

CASE REPORT

TRACHEO-CUTANEOUS FISTULA: A RARE PRESENTATION DUE TO A FORGOTTEN RUBBER BAND IN NECK OF A YOUNG GIRL**Amna Ikram, Amna Idrees, Somia Ilyas, Sikandar Ali, Muhammad Ali Sheikh**

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The presence of foreign bodies in a chronic wound is a well-known phenomenon. The common locations are soft tissues of the wrist and neck, gastro-intestinal foreign objects, foreign objects in the ear and in the Uro-genital tract. aero digestive tract and chest. Retained foreign bodies in soft tissues become reason for formation of sinus which can be superimposed by infection. Most of foreign bodies in soft tissues remain un-detected, becoming the reason for delayed diagnosis. Some of the foreign bodies are inert and stay in the body tissues for prolonged time without causing any deleterious effect but few of them erode into surrounding tissues and cause prolonged illness. Tracheo-cutaneous fistula secondary to a foreign body in a child is rarely reported. In this case, the patient had retained foreign body in her neck for almost eight years which remained undiagnosed while being treated for various types of skin disease.

Keywords: Fistula; Foreign body; Neck sinus; Rubber band; Chronic inflammation

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INTRODUCTION

A sinus is a blind tract leading from epithelial surface down to the underlying tissue which is basically due to connection with a deep-seated focus of suppuration. It is usually lined by granulation tissue which later may be epithelialized.¹ According to a study done at the Oulu University Hospital, an incidence (29.2 per 100,000) of foreign body injuries in children was found during the 6-year study period.

A chronically retained foreign body can lead to formation of a sinus, which could lead to formation of a long narrow tract that is unable to drain itself. This would eventually lead to tortuousness of the sinus tract and dense fibrosis which will eventually halt healing by causing ischemia and preventing the closure of the tract. Certain types of infections like tuberculosis and actinomycosis are well known to be associated with formation of chronic sinuses. Malignant diseases may also cause formation of chronic sinuses. Similarly, ischemia of the tissues, various drugs like steroids, cytotoxic drugs, malnutrition or radiotherapy may also be the contributing factors.²

Some wounds containing foreign bodies have the ability to heal without any complications. Granuloma formation and infection are common sequelae of retained foreign bodies. Toxic and allergic reactions are rarely associated with retained foreign bodies.³

Prolonged wearing of rubber bands, strings or any other such object eventually starts causing constrictive effects on limbs and even formation of burrows across the skin resulting in distal oedema,

compartment syndrome leading to impaired function of the limbs.⁴ We are reporting an unusual case of retained foreign body in the neck of a young girl which was the cause of multiple chronic sinuses and single trachea-cutaneous fistula because some skin lesions are remained undiagnosed for many years which lead to irrelevant investigations and treatment.

CASE REPORT

A 11-year-old female was referred from dermatology department for biopsy of multiple chronic neck sinuses. According to her mother these sinuses were initially noticed when she was 3 years of age. They appeared as small, multiple and painful nodules which later eroded through overlying skin.

She had received different treatments and multiple hospitals but her sinuses persisted. These sinuses increased in size gradually and started having purulent discharge. At the age of 6 years anti tuberculosis therapy was commenced that was continued for a period of nine months. After interval of six months, second line ATT was started which she received for 1 year. She underwent biopsy of her lesions four times at two different hospitals and they were all reported as chronic inflammation with granulation tissue.

Her investigations including complete blood count, renal function tests, liver function tests, ESR, IgE, IgA, Ig M, HIV screening, Mantoux Test and hepatitis profile were within normal range. There was no history of fever or weight loss and no history of skin disease in the family. She was also treated on the line of Job's syndrome and scrofuloderma. Her neck lesions at the time of presentation are shown in Figure-1.



Figure-1: Preoperative picture showing multiple neck sinuses



Figure-2: Operative picture showing rubber band coming from wound in the neck



Figure-3: Removed rubber band.

On examination, sinuses were fixed, soft, tender and soft with apparent seropurulent discharge. Her cervical and axillary lymph nodes were impalpable. Systemic examination was unremarkable.

She was admitted in ward and optimized for surgery. Intra-operatively there were multiple skin

lesions all around her neck having excessive granulation tissue with inter-linked fibrous tissue having thick cheesy pus discharge. A foreign body was noted deep inside the lesion. It was a rounded rubber band which was about 18cm long and it came out from right lateral lesion of neck 1 cm below the skin surface as shown in Figure 2 & 3. Upon asking about it later, the parents vaguely mentioned that she might have been playing with the rubber band and wore it during her play and it might have remained unnoticed because the child was covered in winter clothes.

Air was coming out from lateral border of central lesion that was about 0.5mm in size in all directions and was in front of neck. It was a tracheo-cutaneous fistula as air bubbles could be seen coming out when it was washed. Opinion was sought from ENT department regarding her tracheo-cutaneous fistula and it was decided to treat it conservatively because patient did not have any complaints regarding it, so no further investigation was done. A simple air tight gauze dressing was placed on fistula site. Two specimens were taken for biopsy and sent for histopathology.

