

## Case Report

# Thoracic Rib Cage Lesions Caused by Thiamin Deficiency - Etiology and Physiologic Therapy of Tietze's Syndrome

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**Abstract**

This paper describes abnormalities of the thoracic rib cage observed in a huge labor farm, where every laborer should work terribly hard with inadequate food for several years. The lesions involved the sternum and its surrounding rib cartilages articulated with it. A horizontal ridge developed at the sternal angle; xiphoid processor could be very painful and disfiguring; the costal margin might become opening to the lateral sides and upward.

Twenty-five cases of costochondritis with swelling had been observed within 4 years and cured with small dose of thiamin by local infusion because of medicine under supply. The dramatic therapeutic response confirmed that thiamin deficiency is the etiology of Tietze's syndrome, which was found worldwide only 159 case within 35 years as reported in 1956, and another worldwide 131 cases were added 1955-1960.

**Keywords:** Costochondritis; Costal margin; Rib cage; Sternal angle; Sternum; Thiamine deficiency; Tietze's syndrome; Xiphoid processor

**Case Presentation**

Human thoracic rib cage contains and protects lungs, heart and large vessels and is responsible to help respiration and circulation. Its building framework must be not too hard, strong or fixed and should have some degree of mobility, elasticity and multiple joints to adapt the expansion and contraction of the thoracic cavity. Therefore, sternum and its surrounding articulated rib cartilages become very important and very vulnerable to daily tear and wear. Some lesions might occur at the sternum and its surrounding cartilages especially following exhausted hard labor and hunger as seen in a labor farm 6 decades ago, 1958-1962.

**Lesions Observed****General appearance of the thoracic cage**

**A. The compressible chest cage:** In some cases, the resistance of the chest cage greatly decreased after prolonged malnutrition. The anterior-posterior thickness of the chest cage could be readily pressed to ½ or 2/3 of its original as pressing in a child. In advanced cases, finger pressure might be powerful enough to have accordion-like movement of the chest. This phenomenon was obviously due to bone and muscle weakness. Fortunately, it was rare and found only in severe cachexic and marasmus cases with general weakness before imprisonment. Actually, the clinicians could do nothing for its correction because it was caused chiefly by calorie and protein deficiencies and irreversible. The prognosis was very poor.

**B. Unilateral depression of the chest:** In a case of 55 years old, the entire left side of his chest became progressively depressed within 2 years until its surface became about two cm lower than the right side. The depression was conspicuously visible but no abnormality in heart beats and respiration was found. This was definitely due to local muscle sarcopenia and bone weakness due to malnutrition over years. No treatment could be tried.

**C. Slightly opened costal margins:** In a few cases, the two costal margins on both sides opened slightly wider to both lateral directions and slightly bent upward at their thicker edges. It caused no trouble to the patients' daily activity, however, sudden, sharp and unbearable chest pain mimicking myocardial infarction had occurred in three cases from the margin close to 7<sup>th</sup> and 8<sup>th</sup> cartilage conjunction.

In one case, the sharp pain was promptly and completely relieved within an hour after intramuscular thiamin 100mg, while the other 2 cases, each was locally injected with thiamin 50mg and pain became mild and tolerable. Home made yeast liquid two cups a day were then supplied because of medicine destitution. The liquid yeast was fermented with crude extract from sugar beet. It worked and soothed the pain mildly until pain free within 3-5 days. In a regular medical service, intramuscular thiamin 200-300 mg once, twice or twice daily could stop pain immediately regardless how severe the sudden pain. This indicated that the combined rib cartilages were also vulnerable to nutritional deficiency. It should be also considered as a part of Tietze's syndrome.

**The sternum**

The sternum has a series of bilateral notches to articulate with clavicles and rib cartilages on both sides. Each articulation can be enveloped with enlarged cartilage potentially. The following abnormalities were observed:

**A. Enlarged sternal angle:** The inferior border of manubrium conjugating with the body part of the sternum between the articulated cartilages from the second ribs on both sides developed a horizontal cartilage ridge about 3-5 mm in height without pain. It was found over 10% of laborers imprisoned more than 5 years. After release, the ridge could be vanished within a year or more.

**B. Xiphoidalgia and deformity:** The small lower part of the

sternum is very troublesome clinically. Xiphoidalgia and tenderness were not only common but often causing gastric or respiratory signs including nausea, vomit, or cough. Local infusion of thiamin was constantly required to relieve pain because regular pharmaceutical analgesics was useless.

Deformity of this process was also common, either increasing in size or changing in shape and even twist. Normally, this process makes a depression in the epigastric region of the abdominal wall, which is called epigastric fossa or pit of the stomach. In 20% of the laborers over 5 years, this fossa became enlarged and might reach the size to fill in the ventral portion of a man's thumb or to contain a hen's egg. Its lower apex flexed upward and become a projected point on the abdominal wall.

