posterior membrane [Yang, 2022]. A variety of different disease processes can disrupt this support structure, causing narrowing of the lumen of the trachea (reduced anterior-posterior airway caliber) with associated loss of the semicircular shape and bulging of the posterior membranous wall. A tracheal collapse capable of producing an obstacle to lung ventilation must be greater than 50% of the tracheal diameter. These means that, clinical evidence of tracheomalacia occurs when greater than 50% of the trachea is affected [Yang, 2022].

Tracheomalacia is a condition in which the supporting cartilage of the trachea is distorted leading to weakness and floppiness of the trachea. It is either of congenital or acquired origin. The congenital conditions cause immaturity of the trachea while the acquired conditions cause a normally developed trachea to undergo a degenerative process. These acquired conditions either weaken the whole or part of the tracheal structure, or cause extrinsic compression of the airway [Yang, 2022; Wallis et al., 2019]. The occurrence in adults is usually of acquired origin. The cause is mostly unknown but associated risk factors may be present suggesting the cause. Tracheomalacia in adults becomes clinically evident when greater than 50% of the tracheal structure is compromised [Yang, 2022]. This may explain why our patient had no clinical symptoms pre-operatively. Although the aetiology of tracheomalacia in adult is mostly unknown, in this patient; highest on the list of our suspicion is the effect of prolonged use of steroids. Our patient had been on a cream with 2 very potent topical steroids (betamethasone and clobetasol propionate). She had presented with a depigmented (bleached) skin with discolorations, striae, (stretch mark) on the upper arms, trunks, her buttocks and thighs. She had prominent greenish vessels and very thin, fragile skin with reduced elasticity and she also had beards. All this had made the team to be suspicious of a possible depression of the adrenal gland. Other acquired causes such as infection, external compression from the mass, malignancy that could have been close differentials are less likely as patient did not have any pre-morbid sign or symptom of airway distress. The thyroid mass was small sized, benign, recently occurring (about 9 months) and asymptomatic hence less likely to cause tracheomalacia.

Tracheomalacia when diagnosed peri-operatively in a patient scheduled for thyroidectomy, is commonly as a result of external compression effect from a huge long standing thyroid. [4] In our patient, the thyroid mass was a simple goitre with no history of compression or respiratory effect and it had developed just over a 9 months duration. Also, a thyroid mass impeding on the trachea to the extent of causing tracheomalacia would likely cause some degree of difficulty at intubation. This patient who weighed about 60kg was intubated with ease at the first attempt with a 7.5 mm ID tube. Available literatures do not associate small sized recently occurring asymptomatic thyroid mass with tracheomalacia. The usual trend is to have a patient with a huge long standing thyroid mass with associated symptoms of compression such as dysphonia/hoarseness, change in voice volume, problems with deglutition and features of respiratory