Wound was covered with simple dressing. Her post op recovery was uneventful. She was discharged after 2 days on oral analgesia and antibiotics.

On follow up after 7 days, patient had recovered with healed sinuses and tracheo-cutaneous fistula had closed. Histology of the specimen was reported as chronically inflamed nonspecific fistulous tract.

DISCUSSION

Cutaneous wounds that contain undetected foreign bodies could be considered as a minor focus and does not necessarily mean that should be accompanied by any serious symptoms. This could lead to delayed diagnosis and hence it could be the reason of early specific treatment failure.

Some foreign objects are harmless and hence left undetected in the wound but some may cause intense reaction, inflammation and even infection which can result in serious consequences such as sinus, fistulous tract formation and therefore, these must be removed. Foreign bodies which cause soft tissue infections are generally resistant to treatment unless the foreign object is removed.⁵

There are also some household things which are commonly encountered as foreign body in limbs such as thread which is tied very frequently among Indian population as a part of their culture. They do not remove this thread from their wrist by themselves which results in constriction and embedding of thread in the skin by chronic cutting through soft tissues, so

that the healing potential of tissue in children covers the dermis and that thread gets completely buried underneath. Other examples of common foreign bodies are rubber band, one of the favourite objects for children and thread. Elastic bands have constrictive effects on the limb depending upon its mechanical and tensile properties. It can either have an acute or chronic presentation.⁶

In our case, probably the child wore the rubber band around her neck while playing, which remained there un-noticed for quite some time and eventually got embedded secondary to pressure necrosis on skin which results in repeated inflammatory reactions with overlying skin growth Rubber band foreign body embedment can be seen in areas like wrist, neck, leg and ankle. In some communities, tying of band like structures around wrist or neck can be part of certain religious rituals, which can remain there neglected for several years.⁷

When an inert foreign body is inaccessible or lodged close to a vital organ, conservative management is generally recommended, surgical intervention could be damaging for the vital structures if the foreign body is present in close proximity to them. The common example is stray bullets found in chest or soft tissues which are sometimes left alone. Unless foreign body is causing problems and eroding surrounding structures, then they should be removed, like in our case trachea was damaged and tracheo-cutaneous fistula was found during surgery. Surgery should be decided after evaluation of several parameters like accessibility, anatomical location and nature of the foreign body.⁸

Another important thing to rule out in such cases is child abuse, the embedded foreign body might be evidence of some form of physical abuse or manhandling. Several cases of children who presented with a circumferential scar on the neck have been reported, which appeared due to strangulation either by accident or by abuse.⁹ Both

parents were interviewed but after detailed history and investigation, no evidence of child abuse was identified in our case.

In conclusion. Even though chronic non healing sinuses with all normal investigations are a great challenge to treat, rare and unusual cause for such disease presentations should be kept in mind while managing and diagnosing these cases.

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REFERENCES

1. Kamath SU, Kamath SS, Tejaswi P. A rare and unusual cause of discharging sinus around the knee. *Asian J Pharm Clin Res* 2016;9(4):1–2.
2. Bhat S. *SRB's Manual of Surgery*. Jaypee Brothers Medical Publishers; 2019.
3. Chakrabarti S, Pal S, Biswas BK, Bose K, Pal S, Pathak S. Clinico-pathological study of cutaneous granulomatous lesions-a 5 yr experience in a tertiary care hospital in India. *Iran J Pathol* 2016;11(1):54–60.
4. Yang G, Huang Y, Ye W, Yu H, Mei H. A rare case report of acquired rubber band syndrome due to an unnoticed rubber band on a baby's ankle. *Transl Pediatr* 2020;9(1):66–9.
5. Rupert J, Honeycutt JD, Odom MR. Foreign bodies in the skin: evaluation and management. *Am Fam Physiciaan* 2020;101(12):740–7.
6. John R, Khurana A, Raj NG, Aggarwal P, Kanojia R, Chayapathi V. The 'forgotten rubber band'syndrome–A systematic review of a uniquely 'desi'complication with a case illustration. *J Clin Orthop Trauma* 2019;10(4):822–7.
7. Kumar M, Sharma KK, Chauhan LK, Mehta P. Rubber band (Dhaga) syndrome of the wrist. *Indian J Pediatr* 2018;85(12):1136–7.
8. Clarós P, Fokouo J, Clarós A. Intraorbital foreign body: A rifle bullet removed 20 years after the accident. *Eur Ann Otorhinolaryngol Head Neck Dis* 2017;134(1):63–5.
9. Yetim A, Aygüler E, Özçetin M, Bayramoğlu Z, Çelik M, Güdek K, *et al*. Foreign Body Reaction Due to a Forgotten Rubber Band on the Neck of a Toddler Girl. *Pediatr Emerg Care* 2019;37(8):e491–4.
10. Sinikumpu JJ, Serlo W. Confirmed and Suspected Foreign Body Injuries in Children during 2008–2013: A Hospital-Based Single Center Study in Oulu University Hospital. *Scand J Surg* 2017;106(4):350–5.

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