In some cases, the process enlarged transversely even close to both side rib costal margins. These indicated that the shape and size of xiphoid process might become bigger and spoon- or spade-like, or even twisted. No accompanied enlargement of cartilage of the xiphisternal joint was observed by us. However, Tietze's syndrome lesion at xiphisternal joint had been reported by Jelenko, 1974 [1].

### Tietze's syndrome

The swelling costochondritis of the rib cartilage constitutes Tietze's syndrome. It occurred usually only in one rib cartilage, mostly the second or the third rib cartilage. The costal margins on both sides also can become sudden sharp painful but no local swelling had observed. Within 4 years, twenty-five typical cases of Tietze's syndrome were diagnosed among thousands of laborers. The involved cartilage swelling was fusiform, firm, and terribly painful with severe tender. Acute sharp pain was so severe that frequently causing debilitating as in myocardial infarction. No injury or any other cause could be found except for prolonged terribly hard labor. Vitamin C and regular analgesics, such as pyramidon or paracetamol, were useless while local infusion of thiamin 50mg could relief pain within hours and reduced or eliminated the swelling after 3-5 injections, which were injected within a week. Definitely, this dose was too low than it should be due to medicine destitution. When pain became milder, thiamin infusing was replaced with home-made liquid yeast two cups a day. In this way, twenty-five cases were cured without any consequence. Of course, in case of intramuscular or local infusion thiamine 200-300 mg twice or trice daily the painful swelling would be eliminated within 2-4 days and no more.

## Discussion

Since all the laborers were under the same prolonged semi-starvation and the same arduous physical labor, nutritional deficiency should be the etiology for some commonly associated lesions if confirmed with nutritional therapy.

### The sternum

The enlarged sternal angle was symptom free. It may be useful as a standard for evaluation of patients' nutritional status. Its surrounding rib cartilages may be a potential resource of new rheumatic disease.

The xiphoid process was very vulnerable and troublesome. Its pain and tenderness were common and local infusion of thiamin often required. Since it is too vulnerable and connects with abdominal diaphragmatic muscle, and sternopericardial ligament, therapeutic

practice must be very careful and gentle, therefore, local infusion of thiamin was usually held.

The rib cartilages and the costal margins are critically important because both of them yielding sudden sharp intolerable pain of Tietze's syndrome.

### Etiology and therapy of Tietze's syndrome

Tietze [2] and several other authors had implicated nutritional deficiency as its etiology. Evans and Eames [3] had pointed out that it might be due to deficiency of vitamin C and B. Our observation supported their hypothesis but exclude vitamin C because it had no effect.

**a. High incidence:** Among the malnourished population: Twenty-five cases were observed within 4 years among 5,000 individuals with definite malnutrition, while Kayser collected only 159 cases within 35 years from world literature in 1956 [4]. Aeschlimann and Khan added another 131 cases of 1955-1960 worldwide [5], totally 290.

**b. Dramatic therapeutic effect:** Regular analgesics were useless while parenteral thiamin relieved sharp pain promptly and reduced the swelling if continued. In a case with a sudden, sharp and debilitating pain from costal margin mimicking myocardial infarction, two injections of thiamin 100mg relieved pain completely in an hour. In typical case with swelling rib cartilage, thiamin 100mg was usually injected to reduce pain or make it tolerable. Then followed by local infusion of thiamin 50mg close to the swelling for 3-5 times. Homemade liquid yeast might be supplied. The purpose was to save thiamin. In regular medical service, parenteral thiamin 200-300 mg twice or trice daily could eliminate the abnormality no more than 3 days.

**c. Dietary improvement:** Among the released, Tietze's syndrome was either absent or cured after dietary improvement. Therefore, thiamin deficiency would be the primary etiology for Tietze's syndrome while the role of other nutrient deficiency would be only minimal.

### Possible mechanism of rib cartilage disorders in thiamin deficiency

Thiamin plays a critical role in energy metabolism. When thiamin is deficient, the activities of enzymes in glycolysis decrease. Glutathione content in blood and tissues also decreases. Some deleterious metabolites may be excessively produced or accumulated, including free radicals,  $\alpha$ -oxoaldehydes, lipid peroxides, Advanced Glycation End-Products (AGEs), etc [6,7]. They may contribute to the pathogenesis of cartilage lesions by [8-10]:

- a. Causing degradation of chondrocyte and the matrix including proteoglycan and collagen.
- b. Reducing the capacity of chondrocyte to remodel their extra-cellular matrix.
- c. Interfering with cellular processes, such as adhesion of cells to the matrix, proliferation and gene expression.
- d. Increasing the stiffness, brittleness of articular cartilage and make it more prone to mechanical damage.

e. Cross-linking and increasing the dimension of the collagen fibers by opening the double bonds of glyoxals.

## Conclusion

1. Thiamin deficiency should be the etiology of Tietze's syndrome. The swelling costochondritis or sharp pain from cartilage margin can be promptly eliminated within a few days when injecting large dose of thiamin.

2. Regular analgesics had no effect and steroids should not be used.

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