difficulty/insufficiency. Interestingly, some authors have argued that tracheomalacia is rare even in patients with huge long standing thyroid masses [Valizadeh, 2020; Findlay, 2011]. The gold standard investigation for the diagnosis of tracheomalacia is bronchoscopy which directly visualizes the airway during spontaneous respiration. This is done along with other investigations. Bronchoscopy will demonstrate the narrowing of the tracheal lumen, loss of the semicircular shape of the tracheal cartilages and the bulging of the posterior membranous wall [Valizadeh, 2020]. Bronchoscopy was not requested for in the index case as our patient did not present with any complaint or sign indicative of a compromise of either the upper or lower respiratory airways. The diagnostic imaging had been normal, scan had not shown any evidence of a retrosternal extension and the fine needle biopsy was in keeping with a benign mass. The diagnosis of tracheomalacia in this patient was an intra-operative incidental finding made at direct visualization of the trachea. There was narrowing of the lumen of the trachea, reduced anterior-posterior airway caliber with associated thinning and loss of the semicircular shape of the tracheal rings with bulging of the posterior membranous wall all being evidence of distortion in the supportive structure of the trachea. With the chronic use of topical steroids, there is an inhibition of the hypothalamo-pituitary-adrenal axis by the negative feedback mechanism with subsequent elevated serum and urinary cortisol and low adrenocorticotrophic hormone, (ACTH) levels which may be indicative of Cushing syndrome (10). In normal healthy patients, dexamethasone, which is similar to cortisol, lowers the amount of ACTH released by the pituitary gland. This in turn lowers the amount of cortisol released by the adrenal glands. Conversely, patients who have been on long term steroid use may present with high cortisol levels despite low ACTH after a dose of dexamethasone. (10). Hence the assessment of cortisol and ACTH is essential in the diagnosis of conditions suggesting hypercortisonism. In our patient, because of our high index of suspicion for adrenal insufficiency from the chronic use of topical steroids, she was commenced on intravenous dexamethasone 8mg prior to surgery. With the intra-operative finding of tracheomalacia, samples were immediately taken for baseline serum cortisol and a 24 hour urine cortisol subsequently. Both the serum and urine cortisol levels were elevated albeit minimally. Her baseline serum cortisol level was 28mcg/dl (reference range is 10-26mcg/dl) while her 24 hour urinary cortisol sample yielded a cortisol level of 55mcg over 24 hours (reference range of 3.5 to 45mcg/24 hour). Considering the fact that she had been given dexamethasone prior to surgery, both the serum and urine cortisol levels should not be raised (10) This further supports the suspicion of a systemic response to chronic use of topical steroids. However, a serial cortisol test with adrenocorticotrophine (ACT) assessment would have been more appropriate to further substantiate our hypothesis. The dermatologists were invited to review the patient and the team confirmed that they had several cases of suspected systemic effects due to chronic topical steroid use in form of lightening cream. They had been collating supporting data to help in their campaign and enlightenment on the effect of chronic steroid use.

There is evidence that topical steroids are absorbed systemically. The aim of applying topical steroid is to deliver therapeutically effective dosage of the drug to the target organ with least possible side effects mainly the systemic ones. Overtime, data have accumulated indicating percutaneous absorption of topical steroids depends on a number of factors (11). The horny layer of the skin serves as a reservoir from which drug penetration into the body continues even after a single application [Wester, 1983] Therefore, even small doses of potent topical steroids can produce systemic side effects like suppression of hypothalamic-pituitary-adrenal axis with subsequent adrenal gland atrophy and iatrogenic Cushing's syndrome from the suppression of the hypothalamic cortisol releasing hormone and pituitary adrenocorticotrophic hormone (ACTH) [Wester, 1983]. Patient may present with features of iatrogenic Cushing's syndrome such as diastolic hypertension, diabetes, buffalo hump, facio-truncular obesity, hirsutism, striae, telangiectasia, skin fragility and others. Whitening creams contain steroids in doses up to a thousand times more than topical creams used in therapeutic treatment of skin conditions like eczema hence patients especially young adults who are

in the habit of using these agents are at potential risk of complications associated with long term steroid use such as tracheomalacia as it causes thinning (atrophy), softening and hypotonia of the tracheal cartilages. With thinning of the skin, there is increased absorption of these topical steroids leading to worsening of the complication. Tracheomalacia in the index case could be referred to as an incidental finding. Management of tracheomalacia is multipronged involving a multi-professional approach. Professionals such as Anaesthetists, Perioperative nurses, Surgeons, Dermatologists, physicians and others are involved. Generally the decision to perform a tracheostomy is based on clinical judgement (12) Therefore, after our diagnosis of tracheomalacia, a tracheostomy was performed. The prognosis of a secondary acquired tracheomalacia depends on the pathology that produced the alteration in the trachea and on the degree of the lesion. (2)

CONCLUSION

Tracheomalacia is a life threatening condition that is mostly of acquired origin in adults. The cause is usually unknown however associated risk factors may elucidate the origin better. Chronic use of steroids has been associated with systemic complications such as atrophy, and weakening of airway cartilages but these effects has been sparsely discussed or documented in literatures particularly relating to anaesthesia. This report is an effort to sensitize anaesthetists, critical care workers and other health professionals to an impending pandemic particularly relating to the African Nations considering the present passion and preference of our people for the white skin.

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